# Colour Perception in Autism Spectrum Condition and Williams Syndrome

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# Abstract

Autism Spectrum Condition (ASC) and Williams syndrome (WS) are neurodevelopmental conditions associated with socio-communicative deficits. Also, present in both conditions are sensory sensitivities and reactivities. In ASC extreme sensory reactivity/sensitivities in one or more of any sensory domain have recently been recognised as new diagnostic criteria in DSM-V. Whilst in WS there are reported visuo-spatial and auditory atypicalities. There is increasing importance in identifying both the typical and atypical development of sensory processing, as well as establishing condition-specific and condition-general aspects of sensory processing. Traditionally sensory processing has been studied using a cross-sectional design using either psychophysical tasks or behavioural questionnaires. However little work has attempted to link between these different methodologies resulting in a disconnected study of sensory processing in both typical and atypical development. Colour perception is useful domain to study sensory processing because it can be characterised through psychophysical/cognitive tasks and behavioural questionnaires. Colour perception is also relatively understudied in both ASC and WS despite anecdotal reports of behaviour being influenced by colour. The present research aims to investigate colour perception in ASC and WS relative to mental age typically developing (TD) controls using the same participants across a combination of psychophysical (chromatic discrimination - chapter 3), cognitive (chapters 4 and 5, colour preference and naming), questionnaire (chapter 6) and case studies (chapter 7) methodologies to establish a rounded representation of colour perception in ASC and WS through using these mixed methodologies. The results show condition specific atypicalities across all tasks relative to TD controls. For the ASC group, there was poorer chromatic discrimination, different colour preference patterns and increased frequency of colour affected behaviours. Whilst the WS group showed less pronounced colour preference patterns and atypical colour naming. The differences in results between different measurements of colour perception and condition-specific responses between the ASC and WS suggest that sensory processing is not a homogenous concept but should be considered in relation to the measurements chosen. The results are discussed within the context of diagnostic criteria, approaches to studying sensory processing and syndrome-specificity.

# Dedication

This thesis is dedicated to those individuals with autism and Williams syndrome and their families. These children who served as an inspiration to me through their personality as well as their struggles, and also their parents who are so dedicated to their children, were a major motivation for this body of work.

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# Table of Contents

**Chapter 1 – General Introduction** ..... 1

**1.1 Overview** ..... 1

**1.2. Overview of the neurodevelopmental disorders Autism Spectrum Condition and Williams Syndrome** ..... 1

**1.2.1 Overview of Autism Spectrum Condition** ..... 1

**1.2.2 Sensory Processing Atypicalities in Autism** ..... 3

**1.2.3 Overview of Williams Syndrome** ..... 5

**1.2.4 Sensory Processing Atypicalities in Williams Syndrome** ..... 6

**1.3. Overview of Human Visual Processing** ..... 7

**1.3.1 Colour Vision** ..... 10

**1.4. Atypicalities of Visual Processing in the Developmental Conditions Autism and Williams syndrome** ..... 14

**1.4.1 Visual Processing in Autism Overview** ..... 14

**1.4.2 Visual Processing in Williams Syndrome Overview** ..... 20

**1.5. Neural correlates and theoretical explanations of sensory processing atypicalities in developmental disorders** ..... 21

**1.5.1 Perceptual theories of atypical sensory processing in Autism** ..... 21

**1.5.2 Neural correlates of sensory processing atypicalities in autism** ..... 23

**1.5.3. Perceptual Theories of Williams Syndrome** ..... 25

**1.5.3.2 Neural correlates of atypical sensory processing in WS** ..... 26

**1.6. Comparison of Sensory Sensitivities between Autism and Williams Syndrome** ..... 27

**1.7. Why Study Colour Perception in Autism and Williams Syndrome** ..... 28

**1.8. Aims of Thesis** ..... 29

**Chapter 2 – General Methods** ..... 31

**2.1 Overview** ..... 31

**2.2 Characterisation of Sample** ..... 31

**2.3 Participant Recruitment** ..... 33

**2.3.1 Sample Size** ..... 34

**2.3.2 Inclusion and exclusion criteria** ..... 35

**2.3.3 Final Sample Criteria** ..... 35

**2.4 Developmental Methods** ..... 37

**2.4.1 Matched Groups Design** ..... 37

**2.4.2 Developmental Trajectories** ..... **Error! Bookmark not defined.**

**2.5 The Psychophysics Method** ..... 42

**2.5.1 Forced Choice Method** ..... 43

**2.5.2 Staircase Method** ..... 44

**2.6 Colorimetric specification of colour stimuli** ..... 45

**2.6.1 Physiological cone-contrast space** ..... 46

**2.6.2 Hue Saturation Lightness (HSL) Colour Space** ..... 47

**2.7 Screening for Anomalous Colour Vision** ..... 48

**2.8 General Test Protocol** ..... 49

**2.9 Conclusion** ..... 50

**Chapter 3 – Chromatic Discrimination** ..... 52

**3.1 Overview** ..... 52

**3.2 Introduction** ..... 52

**3.2.1 Chromatic Discrimination Tests** ..... 52

**3.2.3 Chromatic Discrimination across the Lifespan** ..... 55

**3.2.4 Discrimination in Atypical Development** ..... 56

**3.2.5 Hypothesis** ..... 59

**3.3 Methods** ..... 59

**3.3.1 Participants** ..... 59

**3.3.2 Farnsworth-Munsell 100-Hue Test (FM100)** ..... 60

**3.3.3 Chromatic Contrast Discrimination Threshold Test (CCDT)** ..... 61

**3.4 Results** ..... 64

**3.4.1 Farnsworth-Munsell 100-Hue Test Results** ..... 64

**3.4.2 CCDT Results** ..... 67

**3.4.3 Cross Task Performance between FM100 and CCDT** ..... 69

**3.4.4 Correlation with Chronological Age** ..... 69

**3.4.5 Drivers of Task Performance on FM100** ..... 70

**3.5 Discussion** ..... 72

**3.5.1 Appropriateness of Chromatic Discrimination Tasks** ..... 77

**3.6 Conclusion** ..... 79

**Chapter 4 – Colour Preference** ..... 81

**4.1 Overview** ..... 81

**4.2 Introduction** ..... 81

**4.2.1 Individual Differences in colour preference** ..... 81

**4.2.2 Models of colour preference** ..... 86

**4.2.3 Colour preference in Autism and Williams syndrome** ..... 89

**4.2.4 Study Rationale and Hypothesis** ..... 91

**4.3 Methods** ..... 91

**4.3.1 Participants** ..... 91

**4.3.2 Apparatus** ..... 92

**4.3.3 Stimuli** ..... 92

**4.3.4 Design** ..... 94

**4.3.5 Data Analysis** ..... 94

**4.3.6 Procedure** ..... 95

**4.4 Results** ..... 95

**4.4.1 Hue Variants** ..... 95

**4.4.2 Saturation Variants** ..... 97

**4.4.3 Lightness Variants** ..... 99

**4.4.4 Strength of colour preference** ..... 101

**4.4.5 Preferences of Individual colours** ..... 104

**4.4.6 Cone-opponent contrast components of hue preference** ..... 105

**4.4.7 Relationship between colour preference, age, and chromatic discrimination** ..... 111

**4.5 Discussion** ..... 112

**4.5.1 Summary of results**.....112

**4.5.2 Relationship to previous Studies of Hue, Saturation and Lightness Preference**.....114

**4.5.3 Relationship of findings to models of colour preference.** .....116

**4.5.4 Limitation**.....118

**4.6 Conclusion**.....119

**Chapter 5 - Colour Naming**.....122

**5.1 Overview**.....122

**5.2 Introduction**.....122

**5.2.1. Overview of colour naming**.....122

**5.2.2 Universal or Relative colour categories?** .....122

**5.2.2. Adult colour naming studies** .....125

**5.2.3. Development of colour naming** .....126

**5.2.4. Colour Naming in Williams syndrome and Autism** .....129

**5.2.5 Rationale and Hypothesis** .....131

**5.3 Methods**.....131

**5.3.1 Participants**.....131

**5.3.2 Stimuli** .....132

**5.3.3 Coding of Colour Terms** .....132

**5.3.4 Concordance Rating**.....133

**5.3.5 Comparison with other variables**.....133

**5.3.6 Procedure**.....134

**5.4 Results**.....134

**5.4.1 Frequency of Colour Term Use**.....134

**5.4.2 BCT/NBCT, Use of Modifiers** .....136

**5.4.4 Concordance** .....138

**5.4.4 Relationship between Colour Naming and Verbal ability**.....141

**5.4.5 Relationship between Colour Naming and Chromatic discrimination** .....141

**5.4.6 Relationship between Colour Naming and Chronological age** .....142

**5.4.7 “Favourite” Colour Consensus between groups.** .....142

**5.4.9 Parental Questionnaire: Colour Naming** ..... 142

**5.5 Discussion** ..... 143

**5.5.1 Limitations:** ..... 147

**5.6 Conclusion** ..... 148

**Chapter 6 – Parent Reported Colour Affected Behaviours** ..... 150

**6.1 Overview** ..... 150

**6.2 Introduction** ..... 150

**6.2.1 Background to questionnaires** ..... 150

**6.2.2 Sensory Questionnaires Summary** ..... 157

**6.2.3 Qualitative Studies on Sensory Experiences** ..... 157

**6.2.4 Summary Questionnaire and Interview studies on Sensory Processing** ..... 159

**6.2.5 Chapter Rationale and Aims** ..... 159

**6.3 Method** ..... 159

**6.3.1 Participants** ..... 159

**6.3.2 Questionnaire Development** ..... 160

**6.3.3 Procedure** ..... 162

**6.3.4 Data Analysis** ..... 162

**6.4 Results** ..... 163

**6.4.1 Questionnaire Psychometric Properties** ..... 163

**6.4.2 Questionnaire Results** ..... 168

**6.5 Discussion** ..... 175

6.6 Conclusion ..... 178

**Chapter 7 - Case Studies** ..... 180

**7.1 Overview** ..... 180

**7.2 Introduction** ..... 180

**7.2.1 The Mixed Methods Method** ..... 180

**7.2.2 Colour affected behaviours as Restricted and Repetitive Behaviours** ..... 181

**7.2.3 Rationale for Chapter:** ..... 182

**7.3 Method** ..... 182

**7.3.1 Selection of Cases** ..... 182

**7.3.2 Data Analysis** ..... 182

**7.3.3 Classification of Cases** ..... 182

**7.4 Results** ..... 183

**7.4.1 TD group** ..... 183

**7.4.2 Autism Case Studies** ..... 184

**7.4.3 WS Case Studies** ..... 189

**7.4.4 Summary of Case Studies** ..... 195

**7.4.5 Frequencies of Classification between groups** ..... 196

**7.5 Discussion** ..... 196

**7.6 Conclusion** ..... 203

**Chapter 8 - General Discussion** ..... 205

**8.1 Summary of Results** ..... 205

**8.1.1 Profile of Colour Perception in Autism** ..... 206

**8.1.2 Profile of Colour Perception in Williams Syndrome** ..... 207

**8.1.3 Comparison of Colour Perception Profiles between Autism and Williams Syndrome** ..... 207

**8.1.4 Relation to Wider Literature** ..... 208

**8.1.4.2 Applied uses of colour** ..... 214

**8.2 Appraisal of Research** ..... 216

**8.2.1 Strengths** ..... 216

**8.2.2 Limitations** ..... 217

**8.3 Future Directions for Research** ..... 220

**8.4 Conclusion** ..... 221

**Appendices** ..... 223

**Appendix 1 – Parental Information Sheet** ..... 223

**Appendix 2 – Colour Checker HSL Values** ..... 227

**Appendix 3 – Colour Preference Hue Comparisons** ..... 228

**Autism Analysis** ..... 228

**Williams Syndrome Analysis** ..... 228

**Appendix 4 – Parental Questionnaire Version 1**..... 229

**Appendix 5 – Parental Questionnaire Version 2**..... 237

**Appendix 6 – Additional Case Studies**..... 247

**Extreme Hue Preference**..... 247

**Colour Insistent Behaviours** ..... 247

**References**..... 249

# List of Figures

Figure 1-1 Major anatomical structures of the retino-geniculate striate pathway. ..... 9
Figure 1-2- Illustration of the cortical dorsal and ventral pathways. ..... 10
Figure 1-3 - Representation of the spectral sensitivities with respect to wavelength (nm) for each cone class. ..... 11
Figure 1-4 - Visual depiction of the colour opponent channels that are present in the retinal ganglion cells and maintained through the LGN. ..... 12
Figure 2-1 - Example of a Cone Contrast space. ..... 47
Figure 2-2 - HSL colour space. ..... 48
Figure 3-1 - Total Error Scores (TES) for the FM100. ..... 66
Figure 3-2 – NVIQ scores plotted against FM100 Total Error Scores. ..... 67
Figure 3-3 – Chromatic Contrast Discrimination Threshold discrimination thresholds for the luminance, red-green and blue-yellow colour axes. ..... 69
Figure 4-1 – Mock ups of Colour Preference stimuli. ..... 94
Figure 4-2 - Colour preference across Hue variants split by male and female participants for the TD and autism groups. ..... 96
Figure 4-3 – Colour preference across Hue variants split by male and female participants for the TD and Williams Syndrome groups. ..... 97
Figure 4-4 - The preference for different saturations variants across both autism and TD groups. ... 98
Figure 4-5 The preference for different saturations variants across both Williams syndrome and TD groups. ..... 99
Figure 4-6 – Colour preference for different lightness variants across both Williams Syndrome and TD groups. ..... 100
Figure 4-7 – Colour preference for different lightness variants across both Williams Syndrome and TD groups. ..... 101
Figure 4-8 – Strength of colour preference (as measured by standard deviation of proportion preferred) for the autism and TD groups. ..... 102
Figure 4-9 - Strength of colour preference (as measured by standard deviation of proportion preferred) for the Williams syndrome and TD groups. ..... 103
Figure 4-10– The proportion that the selected “favourite” colour was selected. ..... 105
Figure 4-11 – The cone contrast weights for the autism and TD groups. ..... 107
Figure 4-12 - The cone contrast weights for the Williams Syndrome and TD groups. ..... 108
Figure 4-13– The total amount of hue colour preference variance explained by cone contrasts for autism and TD groups. ..... 110
Figure 4-14– The total amount of hue colour preference variance explained by cone contrasts for autism and TD groups. ..... 111
Figure 5-1 – The eleven BCTs proposed by Berlin and Kay (1969). ..... 124
Figure 5-2 - Frequency of colour terms used by both TD and Williams syndrome groups. ..... 136
Figure 5-3 - BCTs, modified BCTs or non-BCTs use. ..... 138
Figure 5-4 – Group Concordance ratings across all, chromatic and achromatic colours. ..... 140
Figure 5-5 - Concordance ratings for all participants across all colours, chromatic and achromatic colours only. ..... 140
Figure 5-6 - Overall chromatic consensus rating plotted against verbal ability. ..... 141

Figure 5-7: Group chromatic consensus rating plotted against Chromatic Discrimination thresholds. ..... 142
Figure 6-1- Sensory processing as assessed by the Sensory Profile. ..... 151
Figure 6-2 – Mean scores on the visual questions of the Sensory Profile Questions. ..... 168
Figure 6-3 – Average frequency of colour affected behaviours. ..... 169
Figure 6-4 – The distributions of the TD, Autism and WS groups. ..... 171
Figure 6-5 - Proportion of colour affected actions for the autism and TD groups. ..... 171
Figure 6-6 – Proportion of participants with emotional responses to colour. ..... 172
Figure 7-1 - Individual colour preference curves for Case Study TD 1. ..... 184
Figure 7-2 - Preference curves for Autism Case Study 1. ..... 185
Figure 7-3 - Preference curve for Autism Case Study 2. The x-axis is the hue radians, whilst the y-axis is the proportion that the colour was preferred. ..... 187
Figure 7-4 - Preference curves for Autism Case Study 3. ..... 188
Figure 7-5 - Preference curves for Williams Syndrome Case Study 1. ..... 190
Figure 7-6 - Preference curves for Williams syndrome Case Study 2. ..... 192
Figure 7-7 - Preference curves for Williams Syndrome Case Study 3. ..... 193
Figure 7-8 - Preference curves for Williams syndrome Case Study 4. ..... 194
Figure 7-9 – Example of colour preference curves for individuals who did not display a colour affected behaviour. ..... 195

# List of Tables

Table 2-1 – Participant demographics for all participants that were recruited and passed the inclusion/exclusion criteria. ..... 37
Table 4-2- Participant demographics for Chapter 4. ..... 92
Table 4-3 - The colour coordinates of colour preference stimuli. ..... 93
Table 4-4 - Cone contrasts values for the 8bit and 10bit setups. ..... 105
Table 4-5 - Results for individual cone contrast weights for autism and TD groups. ..... 106
Table 4-6 – Results for individual cone contrast weights for Williams Syndrome and TD groups. .... 108
Table 5-7 – Colour Naming participant demographics. ..... 132
Table 5-8– Colours that significantly differed between TD and Williams syndrome groups..... 138
Table 6-10 – Initial factor loadings for each individual question item onto the five different factors. ..... 164
Table 6-11- Revised factor loadings for each individual question item onto the four different factors. ..... 167
Table 7-12 - Percentage of matches/mismatches between hue colour preference curves and colour affected behaviours and no colour affected behaviours for each group. ..... 196

# Chapter 1 – General Introduction

## 1.1 Overview

Despite the recent addition of abnormal sensory sensitivities or interests to diagnostic criteria for autism in DSM-V, little is known about sensory processing in autism or whether sensory processing in autism is similar or different in relation to other developmental conditions (Rogers & Ozonoff, 2005). In this thesis, the comparison of how autism and Williams Syndrome vary (if at all) relative to typically developing controls will enable issues relating to condition specificity of colour perception between autism and Williams Syndrome. Firstly, this chapter will introduce diagnostic criteria and behavioural phenotype, with a focus on sensory atypicalities of both conditions. Next there will be an overview of visual processing in typical developing individuals, and how atypicalities in visual perception manifest in autism and Williams syndrome, including a description of colour perception in both conditions. Finally, current theoretical frameworks for this visual atypicalities will also be reviewed, focusing on underlying neural mechanisms and cognitive theories.

## 1.2. Overview of the neurodevelopmental disorders Autism Spectrum Condition and Williams Syndrome

### 1.2.1 Overview of Autism Spectrum Condition

Autism is a neurodevelopmental condition characterised primarily by persistent deficits in social communication and social interaction as well as restricted and repetitive behaviours (American Psychiatric Association, 2013). Social communication and interaction problems include difficulties in reciprocating social behaviours or using non-verbal communicative behaviours, such as gestures or facial expressions. Restricted and repetitive behaviours include stereotyped or repetitive movements, such as lining up toy cars, and an insistence on sameness such as an inflexible following of a routine. Also, included under this behaviour is the so-called “hyper”- or “hypo”- reactivity to sensory input (see next section for further definition), which may manifest itself in behaviours such as the excessive smelling of objects or fascination with lights/spinning objects. To be considered diagnostic of ASC, these symptoms must be present early in life and unexplained by intellectual disability or a global developmental delay. At present, there are no reliable biomarkers for autism therefore diagnosis is reliant on a combination of interviews with the parent and child, behavioural assessments and a review of the child’s history.

The historical diagnosis of autism has changed over the course of history with many terms being used to describe autism or similar behavioural phenotypes including (but not limited to); autism spectrum disorder, childhood schizophrenia, infantile autism and childhood psychosis (Le Couteur & Szatmari, 2015). In DSM-III autism was separated from childhood schizophrenia and psychosis, and pervasive

developmental disorder was added as an additional diagnostic term (American Psychiatric Association, 1994). DSM-IV included further changes, such as multiple sub-categories of autism such as; autism, Asperger's syndrome (social characteristics of autism without language or cognitive delay), Rett Syndrome (developmental regression manifestation of autism phenotype), pervasive developmental disorder not otherwise specified (characteristics of autism but not enough to reach criteria for either autism or Aspeger's syndrome) (American Psychiatric Association, 2000). With the publication of DSM-5 there were further changes to autism diagnoses. Subcategories of autism were removed due to uncertainty over clinical specificity between them (Lord et al., 2012) and were replaced with a general diagnosis of autism. The criteria of autism in DSM-V focusing on social communication and repetitive stereotyped behaviours (including speech, sensory behaviours and sensory-motor).

Autism occurs on a spectrum and behavioural phenotypes are highly heterogeneous (Jeste & Geschwind, 2014; Waterhouse, 2013). Wide inter-individual in the behavioural phenotype of autism is reflected in the DSM-V criteria which describe three levels of severity, from Level 1, which "requires some support", to Level 3, with "substantial support required", based upon the severity of the social symptoms and rigidness in repetitive behaviours (American Psychiatric Association, 2013). Given the high levels of heterogeneity in the autism, it has been argued that treating autism as a single homogenous group is not always beneficial and that this variation need also to be investigated. For example, islets of ability (e.g. as seen in savants) have been identified in some individuals with autism, (Heaton, Hermelin, & Pring, 1998) but these are not present in every case. Appreciation of this heterogeneity may see different subgroups and risk factors for autism be identified that are dependent on how autism manifests itself in those individuals (Jeste & Geschwind, 2014) but with some overlapping underlying neural and genetic characteristics (Geschwind, 2008; Geschwind & Levitt, 2007). Despite this overlap, it is unlikely that there is one single underlying biomarker or theory to account for all phenotypes of autism (Rutter, 2011; Szatmari, 2011).

It has also been proposed that there are autistic traits which lie on a continuum that includes the whole population, but that only those at the extreme end meet diagnosis criteria (Baron-Cohen, Wheelwright, Skinner, Martin, & Clubley, 2001). This "broader autism phenotype" can be seen in relatives of individuals with autism, who do not have a diagnosis themselves. These relatives are more likely to show isolated autistic traits than the wider population (Mandy & Skuse, 2008). The notion of a "broader autism phenotype" has been used to study the traits of autism in general population (both adults and children) who do not meet requirement for autism and to establish whether the presence of these traits influence performance on social and perceptual tasks (Constantino & Todd, 2003; Jackson et al., 2013; Robertson & Simmons, 2013; Skuse et al., 2009; Walter, Dassonville, & Bochsler, 2009). Measures of the "broader autism phenotype" such as the

Autism Quotient (AQ) (Baron-Cohen et al., 2001) have been suggested to characterise "autistic traits" in the general population and scores on the (AQ). This highlights the notion that there is a spectrum where there are individuals in the general population who display some characteristics for autism but that these are not severe enough to warrant a diagnosis.

The prevalence of autism is approximately 1/100 (Baird et al., 2006), although it has been reported to be as low as 1/68 (CDC, 2014). Autism is more commonly diagnosed in males (Baird et al., 2006; CDC, 2014). However it is not clear whether this difference in diagnosis rates between males and females is due to an actual difference in prevalence between males and females in autism or due to underlying biological sex differences (Baron-Cohen, 2002; Baron-Cohen, Knickmeyer, & Belmonte, 2005). The sex difference in diagnosis might instead be due to nuanced differences in the manifestation of autism between males and females, with less repetitive and stereotyped behaviour and better social communication abilities occurring in females with autism compared to males (Holtmann, Bölte, & Poustka, 2007; Mandy et al., 2012), or to a specific difficulty in identifying autism in higher functioning individuals who are also female (Frazier, Georgiades, Bishop, & Hardan, 2014) (see (Halladay et al., 2015) for review).

### 1.2.2 Sensory Processing Atypicalities in Autism

Individuals with autism often have atypicalities of sensory processing in at least one sensory modality (Tomchek & Dunn, 2007). This sensory atypicality may range from "hypo"-sensitivity (which is typically defined as a reduced response to sensory input) to "hyper"-sensitivity (increased response) to sensory input, and may manifest as sensory seeking behaviours (Ben-Sasson, Carter, & Briggs-Gowan, 2009; O'Neill & Jones, 1997; Pellicano, 2013; Rogers & Ozonoff, 2005). It is important to note that there are multiple inconsistencies in terminology and different ways of measuring hyper-/hypo-sensitivity between research fields. As defined above they relate to the term use in sensory questionnaires and how sensory processing relates to general behaviours. However, it is unclear the extent to which these hyper-/hypo- questions relate to actual sensory behaviours, for example many of the questions could be characteristic of either a hyper-/hypo- sensitivity. Whereas in psychophysics sensitivity relates to the ability to detect a stimulus. This issue will be expanded upon in Chapter 6. This thesis will use the psychophysical definition of sensitivity, unless otherwise stated. Both hyper-sensitivity and hypo-sensitivity responses may occur within the same or different modalities in a single individual in both questionnaires and psychophysical studies (Ben-Sasson et al., 2009; Simmons et al., 2009). Furthermore these sensory atypicalities as measured by sensory questionnaires are found in individuals with autism regardless of their level of ability (Rogers, Hepburn, & Wehner, 2003). There is not a coherent profile of sensory sensitivity across different sensory modalities in autism, but there may be distinct subgroups of different types sensory sensitivities, i.e. individuals who display hyper- or hypo-sensitivity for one sensory modality but not

another (Lane, Young, Baker, & Angley, 2010). Sensory sensitivities are also found across the lifespan in individuals with autism, from toddlers (2 years) (Rogers et al., 2003), and increase severity and frequency up to mid childhood and also continue into adulthood (Tavassoli, Hoekstra, & Baron-Cohen, 2014; Tavassoli, Miller, Schoen, Nielsen, & Baron-Cohen, 2013), but how these sensory sensitivities manifest also change with age (Kern et al., 2006).

Several different methodologies have been used to study sensory processing in autism. These range from questionnaires (e.g. The Sensory Perception Quotient (Tavassoli et al., 2014), Sensory Profile (Dunn, 1997), Glasgow Sensory Questionnaire (Robertson & Simmons, 2013)) to interviews with either the parent or the child/adult with autism (Kirby, Dickie, & Baranek, 2015; Robertson & Simmons, 2015). Individuals with autism have described themselves as having strong reactions to sensory inputs in different modalities (such as particular odours or sounds) that can lead to anxiety or elicit a bodily reaction, as well as believing that this response is exaggerated in themselves when compared to others.

Whilst these questionnaires and interviews can be useful to give prevalence or qualitative details about the sensory experiences of individuals with autism, they are not a direct quantitative measure of sensory processing in these individuals. By their nature, the questionnaires collapse across many different sub areas within a sensory domain. For example, with respect to visual processing the underlying neural structures and networks involved are different for each sub-modality of vision, (e.g. depth, colour, motion etc). Yet, the sensory questionnaires either do not distinguish between these sub areas or omit sub-modalities entirely (e.g. in many sensory questionnaires, there are no questions related to colour) and as such are not sensitive enough to identify specific atypicalities within a sensory domain. Other experimental methods do give precise details of processing in sub areas of vision. For example, the method of psychophysics has been used successfully to uncover phenomena related to processing sensory information, such as reading speeds or reduction of “visual stress”. This has also been identified in both children and clinical populations such as glaucoma and Alzheimer’s disease (Garway-Heath, Holder, Fitzke, & Hitchings, 2002; Legge, Ross, Isenberg, & Lamay, 1992; Peters et al., 2003).

Questionnaires have found that sensory hyper- or hypo-sensitivities have been shown to have a profound impact on the daily routines of families and parents of children with autism (Bagby, Dickie, & Baranek, 2012). Furthermore these sensory symptoms (again as measured by questionnaires) have been found to be associated with repetitive behaviours (Boyd et al., 2010; Boyd, McBee, Holtzclaw, Baranek, & Bodfish, 2009; Rogers et al., 2003) (see below) and anxiety in autism (Green & Ben-Sasson, 2010)(Wigham et al 2015), and in fact, in the current (DSM-V) diagnostic criteria, atypical sensory processing and unusual sensory interests are now classified in the domain of repetitive behaviours. The directionality of association of sensory symptoms with repetitive behaviours or

anxiety is not clear, although there is an association between the sensory sensitivities and behavioural problems such as repetitive behaviours or anxiety (Boyd et al., 2010; Green, Ben-Sasson, Soto, & Carter, 2012; Lidstone et al., 2014). This highlights one the benefits of studying sensory processing in autism and Williams syndrome is that exploration of these low level sensory atypicalities may lead to greater understanding of possible causes for higher level symptoms such as anxiety, intolerance of uncertainty and repetitive behaviours.

It has been proposed that in response to either increased "hyper-sensitive" or "hypo-sensitive" sensory processing, repetitive behaviours develop as a compensatory strategy to compensate for the atypical sensory processing. There is modest evidence to support this hypothesis. There is a moderate correlation between scores on Sensory Profile and repetitive behaviours in young children and adolescents with autism (Baker, Lane, Angley, & Young, 2008; Boyd et al., 2010; Boyd et al., 2009; Chen, Rodgers, & McConachie, 2009; Gabriels et al., 2008). Further division of repetitive behaviours and sensory processing into specific subtypes revealed that hypersensitivity correlated with stereotypy, self-injury, compulsion and ritualistic behaviours; hyposensitivity correlated with stereotypy; and sensory-seeking behaviour was associated with stereotypy, self-injury and ritualistic behaviour. Suggesting that even though sensory behaviours are under the repetitive behaviours domain, that different types of repetitive behaviours may reflect fundamentally different underlying processes for different types of sensory processing atypicalities in autism (Boyd et al., 2010).

The current evidence suggests that hyper sensory sensitivities are associated with increased repetitive behaviours. It may be that certain repetitive behaviours are sensory in nature (e.g. rocking, hand flapping) but other repetitive behaviours are not sensory in nature (e.g. insistence on sameness or unusual preoccupations) (Bishop et al., 2013; Wigham, Rodgers, South, McConachie, & Freeston). However, it should be noted that all of the research relating sensory symptoms to repetitive behaviours relies exclusively on questionnaires and does not measure the actual perceptual or physiological responses to sensory information. It is unknown in the examples above whether the sensory-motor repetitive behaviours correlate with scores on sensory processing measures only because they are the most noticeable repetitive behaviours. Section 1.4.1 will give an overview of visual perception studies in autism. The next section will give an overview to Williams syndrome

### 1.2.3 Overview of Williams Syndrome

Williams Syndrome is a rare genetic neurodevelopmental condition caused by the deletion of approximately 25 genes on the long arm of chromosome 7 (Organization, 1992, 2004). Williams syndrome is often associated with a pattern of physical "elfin" features that include a flattened naval bridge, large mouth and prominent lower lip. One of the deleted genes is the ELN gene which codes for elastin. Individuals with Williams Syndrome also tend to have vascular or changes in arterial

medial hypertension which may lead to heart disease (most commonly supravalvar aortic stenosis or hypercalcaemia) (Morris & Mervis, 2000). Williams syndrome is also associated with several ocular differences such as a stellate iris pattern and high rates of strabismus. The incidence rates of Williams Syndrome are estimated between 1/7,500 and 1/20,000 (Strømme, Bjømstad, & Ramstad, 2002; Wang et al., 1997). There are no sex differences in the prevalence of Williams Syndrome.

Williams syndrome has a common behavioural and cognitive phenotype. Individuals with Williams syndrome are described as overly friendly and highly sociable (Doyle, Bellugi, Korenberg, & Graham, 2004; Gosch & Pankau, 1997) and have a heightened salience to social stimuli such as faces (Frigerio et al., 2006), which is consistent across the lifespan of individuals with Williams syndrome (Mervis & Klein-Tasman, 2000). Nonetheless there are also reported to be higher frequency of behavioural problems such as hyperactivity and anxiety (Dykens, 2003; Udwin & Yule, 1991). There is also a common cognitive profile for individuals with Williams syndrome. Mean full scale IQ scores are in the mild intellectual disability range, with a relative strength in language but severe weakness in visuospatial cognition (M. A. Martens, S. J. Wilson, & D. C. Reutens, 2008; Morris & Mervis, 2000). Despite this relative strength in language, it is still not typical. There is delayed language acquisition, difficulty in use of pragmatics of language, poor relational vocabulary (e.g. spatial terms) but concrete vocabulary (nouns) are a relative strength (M. A. Martens et al., 2008; Mervis & John, 2010; Morris & Mervis, 2000).

### 1.2.4 Sensory Processing Atypicalities in Williams Syndrome

In comparison to autism, less is known about sensory processing in Williams Syndrome, and there have been few studies that use of questionnaires or interviews to address symptomatology in this area. There have been only two studies using the Sensory Profile questionnaire (Janes, Riby, & Rodgers, 2014; John & Mervis, 2010). Results from children and adolescents with Williams Syndrome using the Sensory Profile have suggested that there is hypersensitivity, for auditory, gustatory and proprioceptive sensory responses. By comparison there were few reports of hypersensitivity or hyposensitivity for vision or tactile modalities (Janes et al., 2014). John and Mervis (2010) also used the sensory profile in children with Williams Syndrome. In contrast to Janes and colleagues (2014) they found that using the Short Sensory Profile there was not only hypersensitivity, but also hyposensitivity, in young children with Williams syndrome (John & Mervis, 2010). Similar to autism, sensory processing atypicalities have been associated with repetitive behaviours (Riby, Janes, & Rodgers, 2013) and poorer adaptive and executive functioning (John & Mervis, 2010). Most investigations of sensory processing in Williams Syndrome have used experimental methods, the results of which will be reviewed in the section below on visual perception in the two developmental conditions.

## 1.3. Overview of Human Visual Processing

The human visual system comprises two main biological structures: the eye and the brain. Initially this overview will address the retina – geniculate – striate pathway (see figure 1.1), whilst the second part will overview primary visual cortical pathways (see figure 1.2). Before the retina – thalamus – striate cortex pathway, light emitted by a source, or reflected from a surface, enters the eye, it is here that visual perception begins. As light enters the eye it is refracted as it passes through the cornea where the light is focused onto the pupil and is further refracted as it passes through the lens. The pupil contracts and expands, moderated by the iris, depending on lighting conditions to let the optimal amount of light through the lens. The lens then combines with the cornea to focus the light onto the retina. In the retina light is absorbed by different types of photoreceptor cells. Inside these photoreceptor cells are different proteins called opsins. These opsins absorb light and transmit signals to the retinal ganglion cells. There are at least five types of opsin found in the retina: 3 cone opsins (photopsins I, II, and III), rhodopsin and melanopsin. All three of the opsins differ in their peak spectral sensitivity, but their spectral sensitivities overlap (see section below on Colour Vision). Photopsins are found in the cone photoreceptors and respond at photopic light levels (see section on colour vision). Rods are sensitive primarily in low light conditions. Melanopsin is expressed by the intrinsically photosensitive retinal ganglion cells and are sensitive to short wavelengths with a peak at 480 nm (Hattar et al., 2003; Lucas et al., 2003). Functional splits have also been suggested, with rods in the periphery of the retina more sensitive to motion detection than cones. These distinct photoreceptors' responses are then transformed into signals by the bipolar and ganglion cells. Ganglion cell receptive fields take different forms depending on the type of ganglion cell, with, for example, the midget ganglion cell having spatially opponent centre and surrounds, with the centre fed by a single L or M cone type in the fovea and fed by opposite sign inputs from cones in the surround (Hubel & Wiesel, 1959; Livingstone & Hubel, 1988). Cells are defined as either ON centre or OFF centre depending on whether light falling onto the centre of the ganglion cell is excitatory or inhibitory.

Action potentials from the ganglion cells are collated and sent down the optic nerve, preserving the relative spatial and temporal responses to the visual input. Information from each eye's optic nerve contains information from both visual hemifields and is transmitted to the lateral geniculate nucleus (LGN) of the thalamus. En route to the LGN, at the optic chiasm, the information from each optic nerve partially crosses, i.e. information from both nasal visual fields crosses over, whilst information from the temporal visual field does not cross over. After the optic chiasm, most information is transmitted to the lateral geniculate nucleus (LGN), with the remaining information sent to the suprachiasmatic nucleus or midbrain nuclei. This means that the left LGN receives information from the right visual field and vice versa for the right LGN. The LGN has six different layers which convey

different visual information. Layers 1, 4 and 6 receive input from the contralateral eye, whilst layers 2, 3 and 5 receive input from the ipsilateral eye. Cells within each layer are retinotopically mapped (although distorted to over-represent the central visual field) meaning that cells next to each other process information located in adjacent locations. Subdivisions of colour, form and motion signals from the retinal ganglion cells continue to project into the LGN. The magnocellular cells in layers 1 and 2 process information related to coarse depth and motion. The parvocellular pathway comprises the small cells of layers 3, 4, 5 and 6 and processes information relating to colour and form. A third cell type is also found in intercalated layers, between the major 6 layers of the LGN. These cells are koniocellular types and carry information in the "blue-yellow" colour channel (see section 2.1 for more details about colour vision).

The primary visual cortex (or striate cortex) is located posteriorly at the back of the brain in the occipital lobe. Like the LGN, the primary visual cortex is structured into different structural layers where each layer has distinct functional properties. Much of the input from the LGN (i.e. most visual information) is projected into different sections of layer 4 (V1). The magnocellular and parvocellular pathways predominately input to layers 4Cα and 4C beta respectively (Callaway & Wiser, 1996). Input from the koniocellular layers is less clear, although there is some evidence for them projecting to blobs and layers 2, 3 and 4α within the visual cortex (Hendry & Reid, 2000; Nassi & Callaway, 2010). Neurons in V1 have been identified as being either simple or complex (Hubel & Wiesel, 1962). Simple cells have defined ON and OFF regions that are like those defined in the retinal ganglion cells, but in addition these simple cells also respond selectively to certain orientations, positions or sizes in their receptive field. Complex cells respond similarly to simple cells with the exception that they are not as particular about the properties of the visual image, just whether it falls within their respective receptive field (Movshon, Thompson, & Tolhurst, 1978; Skottun et al., 1991). Other functional specificity has been observed in V1. For example, interblob cells are sensitive to orientation, e.g. simple cells (Hubel & Wiesel, 1959), whilst other cells are responsive to particular spatial frequencies or direction of movement (Bredfeldt & Ringach, 2002; DeAngelis, Ohzawa, & Freeman, 1995; DeValois & both Professors, 1988; Foster, Gaska, Nagler, & Pollen, 1985). This completes the overview of the retino-geniculate-striate pathway (figure 1.1).

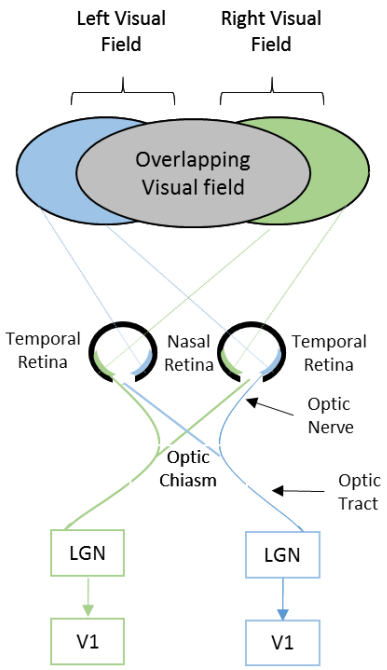



*Figure 1-1 Major anatomical structures of the retino-geniculate striate pathway.* The parallel processing of information from the left and right eyes is denoted by blue and green shading respectively.

From V1, the two functional "what" and where" streams (see figure 1.2) , which originate in the functionally distinct parvocellular and magnocellular retino-geniculate pathways, take distinct anatomical routes (Milner & Goodale, 2008; Ungerleider & Mishkin, 1982). The dorsal stream (or "where" stream) extends from V1 via projections to V2 and V5 and encodes spatial and motion information. The output of the dorsal stream is to the parietal cortex, which may indicate that it has a role in directing attention and planning movements. Conversely, the ventral stream (or "what" stream) extends from V1 via projections to V2 and V4, encoding object and colour information. It should also be noted that V4 receives input from the koniocellular pathway in the LGN (Hendry & Reid, 2000; Lysakowski, Standage, & Benevento, 1988). V4 subsequently outputs to the inferotemporal cortex which is predominantly involved in increasingly specific pattern recognition such as face recognition.

Although the dorsal and ventral streams are functionally different and processed in parallel, there is evidence that they interact at multiple levels (e.g. (Callaway & Wiser, 1996; Merigan & Maunsell, 1993)). In fact, many of the cells in each pathway fire in synchrony to ensure that the same visual stimulus is being encoded (Funke & Wŕgötter, 1997; Neven & Aertsen, 1992; Singer & Gray, 1995). This suggests that the visual system is organised optimally in a way that enables both functionally discrete but also functionally related processing, meaning that such functional separations are simplifications of the way in which visual information is processed in the brain (Schiller, 1996).

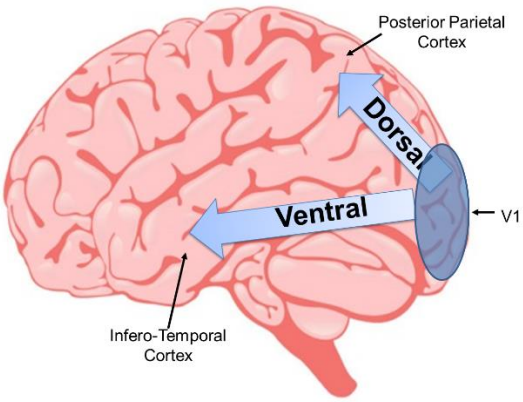



*Figure 1-2- Illustration of the cortical dorsal and ventral pathways.* Both originate from V1 and end in the parietal and temporal cortex respectively.

## 1.3.1 Colour Vision

### *1.3.1.1 Retinal processing of colour*

As discussed above, light waves enter the eye and are subsequently absorbed by different photoreceptors in the retina. The photo-receptors' spectral sensitivity curves overlap with one another, but each photo-receptor has its own peak spectral sensitivity: The L cones, which are sensitive to long wavelengths, with a peak sensitivity of approximately 565nm, the M cones, which are more sensitive to middle wavelengths and have a peak sensitivity of around 545nm, and S cones, most sensitive to short wave-lengths, with a peak around 440nm (see figure 1.3). The L and M cone types are densely packed and randomly distributed within the fovea, but this density decreases with distance from the fovea. There are fewer S cones compared to the L and M cones and the S cones are distributed more regularly and sparsely, and only outside the foveola. Rods are found only in the periphery and are greater numbers than cones. The L cone to M cone ratio varies greatly between individuals and between measurements, from 2:1 to 1:3 in one study and 0.4:1 to 16.5:1 in another (Carroll, Neitz, & Neitz, 2002; Cicerone & Nerger, 1989; Kremers et al., 2000). Photoreceptors are not individually able to identify the wavelengths of the photons which they absorb, i.e. the receptor's response is the same to each photon it captures, regardless of wavelength (the principle of univariance); the spectral sensitivity of the photoreceptor determines only the probability that it captures a photon of a wavelength. Individual photoreceptors are therefore unable to signal the wavelength that they have absorbed. It is not until the next stage of colour processing in the retinal bipolar cells, that some information about the spectral content of the light signal is recovered, through comparison of the outputs of the distinct photoreceptor types (see figure 1-4).

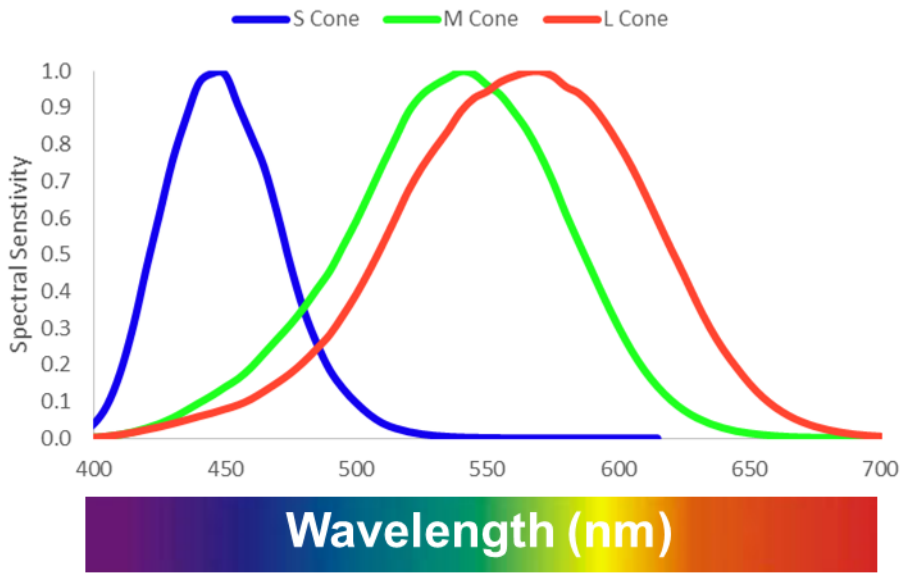



*Figure 1-3 - Representation of the spectral sensitivities with respect to wavelength (nm) for each cone class.* The S, M and L cones are represented by blue, green and red lines respectively. Spectral sensitivities are normalised to the maximal value of each class of cone. Spectral sensitivities are based on the Stockman and Sharpe 2° cone fundamentals (Stockman & Sharpe, 2000)

The bipolar cells output combine to send colour coded signals to the LGN (see figure 1.4). From these post-receptor stages of the human visual system, luminance is calculated from the sum of L and M cones (L + M). Subtracting M cone activity from L cones gives the "Red-Green" (RG) colour-opponent channel (L - M). Finally, the "Blue-Yellow" (BY) opponent channel is where the sum of activation of the L and M cones is subtracted from S cones (S – (L + M)). The three different "cardinal" colour axes are thought to be carried by distinct types of retinal ganglion cells. Although this model works well at simulating the relative inputs of the cone class. This does not account for other features of retinal ganglion cells, such as their spatial receptive field, inter-cell variability and ON and OFF types of ganglion cells. Physiological evidence from non-human primates suggests that S-ON and L+M-OFF P-cells provide a chromatic but not spatially opponent BY channel. The RG channel is both chromatically opponent and spatially opponent from the centre and surround of their receptive fields. In this case, chromatic opponent comes from differencing spectral sensitivities between L and M-ON centre mechanisms for the red dimension, and vice versa for green dimension. However, where these cells are also spatially opponent they also produce an achromatic response. Each possible colour is therefore associated with different On and Off responses of the retinal ganglion cells. The outputs of these channels remain segregated as they are transmitted to the parvocellular and magnocellular pathway in the LGN (Lee, 2011; Stockman & Brainard, 2009). Thus, from the retina the chromatic and spatial properties of the original signal have been processed and preserved as it is transmitted to the LGN.

In the LGN, the magnocellular pathway processes the luminance signal at relatively coarse spatial frequencies and high temporal frequencies, whereas the parvocellular pathway processes luminance information at higher spatial frequencies and chromatic information at lower spatial frequencies. The RG (at low spatial frequencies) and BY signals are carried by the parvocellular and koniocellular pathways respectively (Ingling & Martinez-Uriegas, 1983; Lee, 2011; Lennie & D'Zmura, 1987).

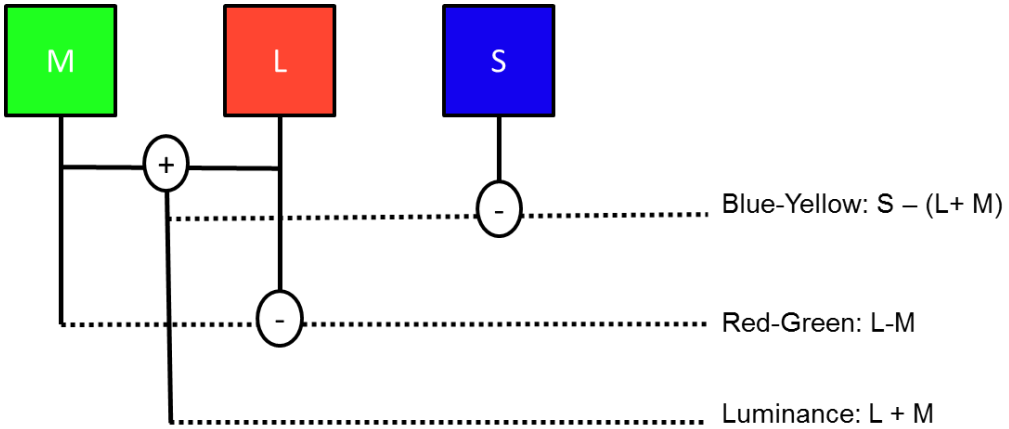



*Figure 1-4 - Visual depiction of the colour opponent channels that are present in the retinal ganglion cells and maintained through the LGN. The model includes the activation of the L, M and S cones which combine to produce chromatic and achromatic channels. Under this model there is assumed to be no contribution of the S cone to the luminance channel.*

### *1.3.1.2 The colour pathway beyond the retina: colour perception*

There has been a long held assumption that visual processing is modular, with specific cortical regions and functional pathways processing information for different visual functions, e.g. colour vision, form, motion etc. This section will assess the extent to which the evidence (predominately from animal studies) supports this modular view for colour perception. Colour information is processed through different levels in visual cortex. In V1 there are a variety of cell types that are colour responsive, the most common being either single to double opponent cells. Single-opponent cells respond to either colour or luminance signals, whilst double opponent cells respond to either colour information combined with spatial information. Despite the presence of both single and double opponent cells, V1 cells are also selectively responsive to luminance information (Lennie, Krauskopf & Sclar, 1990; although for possible RG selective cells see Engel, Zheng & Wandell, 1997). However, this is not to say that colour information is not processed in V1. Chromatic visual evoked potentials (VEPs) have found that there is a specific response for RG for chromatic gratings. The size of this response is attenuated at a certain spatial frequency. Similar findings of chromatic selectivity have also been observed in electrophysiology studies in V1 cells of non-human primates (Conway et al., 2010; Lu & Roe, 2008; Xiao, Casti, Xiao, & Kaplan, 2007). This suggests that colour information is specific in V1 but only for certain spatial frequencies. There is more evidence for colour-specific areas

within V2. Cortical projections from V1 are separate for parvocellular and magnocellular streams into the CO blobs in V2. Specific colour responses are found within the thin and pale CO stripes in V2 which receive input from the double opponent cells in V1 (see Shapley & Hawken, 2011 for review). Although it should be noted that V2 also processes other properties (e.g. orientation) suggesting that their function may be to process object properties but a distinct functional segregation between object properties and object location (Gegenfurtner, 2003).

The area V4 has been suggested as the "*centre for colour*" within the visual system (e.g. Van Essen & Zeki, 1988; Zeki, 1983). Many of these studies have been conducted on macaques and identified colour specific responses for cells within V4 related to colorimetric properties unlike in V1 which are more related to physiological responses (Bohon, Hermann, Hansen, & Conway, 2016; Kusunoki, Moutoussis, & Zeki, 2006; Zeki, 1980). Specifically, this colour specific response has been identified in response to be hue specific (Bohon et al., 2016; Conway, Moeller, & Tsao, 2007; Conway & Tsao, 2009; Harada et al., 2009; Lafer-Sousa & Conway, 2013). Moreover this cortical activity in V4 for response to colour has been seen to reflect the "colour wheel", i.e. cells that are responsive to red are adjacent to those that are responsive to purple (Conway et al., 2007; Conway & Tsao, 2009). However, the notion of V4 as a "colour centre" has been criticised. Firstly, V4 has been identified to have a role in shape, orientation visual attention (Gegenfurtner & Kiper, 2003; Pasupathy & Connor, 2002; Schiller, 1996). Whilst there have been many studies supporting specialised colour processing in V4 its function is not to only process colour but also basic geometric shapes and orientation. For example studies in macaques with ablations to V4 (i.e. induced cerebral achromatopsia) were still found to be able to discriminate between colours (Heywood, Gadotti, & Cowey, 1992; Heywood & Kentridge, 2003). Furthermore a meta-analysis of patients with cerebral achromatopsia found that when an overlap in the ventral occipital cortex of where the lesions was, that the most common region were similar to those that were related to prosopagnosia (face-blindness) (Bouvier & Engel, 2006). This suggests two things, firstly it casts doubt as to whether colour by itself is a single modular function but in fact there is more general visual processing of object information. Secondly that this overlap in function may suggest that impairment of low-level colour perception may be related to visual processing of faces. Other lesion studies have also suggested that the infero-temporal cortex has a role in processing colour.

Further processing of colour, including linguistic categorisation and representation, emotional responses to colour, colour preference or memory of objects with a colour diagnostic colour (e.g. bananas are yellow), is not processed in these basic visual areas, but instead processed in higher order cortical areas such as the frontal regions. It is unlikely that there is a specific colour "module" within the cortex, instead there is evidence that there are functional components (e.g. cells, blobs etc) that specifically respond to colour throughout the cortical processing of visual information from

early partitioning in visual processing and colour specific information is retained throughout the primary and associated visual areas, including some highly specific colour responsive cells in V1, V2 and V4.

## 1.4. Atypicalities of Visual Processing in the Developmental Conditions Autism and Williams syndrome

Studying visual processing in developmental conditions such as autism and Williams Syndrome allows for the different levels and areas of neural processing to be evaluated. Through examining colour, a perceptual property that is processed through multiple stages in the visual pathway, beginning with the initial reception of light at the retina, it is possible to assess visual function at its earliest stage as well as later stages. Colour is processed through the ventral stream and inputs into higher order visual processing such as face and object recognition. Atypical face and object processing has been identified in both autism and Williams Syndrome but it is possible that this atypicality may be related to either more general ventral stream dysfunction or that the atypicality is caused by abnormal upstream processing from face processing (e.g. LGN or V1). This section will focus on psychophysical and neuroimaging studies on visual processing in autism and Williams syndrome. Inclusion of studies relating “autistic traits” to visual perception is beyond the scope of this thesis.

### 1.4.1 Visual Processing in Autism Overview

A varied visual profile exists in autism, this characterised by relative strengths and deficits both within and between different visual functions. Dorsal stream function has been found to be dysfunctional in children and adolescents with autism, with reports both of higher motion detection thresholds (Spencer et al., 2000) and poorer biological motion identification (e.g. (Annaz et al., 2010)). However, not all motion processing is impaired. First-order motion detection (identification of movement) is the same as chronological-age controls but detection of second-order motion (identification of a moving contour defined by contrast) was worse or the ASC group (Bertone, Mottron, Jelenic, & Faubert, 2003). Although motion processing is impaired, variation in experimental procedures between studies may contribute to this. For example, the visual angle varies between studies, making stimulus detection easier or harder depending on whether the visual angle has increased or decreased respectively. Another issue is the wide inter-individual variability within ASC, the studies above predominantly use high functioning ASC participants as opposed lower functioning participants. In addition, diagnosis also affects performance. For example, higher motion coherence thresholds are found for individuals with autism compared to Asperger’s Syndrome (Spencer & O'Brien, 2006), see (Simmons et al., 2009) for further discussion). This shows that there needs to be careful consideration given to diagnosis and level of ability when interpreting and designing studies relating to development, and particularly developmental conditions such as ASC.

In comparison to the dorsal stream, evidence for ventral stream atypicalities is more complex. Form perception has been shown to be relatively intact when compared to motion processing and with respect to TD controls (e.g. (Milne, Swettenham, & Campbell; Spencer et al., 2000); although see (Spencer & O'Brien, 2006)). Orientation discrimination is another ventral stream function that has been studied in autism. There is reported intact or superior orientation discrimination using either gabor or sinusoidal gratings for oblique and veridical presentations compared to non-verbal mental age equivalents (Dickinson, Bruyns-Haylett, Smith, Jones, & Milne, 2016; Schwarzkopf, Anderson, de Haas, White, & Rees, 2014; Shafai, Armstrong, Iarocci, & Oruc, 2015). Interesting, in the study by Dickinson and colleagues (2016) a subset of participants also had induced oscillatory EEG activity recorded in response to gratings. They found that performance on the orientation discrimination task was correlated with peak gamma frequency when collapsed across both control and autism groups.

Faster performance has also been found on the "Embedded figures task" (e.g.(Pellicano, Gibson, Maybery, Durkin, & Badcock, 2005)). It has been claimed that faster performance is the result of a feature-based search strategy whereby individuals with ASC rely more on the processing of local features over global features (Plaisted, Swettenham, & Rees, 1999). Importantly, this is different to the strategy used by typically developing individuals. Recent research suggests that this local bias in autism is consistent for visuospatial tasks but not auditory tasks (D'Souza, Booth, Connolly, Happé, & Karmiloff-Smith, 2015). Face and object perception have also been shown to be atypical. Individuals with autism have been shown to attend more to the local features of faces (e.g. mouth) rather than to process faces holistically (e.g. (Deruelle, Rondan, Gepner, & Tardif, 2004)). Studies using ERPs in children with autism have found reduced neural responses to faces compared to objects (e.g. (Dawson, Webb, & McPartland, 2005)). However decreased ERP amplitudes for objects have also been found in ASC compared to same age controls (Webb, Dawson, Bernier, & Panagiotides, 2006). These findings suggest that there may be a more non-specific visual processing deficit which may be related to a local processing bias in ASC (Behrmann, Thomas, & Humphreys, 2006; Jemel, Mottron, & Dawson, 2006).

#### *1.4.1.1 Colour Perception in Autism*

Colour perception, by comparison, has sparsely been studied relative to other visual functions in ASC. There is anecdotal evidence of both colour obsessions and colour avoidance (Bogdashina, 2003; Ludlow, Heaton, Hill, & Franklin, 2014; Ludlow & Wilkins, 2009), for example, insistence on food or walls being a certain colour or a strong negative response to the presence of certain colour (Ludlow & Wilkins, 2009). However, it is not clear whether these behaviours arise from a specific deficit or difference in colour processing or whether they are similar in nature to other repetitive behaviours seen in autism. The actual prevalence of such exaggerated responses to colour is not known, nor

whether these may reflect atypical processing of colour in the early visual pathway in ASC or higher-level behavioural or cognitive atypicalities, such as more generalised ritualistic or repetitive behaviours.

Reduced chromatic discrimination, measured via both standardised tests and specialised psychophysical tasks, has been found in both young adolescents and adults with ASC ((Franklin, Sowden, Burley, Notman, & Alder, 2008; Franklin, Sowden, et al., 2010; Heaton, Ludlow, & Roberson, 2008; Hurlbert, Loveridge, Ling, Kourkoulou, & Leekam, 2011). Two studies have used the standardised Farnsworth-Munsell 100-Hue Test (FM100) to measure chromatic discrimination. The FM100 is a manual cap sorting task in which the participant's task is to place coloured caps in order along a colour gradient (e.g. purple to pink). The lightness and saturation of the cap colours is constant across the set, with just-noticeable differences in hue only between adjacent coloured caps. Error scores are calculated based on differences between each pair of neighbouring caps. The sum of all error scores is then taken as a measure of chromatic discrimination. Poorer chromatic discrimination has been found for young adolescents with high-functioning autism relative to chronological and mental age controls (Franklin, Sowden, et al., 2010), with no significant differences in the relative deficit between the red-green and blue-yellow cardinal colour-opponent axes. A similar result for FM100 performance has also been reported for young adults with autism (Hurlbert et al., 2011).

Franklin and colleagues (2010) also used a custom psychophysical test of chromatic discrimination, in which participants had to discriminate the colour difference between two halves of a disk displayed on a computer monitor to determine and report whether the bisecting line was slanted to either the right or the left. There were three different conditions based on the cardinal colour axes in Macleod-Boynton colour space. Young adolescents with high-functioning autism completed the task for two of the three cardinal axes (the luminance axis and one chromatic axis). Again, it was found that there was poorer chromatic discrimination relative to chronological and mental age controls. Crucially there was no difference between groups in luminance thresholds, suggesting that the chromatic discrimination difference is not due to a general inability to complete the task. There was also no difference between either red-green or blue-yellow thresholds between participants with and without, implying that there may not be a specific deficit in discrimination of a colour axis. However, because individual participants did not complete both chromatic axes, it is not clear whether there is a specific deficit relative to a chromatic axis.

Other studies have assessed colour discrimination, specifically around colour category boundaries. Franklin and colleagues (2008) assessed chromatic discrimination around the blue-green category boundary using stimuli whose colour differences were equally spaced, in units of 2.5 Munsell steps; stimuli were drawn from within a category (two blue and two green stimuli) or across the blue-green

category boundary. Participants had to locate whether a coloured circle appeared on either the left or right side of the screen. Young adolescents with high-functioning autism had poorer accuracy for both within and between colour categories when compared to mental and chronological age matched controls. There was no significant group by within/between category interaction, suggesting that there is poorer chromatic discrimination regardless of whether colours cross linguistic boundaries. A second study required participants to identify the odd colour out from three coloured patches. Low functioning young adolescents with autism were found to have poorer chromatic discrimination than chronological age matched typically developing controls but similar chromatic discrimination to a moderate learning disability control group (P. Heaton et al., 2008). This result was interpreted as evidence for poorer chromatic discrimination in autism, but it cannot be certain whether this is due to low ability or specifically to autism. There was also no control task, so it is not certain whether the reduced performance is due to poorer chromatic discrimination or the ability to identify "odd one out". An additional problem with both the Heaton et al., (2008) and Franklin et al. (2008) studies is that they both use Munsell colour space to give the stimuli for their discrimination experiments. The Munsell colour space is a perceptual colour space (but only valid directly under Illuminant C, daylight) and not a physiological colour space based on cone activations, making it difficult to map on the discrimination to underlying physiological mechanisms.

McCleery and colleagues (2007) found chromatic discrimination was not different between 6 month infants at high and low risk for developing autism. Instead in the 6-month old infants at high risk for autism there was reduced luminance contrast sensitivity (black-white gratings) but not chromatic contrast sensitivity (red-green gratings) when compared to low-risk infants (McCleery, Allman, Carver, & Dobkins, 2007). There was also greater difference between the luminance and red-green contrast sensitivity thresholds in the high-risk group compared to the low risk group. Interestingly, in a follow-on study, the authors anecdotally note how one infant from each group later developed autism (i.e. one infant from both the low and high risk groups developed autism) both displayed higher differences between luminance/Red-Green thresholds than each of their respective groups. It also contradicts the results in adolescents and young adults with autism where there is poorer chromatic discrimination but no reported difference for luminance discrimination when compared to controls (Franklin, Sowden, et al., 2008; Franklin, Sowden, et al., 2010; P. Heaton et al., 2008; Hurlbert et al., 2011). The extent to which these differences in results reflect either the heterogeneity in autism or different developmental trajectories of luminance and chromatic discrimination between autism and typically development is unclear.

The studies above strongly indicate that there is reduced chromatic discrimination in young adolescents and young adults with autism spectrum disorder. But chromatic discrimination varies with chronological age e.g. (Barbur & Rodriguez-Carmona, In Press; Knoblauch et al., 1987; Paramei

& Oakley, 2014) and it is unclear at what age these differences in chromatic discrimination occur and whether they reflect deviancy or delay. Additional questions have been raised as to the methodology of chromatic discrimination tests and how these relate to the overall intellectual ability of the participant. Performance on the FM100 has been found to be associated with non-verbal ability in adults (Hurlbert et al., 2011). Likewise in the (P. Heaton et al., 2008) task it is difficult to dissociate whether poor performance in the autism group was due to low ability or to having autism. The extent to which ability affects performance on psychophysical tests in the general population is beginning to be investigated more thoroughly in different modalities (Acton & Schroeder, 2001; Li, Jordanova, & Lindenberger, 1998; Melnick, Harrison, Park, Bennetto, & Tadin, 2013) and it is important to assess whether this association exists in measures of sensory discrimination in developmental disorders. In addition, the studies described above do not characterise the severity of autism samples with additional measures, making it difficult to assess whether the severity of autism is related to the severity of chromatic discrimination deficiency or whether the latter is related to differences in visual sensory processing sensitivities.

Colour memory deficits have also been identified in young adolescents with autism (Franklin et al., 2008; Heaton et al., 2008). Heaton and Colleagues (2008) reported that these deficits significantly negatively correlated with Non-Verbal IQ for the ASC group only. Heaton et al. (2008) suggest that lower-functioning ASC individuals adopt a perceptual rather than linguistic strategy to encode and recall colours, in contrast to the TD or moderate learning disability control groups. Franklin et al (2008) found that there was poorer colour, but not form, memory in adolescents with autism compared to chronological controls. However, in both these studies few colours were used. In both studies the stimuli were not equal for lightness or saturation between colours, meaning it is unclear whether these factors are driving better/poorer memory of colours.

Some studies have utilised colour as a mechanism for therapeutic intervention (see also section below). There is evidence that the use of coloured overlays or glasses in young adolescents with autism improves the reading speed (Ludlow, Taylor-Whiffen, & Wilkins, 2012; Ludlow, Wilkins, & Heaton, 2006; Whitaker, Jones, Wilkins, & Roberson, 2015) but not overall reading accuracy, in comparison to chronological age and verbal matched controls. Whilst this is a consistent result it should also be noted that this is a relatively small effect (tantamount to approximately a word per 6 or a 10% increase). Two of these studies also tested the effect of coloured overlays on emotion recognition (Ludlow et al., 2012; Whitaker et al., 2015). Ludlow and colleagues (2012) tested young adolescents on a child version of the Minds Eyes test. In this test participants are required to identify an emotion on the basis of a picture of a face in which only the eyes are present. They found that there was no significant effect of overlay but that the accuracy of the autism group was significantly lower than chronological age and verbal ability matches. There was also a significant group by

overlay interaction, which was driven by relatively higher accuracy in the autism group when a coloured overlay was used compared to without, whilst there was no difference in the control group. However, it should be noted that this effect equated to an increase of only one correct answer from 28 trials. Performance by the autism group when using coloured overlays was correlated between performance on the rate of reading and on the Mind's Eye tasks. It was found that there mild correlation between the differences in performances on both tasks when using and not using the coloured overlays. The authors suggest that this demonstrates the value of using coloured overlays. It should be noted however that 3/15 participants in the autism group showed poorer performance on both tasks using coloured overlays, whilst a further two showed no improvement on the Mind's Eye task, suggesting that coloured overlays do not work for all children with autism and are in some cases counter-productive. Whitaker and colleagues (2015) investigated whether the use of coloured overlays could also increase accuracy in the identification of emotional intensity shown in faces. Here participants asked to select the face that showed the most emotional intensity from two possible faces. They found that the adolescents with autism improved their rate of reading when using coloured overlays. The authors also found that when using the coloured overlays that were significantly better at identifying the "emotional intensity" of a face. This task however is ambiguous and combines many different emotions. Also like previous studies on coloured overlays the study does not address that 7/16 participants with autism had poorer performance on one task when using the overlays.

Despite these studies, it is still unknown whether coloured overlays/tints provide long term benefits to individuals with autism. Likewise, given the wide range of colours available for the overlays/tint, it is unclear as to the mechanisms underlying the role of colour in this intervention, and the extent to which it is related to either initial sensory processing of colour or how colour is used in higher order processes. For example, it is unclear whether the improvement results from a higher-level association (e.g. related to object memories) to specific colours, or whether the coloured overlays act to change the low-level visible contrast of the stimuli. Likewise, it is unclear why certain individuals with autism are either aided or inhibited with the use of coloured overlays/tints and possible reasons have not been explored. It should also be noted that these studies are predominantly done with high functioning individuals with autism, so it is also unclear the extent to which coloured filters would benefit low functioning individuals with autism.

Other evidence for reduced sensitivity in the colour pathway in autism comes from an EEG study (Fujita, Yamasaki, Kamio, Hirose, & Tobimatsu, 2011). Measuring steady state chromatic visual evoked potentials (VEP) to chromatic gratings, Fujita and colleagues (2011) found that peak latency was longer for adolescents and young adults with autism, suggesting a reduced neural response to colour in the primary visual cortex. However, the spatial frequency varied between the chromatic

and achromatic gratings, making it unclear whether the effect is due to difficulty in processing certain spatial frequencies or due to the chromatic contrast of the gratings. Although the mean ages of the two groups was similar, age was not directly controlled for. In both groups, there is a large age range (autism group: 21 years, TD group: 17 years) the autism group was also older than the TD group. The magnitude of chromatic VEPs has been shown to change with age (Crognale, 2002; Crognale, Kelly, Weiss, & Teller, 1998; Tobimatsu, Kurita-Tashima, Nakayama-Hiromatsu, Akazawa, & Kato, 1993). Given that the spread of the different ages was not reported, it is difficult to determine the extent to which age may be influencing the results. Furthermore, it is not known whether individuals with autism will follow the same developmental trajectory for VEPs.

### 1.4.2 Visual Processing in Williams Syndrome Overview

In Williams Syndrome, severe impairments have been found for dorsal stream functions which are present in childhood and persist into adulthood (Atkinson et al., 2006). Visuo-spatial functioning, in particular motion perception (e.g. (Atkinson, 2000; Atkinson et al., 1997) and also planning of motor movements where spatial judgements are required (Cowie, Braddick, & Atkinson, 2012), are particularly impaired. Nonetheless, not all dorsal stream function is atypical. Identification of biological motion is still intact in WS relative to chronological age-matched controls , but form detection from motion is still impaired relative to mental age-matched controls (J.E. Reiss, J.E. Hoffman, & B. Landau, 2005a), suggesting a more selective dorsal stream deficit in Williams syndrome.

In contrast, some evidence suggests that ventral stream function may be a relative strength in Williams syndrome, compared to other visual and cognitive functions. Both form coherence thresholds and facial expression identification have been found to be like mental-age controls (e.g. Atkinson et al., 1997, Farran, Jarrold & Gathercole, 2003; Deruelle et al., 1999; Gagliardi et al., 2003). Nonetheless ventral stream functioning is not typical. Similar to ASC, faces are also processed atypically in WS. WS have been shown to use a featural strategy (e.g. Karmiloff-Smith et al., 2004) with impaired development of configural processing (e.g. Deruelle et al., 1999; although see Deruelle et al., 2006). Form perception is also not typical in WS. For example, there is poorer shape identification in WS when form is defined by global variations (Atkinson et al., 2003).

#### *1.4.2.1 Colour Perception in Williams Syndrome*

Colour perception has scarcely been studied in WS. There has been one direct study of colour perception in Williams syndrome (Farran, Cranwell, Alvarez, & Franklin, 2013) reports that WS adolescents have similar chromatic discrimination relative to mental-age controls but poorer chromatic discrimination relative to chronological-aged matched controls, measured with the FM100. This study also used a forced choice verbal naming across the blue-green colour boundary.

There was no difference in the categorisation of either blues or green in the Williams syndrome group compared to either the mental age or chronological age control groups. A final experiment in this study used a visual search with blue and green coloured targets and distractors. Targets and distractors were either from within the same colour category (i.e. two blues) or between a colour category (i.e. a blue and a green). All groups found between colour category targets faster than within colour categories. The Williams syndrome performance was in line with their mental age but poorer than their chronological age. These results suggest that colour discrimination and cognition may be relatively intact in Williams syndrome with respective to their mental age.

Two other studies have not assessed colour perception directly but have used colour in a control task. Vicari and colleagues (2005) found that adolescents with Williams syndrome were able to group similar colours together (Vicari, Belluci, & Carlesimo, 2005). Farran and Jarrold (2005) used a naming task for colours in the blue/green region of colour space and found differences in the proportion of how colours were named relative to mental age controls (Farran & Jarrold, 2005). Colour has also been used as a mechanism to help aid the learning of a virtual route (Farran, Courbois, Van Herwegen, Cruickshank, & Blades, 2012). In this study, it was also noted that the Williams Syndrome group were more likely to give an atypical name to a colour (e.g. tooth paste). Unfortunately, these responses were only observed and not systematically recorded. However more importantly, it should be noted that these studies the coloured stimuli were not the main focus of the study and as such their stimuli were not precisely controlled and varied in saturation, hue and lightness between stimuli.

## 1.5. Neural correlates and theoretical explanations of sensory processing atypicalities in developmental disorders

### 1.5.1 Perceptual theories of atypical sensory processing in Autism

The weak central coherence theory was originally proposed after observations that individuals with autism have difficulty in processing information in context because of an increased focus on details rather than the “bigger picture” (Frith, 1989; Happé & Frith, 2006). It was proposed that this more detail focused cognitive style enables better performance on tasks where the focus is on the identification of local factors at the expense of global processing. This style may lead to a more detailed focus approach, for example, better performance on the embedded figures task by individuals with autism relative to mental age controls (Pellicano et al., 2005).

The perceptual analogue of the weak central coherence is the enhanced perceptual functioning theory (Mottron & Burack, 2001; Mottron, Dawson, Soulieres, Hubert, & Burack, 2006). However, unlike the weak central coherence theory, enhanced perceptual functioning does not propose a weakness in processing global information. Additionally, it is clear that enhanced perceptual

functioning does not exist in all perceptual domains, given, for example, the reports of reduced chromatic discrimination in autism (Franklin, Sowden, et al., 2010). In light of such results, the theory has recently been revised to distinguish between simple and complex visual percepts (Bertone & Faubert, 2006). Simple visual percepts are those which are predominantly processed in the primary visual cortex (e.g. is there movement?), whereas complex stimuli are dependent on integration of signals (e.g. in which direction is there movement?). Processing of simple information has been found to be similar to mental and chronological age matched controls in the vision for motion perception (Bertone et al., 2003; Bertone, Mottron, Jelenic, & Faubert, 2005). This is also seen in other modalities, for example enhanced pitch discrimination (Heaton et al., 1998). Yet other visual functions show poorer discriminations and this doesn't explain the poorer chromatic discrimination. The lack of a consistent finding of enhancement or reduction in perceptual discrimination in ASC within and across different sensory modalities may also, of course, relate to other differences underlying the ability to process perceptual information that vary between individuals across modalities and with the complexity of the task, which have been suggested to be independent of other factors such as ability (Meilleur, Berthiaume, Bertone, & Mottron, 2014).

Another theory of autism based on differences in perceptual processing posits that there is increased neural noise in autism (Dakin & Frith, 2005; Simmons et al., 2009). Here noise refers to increased neural variability. Although the addition of noise under most conditions would decrease the signal to noise ratio, it is possible under certain circumstances that the signal may be amplified (i.e. increase in signal to noise ratio) under certain conditions of non-linearity of signal via stochastic resonance. This would explain both enhanced and poorer perceptual discrimination findings such as enhanced first order motion but impaired second order motion in autism (Bertone et al., 2003). There is further support for increased neural noise in individuals with autism from findings of higher inter-trial variability reported using both EEG (Milne, 2011) and fMRI (Dinstein et al., 2012) (although see (Schwarzkopf et al., 2014)). But it is not clear whether the source of this neural noise is due to atypical anatomical structures, atypical connectivity or imbalance between excitatory/inhibitory signals in individuals with autism. At present, there are few psychophysical studies that investigate the neural noise theory. One study has used motion coherence to investigate levels of neural noise, using a technique that allows measurement of both internal and external noise. There was no difference between performance of the autism and a mental/chronological control matched group for the internal noise condition, but a better performance in the external noise condition (Manning, Dakin, Tibber, & Pellicano, 2014), suggesting that individuals with autism are better at combining a wider range of direction variability compared despite external noise. It has also been suggested that there is *decreased* neural noise in autism, as appropriate levels of noise can stop a neural network from reaching a local minima (Davis & Plaisted-Grant, 2014). Davis and Plaisted-Grant (2014) argue

that reduced neural noise in this context can affect perceptual discrimination (such as enhanced performance on visual search) and could also lead to increased shifts in neural networks, the lack of which can lead to poorer performance of individuals with autism on tasks such as binocular rivalry where there are perceptual shifts (Robertson, Kravitz, Freyberg, Baron-Cohen, & Baker, 2013). Recent attempts have been made to further link this variability to an imbalance in underlying neuro-transmitters such as GABA (Robertson, Ratai, & Kanwisher, 2016; Rubenstein & Merzenich, 2003). Such links are based upon visual psychophysical tasks which are highly correlated with GABA, for example poorer performance on binocular rivalry but better orientation discrimination (Dickinson et al., 2016; Robertson et al., 2013). Nonetheless whether there is either *increased* or *decreased* neural noise is uncertain due to lack of supporting/discounting evidence and failure to parsimoniously reconcile existing findings by either approach to neural noise. It also unclear, how colour discrimination would fit into neural noise models of colour.

### 1.5.2 Neural correlates of sensory processing atypicalities in autism

The next section will provide an overview of neuroimaging studies in autism with particular focus on studies related to visual processing, although considerations will be given to other areas of sensory processing and neuroanatomical structures.

As autism is a neurodevelopmental condition there has been considerable effort to understand the underlying anatomical and functional neural profile (Amaral, Schumann, & Nordahl, 2008; Lainhart, 2015). There is evidence for accelerated early brain overgrowth followed by subsequent decelerated growth (Courchesne, Carper, & Akshoomoff, 2003; Dawson, 2008). There is evidence for young children with autism having larger head circumferences and brain sizes which may be due to increased cell proliferation of both grey and white matter (Hazlett et al., 2011). Although this difference between grey and white matter may not continue into adulthood, where differences have been found for white matter but not grey matter (Barnea-Goraly et al., 2004; Hyde, Samson, Evans, & Mottron, 2010). Furthermore there is specificity in the cortical locations of these white and grey matter differences, with grey matter increases in the primary auditory and visual cortical areas (Hyde et al., 2010) and also reduced white matter bilaterally in the insula cortex (which receives projections from the ventral stream of the visual pathway) and in the right superior temporal gyrus (Cheng et al., 2010). Other differences are most commonly found for the frontal lobe, where there are increases in volume of the dorsolateral prefrontal cortex and medial frontal cortex (Carper & Courchesne, 2005). However it is not clear whether the extent of these differences continue into adulthood (Amaral et al., 2008; Haar, Berman, Behrmann, & Dinstein, 2014).

Increased cortical thickness has also been reported over the whole cerebral cortex, but especially in the temporal and parietal cortices (Hardan, Muddasani, Vemulapalli, Keshavan, & Minshew, 2006) in 8-12 year olds with autism. In contrast, in adults there is evidence for increased cortical thinning in frontal, parietal and temporal regions suggesting that there are two cortical growth periods (one characterised by cortical overgrowth and the second by increased cortical thinning) in structural MRI (Hadjikhani, Joseph, Snyder, & Tager-Flusberg, 2006; Wallace, Dankner, Kenworthy, Giedd, & Martin, 2010). This suggestion has been further supported by stereological studies using post-mortem autistic brains which find reduced neuronal density (van Kooten et al., 2008). There is also evidence for differences in subcortical architecture where the mini-column structure is smaller and more densely packed in the frontal and temporal regions (Casanova, Buxhoeveden, Switala, & Roy, 2002) suggesting complex differences at different anatomical levels (Schumann & Nordahl, 2011).

These complex brain anatomical differences seen in autism, combined with the age-specificity of the differences, highlight the importance of the developmental context in which the majority of these studies are conducted (Courchesne, Campbell, & Solso, 2011; Lainhart, 2015). It is not possible to say whether the differences in brain sizes are due to accelerated growth because the studies are cross-sectional. Although the ideal study to address these issues would use the same individuals longitudinally, such a study is yet to be conducted. It may also be that the certain differences in structure may relate to specific behavioural subcategories of individuals with autism (Haar et al., 2014).

In addition to these anatomical differences in autism there has also been evidence for atypical functional connectivity in autism. The first and most commonly observed finding is reduced functional connectivity in autism between the frontal lobe and the other remaining lobes (Courchesne & Pierce, 2005; Just, Cherkassky, Keller, & Minshew, 2004). Specifically this reduced connectivity has also been found between the frontal cortex and the insula (Ebisch et al., 2011) and the primary visual cortex (Villalobos, Mizuno, Dahl, Kemmotsu, & Müller, 2005). This atypical connectivity is characterised by local connectivity between neurons within a single cortex but reduced long range connectivity between brain regions.

This suggests that there is not just under connectivity but also over connectivity. Therefore a better conceptualisation may be one of atypical connectivity dependent on the brain regions and underlying neural circuitry involved (see (Maximo, Cadena, & Kana, 2014) for review). This disrupted connectivity may cause imbalances in the excitatory and inhibitory responses in the brain which in turn may cause altered firing in local areas and introduce more neural noise into the signal (Rubenstein & Merzenich, 2003).

With respect specifically to functional circuitry of the visual pathway, the primary visual cortex has shown typical retinopic mapping (Hadjikhani et al., 2004; Schwarzkopf et al., 2014) but larger receptive fields in adults with autism relative to chronological age matches (Schwarzkopf et al., 2014), though whether these findings also apply in children is unknown.

Similar to the anatomical differences, there is also the need to consider developmental changes in functional connectivity (Maximo et al., 2014; Uddin, Supekar, & Menon, 2013).For example in typical development it has been found that there is an increase in the strength of these connections with age from childhood to adulthood (Hagmann et al., 2010) but not necessarily in autism where there is increased functional connectivity compared to typical controls but reduced functional connectivity in adults (Uddin et al., 2013). Again, this example highlights the need to interpret both anatomical and connectivity results in a developmental context. One notable study has attempted to bridge neuroimaging studies with behavioural correlates of behaviour. Green and colleagues (2015) measured sensory processing using the Sensory Profile and low-level sensory stimuli (rotating colour wheel and beep sound, either singularly or jointly) in adolescents with autism using fMRI and found that there was a significant positive correlation between scores of sensory over-responsivity and increased neural activity in the amygdala, primary auditory and somato-sensory cortices (Green et al., 2015). Furthermore, those children who were not classed (by the Sensory Profile) as being over responsive showed different functional connectivity to frontal regions, suggesting that these individuals could be better able to regulate their behaviour. The results from this study suggest that there is a link between sensory over-responsivity, for auditory and tactile sense, and increased activation in primary sensory cortices (Green et al., 2015), but it is still unclear whether how this relates to children who have Sensory Profile scores which are classed as under responsive. There were also a low number of high-functioning participants, and a wide age range, meaning that it is unclear to which extent these are representative of cortical to behaviour relationships in autism.

The studies outlined above show that there are clear anatomical, connectivity and functional differences in adults with autism compared to adults without autism. With specific reference to primary visual areas, there are larger receptive fields, but similar retinopic mapping, and increased cortical activity in primary visual areas.

### 1.5.3. Perceptual Theories of Williams Syndrome

The profound visuospatial deficits observed in Williams Syndrome have led to a hypothesis that there is a specific *dorsal stream deficit* in Williams Syndrome (Atkinson, 2000; Atkinson et al., 2006; Atkinson et al., 1997). The hypothesis assumes that the dorsal stream is at increased risk because it follows a longer developmental trajectory than the ventral stream, making it more at risk to insult

during development (Braddick, Atkinson, & Wattam-Bell, 2003). The differential neural architecture in Williams Syndrome in dorsal stream areas (e.g. Intra-parietal sulcus) supports this idea (Eckert, Galaburda, et al., 2006). The profound visuospatial and motion processing difficulties in Williams Syndrome previously found support that there is a dorsal stream deficit in Williams Syndrome. On the other hand, ventral stream visual function is not typical either, suggesting that the entire visual phenotype of Williams Syndrome cannot be explained by a dorsal stream deficit.

#### 1.5.3.2 Neural correlates of atypical sensory processing in WS

Like autism, Williams Syndrome is a neurodevelopmental condition and so understanding the underlying neural anatomical structure will help to understand the condition further. In addition, given that the genetic deletions that cause Williams Syndrome are known, the effect of these genes on brain development may be directly studied.

Generally, the whole brain volume of individuals with Williams Syndrome is reduced by approximately 11-13% (Boddaert et al., 2006; Cherniske et al., 2004; Eckert, Tenforde, et al., 2006; Galaburda & Bellugi, 2000; Reiss et al., 2004; Reiss et al., 2000; Thompson et al., 2005) and this volume difference is consistently found regardless of age (Cherniske et al., 2004). Yet the reduction is not uniform across the whole brain. There is reduced grey matter volume in the thalamus and occipital lobe, as well reduced grey matter density in areas that are strongly related to visuospatial ability (which is a relative weakness in Williams Syndrome) (Boddaert et al., 2006; Reiss et al., 2004). There is also greater gyrification (Gaser et al., 2006) and sulcus depth (Van Essen et al., 2006) in the occipital cortex. Furthermore there is increased neural density in the visual cortex, but this is only significant in the left hemisphere layer IVCβ, which is a layer which receives input from parvocellular cells in the lateral geniculate nucleus, although there are also trends towards increases in small neuron numbers/densities in other left hemisphere V1 layers IVA, IVCα, IVCβ, V and VI (Galaburda & Bellugi, 2000; Galaburda, Holinger, Bellugi, & Sherman, 2002). Related to colour, layers IVA and IVCβ are layers receive colour information from the koniocellular and parvocellular layers within the LGN. As in autism, it is possible that these differences in anatomical structures in primary visual areas may affect visual processing in Williams Syndrome.

There are also increases in the grey matter of areas that underpin relative strengths in Williams Syndrome: face processing (orbital/medial prefrontal cortices, anterior cingulate, insular cortex, fusiform gyrus and the superior temporal gyrus) and those with emotion (amygdala) (Golarai et al., 2010; Reiss et al., 2004). However, the ventral stream is not typical either, where there is also atypical anatomical connectivity between the fronto-parietal and temporo-parietal regions in Williams Syndrome. There is also increased gyrification and cortical thickness in the perisylvian region, which includes the temporal gyrus (Meda, Pryweller, & Thornton-Wells, 2012; Thompson et

al., 2005), as well as reduced white matter connectivity between the anterior insula, amygdala and orbitofrontal cortex in the left hemisphere (Jabbi et al., 2012). However there is also over-connectivity of white matter in the right hemisphere along the superior longitudinal fasciculus (SLF, a white matter pathway which is associated with the dorsal stream) and the inferior longitudinal fasciculus, a white matter pathway which is associated with the ventral stream (Hoeft et al., 2007).

In addition to the increase in amygdala size, there are also multiple atypical connections. Decreased white matter density is observed between the amygdala and the orbitofrontal cortex (Avery, Thornton-Wells, Anderson, & Blackford, 2012) which may underlie the "stranger danger" difficulties seen in Williams Syndrome. There is also reduced FA connectivity between the amygdala and the fusiform face area, as well as less functional connectivity between the temporal lobe and prefrontal/parietal regions (Sarpal et al., 2008). There are also links between brain structure in Williams Syndrome and cognitive function, e.g. intra-parietal sulcus and spatial performance (Meyer-Lindenberg, Mervis, & Berman, 2006) or reduced amygdala activation to threatening faces (Meyer-Lindenberg et al., 2005) which may further explain why individuals with Williams Syndrome do not show "Stranger Danger".

Due to its rarity, there are fewer studies of brain imaging in Williams Syndrome compared to autism. This means that there are still issues about how development itself affects the neural development of someone who has Williams Syndrome. Two studies examining ageing in Williams Syndrome with respect to brain volume report that despite the overall reduction, the relative ageing curve of brain volume was the same as typically developing controls (Cherniske et al., 2004; Koran et al., 2014). However, these were adults with Williams Syndrome and as such whether the brain volume growth curves are the same as typically developing children is not known. Likewise, atypical connectivity in individuals with WS is not surprising given the differences in brain size and structure in Williams Syndrome.

Overall the underlying neural structure in Williams Syndrome reflects many of the cognitive and behavioural characteristics found in the condition, with specific anatomical differences reflecting the poor visuospatial difficulties and facial perception. Abnormal functional connectivity and anatomical structure in the ventral stream and primary visual areas may impact on the ability to process visual information and on the ability to process colour information. Atypical functional connectivity of the ventral stream may affect cognitive uses of colour after initial sensory processing.

## 1.6. Comparison of Sensory Sensitivities between Autism and Williams Syndrome

As outlined above, despite the recent addition of abnormal sensory sensitivities or interests to diagnostic criteria for autism in DSM-V, little is known about sensory processing in autism or whether sensory processing in autism is similar or different in relation to other developmental conditions

(Rogers & Ozonoff, 2005). In this thesis, the comparison of how autism and Williams Syndrome vary (if at all) relative to typically developing controls will enable clarification of issues relating to condition specificity of colour perception between autism and Williams Syndrome. With reference to colour perception, previous research has found that chromatic discrimination is impaired relative to chronological and mental age controls in autism (Franklin, Sowden, et al., 2010), but in Williams Syndrome chromatic discrimination has been found to be impaired relative to chronological age only (Farran et al., 2013), although for all previous studies certain caveats apply, given the restricted nature of the populations studied and elements of the methodology. There is some suggestion that in autism, the ability to use colour information may be poorer relative to controls but it is unclear whether this is the case in Williams Syndrome.

## 1.7. Why Study Colour Perception in Autism and Williams Syndrome

There have been wide ranging uses of colour in investigations of Autism and Williams Syndrome, including both basic science research (both as a topic and in other studies such as visual search) and intervention. Both people with autism and Williams Syndrome have been found to have atypical ventral stream function in other visual functions, e.g. face processing. Since the ventral stream underlies key aspects of social cognition (e.g. face processing) it may be that this dysfunction is caused by differences in processing of lower level visual factors in the ventral stream. Additional understanding of lower level ventral stream function may help to understand later downstream higher order ventral stream visual processing.

Colour has also been used in clinical guidelines to mixed effect. The National Institute for Health and Care Excellence (NICE) guidelines for adults with autism states that the colour use in the room should be monitored, specifically recommending “low-arousal colours such as cream” ((NICE, 2012) p12). However, there is no research to support this statement, let alone the use of a single specific colour for individuals with Autism. There is also no mention of colour in the NICE guidelines for children or young people with autism (NICE, 2011). Udwin, Yule and Howlin (1991) warn against the use of *too* much colour in books and programmes for children with Williams Syndrome as this may cause visual overstimulation and as such distract the child from the classroom activity (e.g. reading), although colour coding may be useful to improve independence and to teach new skills (Udwin, Yule, & Howlin). One experimental study utilises colour in this way in Williams Syndrome. Farran et al. (2012) found that verbalising a colour aided the learning of a virtual route for both WS and TD groups, but the colour itself did matter. Focal colours (prototypical representations of colours across languages) on route were more accurately remembered than non-focal colours, but there was no significant interaction between group and focal/non-focal colours (Farran et al., 2012).

A second line of research that heavily involves colour is the use of coloured overlays and colour tinted glasses. These interventions require the participant to wear colour tinted glasses or use a coloured overlay; the results suggest that these interventions can help to reduce the visual stress that the individual experiences (see section 1.4.1.1 for in depth discussion of studies). Yet it is also unclear what lasting benefit the overlays have, coupled with the lack of experimental data in other domains outside of reading performance and anecdotal case studies. This demonstrates that despite the apparently convincing results on the surface, the use of coloured overlays is still not effective for all children with autism to improve their functioning. Sensory rooms and toys have also been suggested to induce "calmness" in the children. These sensory-based interventions typically involve the use of colour either through user-controlled illuminations or coloured surfaces. There is modest evidence for sensory interventions in reducing hyperactive behaviour but little on whether this effect continues outside of the sensory room.

These studies and interventions involve a pivotal use of colour that is central to the outcome of the study, but without sufficient scientific basis, given that knowledge of colour perception (at both sensory and cognitive levels) in both autism and Williams syndrome is very sparse. For example, will a prescribed colour for relaxing an individual work if that colour is disliked? The outcome of the studies in this thesis will enable greater efficacy in the use of colour in these interventions.

## 1.8. Aims of Thesis

The aim of this body of research is to give a more comprehensive characterisation of colour perception in both autism and Williams syndrome. Firstly, this study will include assessment of low-level perceptual discrimination of colour and the appropriate methods to use with typically developing children and children with either autism or Williams Syndrome (chapter 3). Previous research has either used inappropriate or incomplete methods (for example, not measuring all cone opponent axes for each participant). The second aspect of this thesis entails higher-level responses to and uses of colour (chapters 4 and 5). These investigations will include colour preference and colour naming. Finally, possible links between the lower-level sensory processing of colour and how (if at all) this relates to the higher-level uses of colour will be explored (chapter number). Unlike previous studies on colour perception, almost all participants will have completed all experimental tasks, making it possible to establish associations between different levels of colour processing within the same individual. Establishing whether there is a link between low-level sensory processes and higher-order responses including cognitive functions such as naming may give further indication of the extent to which atypical sensory processing (for example, "hyper" and "hypo" sensitivities) observed in both autism and Williams syndrome is related to behaviour. When previous studies have attempted to investigate this link, they have relied purely on questionnaire responses, whereas the work in this thesis instead uses direct measurements of both sensory processing and higher-level

functions. Colour is an ideal visual domain in which to make these links, because it is well characterised at a sensory level (chromatic discrimination) but also at higher levels (e.g. colour preference, naming and memory). It will therefore be possible to establish any links (if any) between sensory processing and affective/ cognitive/behavioural performance in autism and Williams Syndrome relative to typically developing controls.

# Chapter 2 – General Methods

## 2.1 Overview

This chapter will give an outline of how colour perception will be examined in autism and Williams Syndrome (WS) throughout the thesis. The chapter will provide a discussion of the relevant background and methodological issues relating to the central tenets of this thesis: developmental conditions and colour vision. The chapter will include participant recruitment, inclusion criteria and characterisation of the autism, Williams syndrome and Typically Developing (TD) control groups, as well as how comparisons of task performance are made between clinical groups and TD controls (e.g. matched-groups design). Finally, the general underlying principles of psychophysical tests and the stimuli that will be used in the experiments throughout the thesis will also be considered.

## 2.2 Characterisation of Sample

Accurate diagnosis and characterisation of participants is crucial to research involving developmental conditions. For the autism group recruited through the North East regional autism database and local North East England special schools, a diagnosis had already been conducted by trained multi-disciplinary team (including but not limited to; clinical and educational psychologists, Paediatricians, General Practitioner) according to DSM-IV and DSM-IV-TR criteria and ICD-10 definitions of autism (American Psychiatric Association, 1994, 2000; World Health Organization, 2008). To confirm the autism diagnosis, the Social Communication Questionnaire (SCQ, formerly Autism Screening Questionnaire; (Rutter, Bailey, & Lord, 2003)) was used. The SCQ is a short 40 item parental questionnaire which is used for screening individuals from two years old for autism (Rutter, Bailey, et al., 2003). Each item scores either 0 or 1, where scores of 1 indicate the presence of an "autistic behaviour". The questions have been adapted from the Autism Diagnostic Interview Revised (ADI-R (Lord, Rutter, & Le Couteur, 1994; Rutter, Lord, & Le Couteur, 2003)) to identify behaviours that are characteristic of individuals with autism. Sub-scores can be calculated for the three central aspects of autism, and parallel those of the ADI-R, Reciprocal Social Communication, Restricted, Repetitive and Stereotyped Patterns of Behaviour and Qualitative Abnormalities in Communication. In DSM-V, atypical sensory sensitivities and interests for autism has been posited to be a part of the Restricted, Repetitive and Stereotyped Patterns of Behaviour, SCQ scores are of these sub-domains and are of interest to appropriate sub-domain for atypical sensory processing. The SCQ has been shown to have good discrimination validation between individuals with and without autism (Berument, Rutter, Lord, Pickles, & Bailey, 1999; Chandler et al., 2007; Charman et al., 2007) and is also independent of age, IQ or language ability (Bishop & Norbury, 2002; Rutter, Bailey, et al., 2003). There is also good concurrent validity with the ADI-R at the domain level, particularly high correlations for Reciprocal Social Interaction and Communication domains and medium correlations

for the Restricted, Repetitive and Stereotyped Patterns of Behaviour domains (Bishop & Norbury, 2002; Rutter, Bailey, et al., 2003). There is also good internal consistency of this measure (from 0.84 increasing to 0.93 from 2-4 years to 11+ years respectively) with similar standard deviations across different age groups. In comparison to similar alternative measures such as the Social Responsiveness Scale (SRS) (Constantino & Gruber, 2002) and Children's Communication Checklist (CCC) (Bishop, 1998), the SCQ has been shown to have good sensitivity (0.86) and specificity (0.78) when compared to the SRS (sensitivity = 0.78, specificity = 0.67) and the CCC (sensitivity = 0.93, specificity = 0.46). The SCQ clearly outperforms the SRS on sensitivity and specificity and although the CCC has higher sensitivity than the SCQ this is offset by its considerably lower specificity (Charman et al., 2007). This suggests the SCQ is a reliable method to confirm autism of diagnosis, independent from other developmental conditions.

Participants with WS were recruited from the Williams Syndrome Foundation (WSF), UK, thereby guaranteeing that participants have a diagnosis of Williams syndrome. Prior to widespread availability of genetic testing, a diagnosis of Williams syndrome was made on the phenotypic expression of the disorder (Preus, 1984); however this was inadequate due to individual variability in the expression and thus there was not a single behavioural phenotypic feature that was consistently found in all individuals with WS (Morris, 1999). Recent advances in genetic testing combined with the key physiological and cognitive characteristics of WS, have enabled genetic markers for WS to be identified. The fluorescent in situ hybridization (FISH) is used to detect the micro deletion of the elastin gene (one of the approximately 25 sequential genes missing in the region of 7q11.23 which is known to be associated with WS). The FISH technique on this region allows for the 98% identification of WS (Ewart et al., 1993; Frangiskakis et al., 1996; Morris, 1999). All participants in this study had a previously had their diagnosis confirmed with a positive FISH diagnosis for WS.

TD individuals were screened using the Strengths and Difficulties Questionnaire (SDQ (Goodman, 1997)), was used to indicate the presence or absence of possible behavioural and emotional problems in the TD sample. The SDQ is a short 25 item parental/teacher questionnaire in which parents' rate behaviours on a three-point scale from 0-2. The questionnaire has a five-factor structure relating to behaviour: Emotional Symptoms, Conduct Problems, Hyperactivity/Inattention, Peer Problems and Prosocial Behaviour. The SDQ has been used successfully in screening for hyperactivity and psychiatric conditions, such as depression or anxiety (Goodman, Ford, Simmons, Gatward, & Meltzer, 2000). There is good specificity and sensitivity to detection of behavioural problems in TD children, where the area under the receiver operating characteristic curve measured at 0.87 (Goodman, 1997), although other studies have found this to be higher (Goodman et al.,

2000). In addition, there is also good agreement between parent and teacher completed SDQ's. In comparison to other measures such as the Rutter Parent Questionnaire (Rutter, 1967), the SDQ was found to have similar levels of sensitivity to identifying children with behavioural problems. There is also good agreement with psychiatric diagnoses based on ICD-10 criteria (Goodman et al., 2000). Furthermore the SDQ has also been shown to have higher overall scores in individuals with autism compared to children without autism (Chandler et al., 2007), and to have clinical specificity between high-functioning autism and attention deficit hyperactivity disorder (Iizuka et al., 2010). Therefore, the SDQ is a good screening questionnaire for behavioural problems in TD children. For the purposes of this thesis, the SDQ is used to screen only for behavioural difficulties in the TD sample. Parents filled out and return the SDQ as part of a wider questionnaire pack (see section 2.8 General Protocol). A threshold cut-off score of 13 or more has been identified in UK general population studies as a likely indicator of a psychiatric condition, where scores above 13 indicate increased likelihood of having a psychiatric condition. This cut-off score of 13 has been recommended as the optimal score to balance between false positives and false negatives (Goodman, 1997; Kessler, Barker, Colpe, & et al., 2003). All the TD sample recruited here had SDQ scores below the cut-off score of 13 indicating no behavioural difficulties in the TD sample.

## 2.3 Participant Recruitment

This section will outline the participant recruitment protocols of participants in all three groups. As each participant group was recruited via different means, three different recruitment protocols were required and these will be outlined separately for each group. (See appendix NUMBER for sample recruitment letters for the three groups). Generally, participants were recruited through local schools or for the autism and Williams syndrome groups, recruitment was aided through charities or databases associated with each condition respectively.

Participants with autism were recruited through a combination of local special schools and through Daslne (the Database for children with Autism Spectrum disorder Living in the North East. This led to fourteen participants who were recruited through local special education schools, with a further seven from the Daslne database. Approximately 55% of children between the ages of 6-18 years old with autism living in the North East of England (Newcastle, North Tyneside, Gateshead, Northumberland, South Tyneside and Sunderland) are recruited to the database (McConachie et al., 2009; Warnell et al., 2015). The families in Daslne have been shown to be representative of families in the UK with a child who has autism (Warnell et al., 2015). Daslne has several purposes including with parents' consent aiding recruitment of participants for research studies (McConachie et al., 2009). For this thesis, information sheets about the study were sent to fifty parents. Of these, nine families expressed interest in taking part (18%), and seven gave informed consent. Two families did not take part: one was unable to be contacted, whilst the second family withdrew from the study

after cancelling two appointments. To recruit through special education schools, initial contact letters were sent out to three special education schools in the Newcastle, Tyne and Wear and Northumberland regions. Two out of three (66%) special education schools who were contacted agreed to take part in the study. Once a school had agreed to take part, parental and young person information sheets and consent forms were sent to potential participants. These were then returned to the school if the parent and child wanted to participate in the study. A total of forty sets of information sheets and consent forms were sent out. Of these seventeen (42.5%) consent forms were returned. Thus, a combined total of twenty-four participants were recruited in the autism group.

The rarity of WS meant that it was necessary to recruit from as wide a geographical area as possible, to ensure a sample greater than the minimum required size. For this thesis, participants were recruited from Scotland, North England, Yorkshire and Midlands regions through the Williams Syndrome Foundation, UK (WSF). When families sign up to the WSF, they are given the option to be contacted with the opportunity to take part in research. The WSF was asked to provide a list of members in these regions between the ages of 7-20 years old. The families of all potential participants on this database were contacted and asked to take part in the study. The WSF identified individuals within the age range of 6-20 years, were contacted within Scotland, Northern, Midlands England and Welsh Regions. A total of 34 letters were sent out with a response rate of 79.4%, thus a total number of twenty-seven participants with Williams Syndrome were recruited in this way. One participant did not take part due to an inability to establish further contact. One additional WS participant was not recruited through the WSF but was recruited through a local special education school.

For the TD group, schools were contacted in the same way outlined above for special education schools. A total of 12 schools from Newcastle and Northumberland area were contacted, of which three agreed to take part (25%). From these schools, 79 individuals returned consent forms (return rate 49%). This is the total number of TD participants recruited, from this, a selection of appropriate mental age matches was chosen (see section 2.4.1 for more details)

### 2.3.1 Sample Size

The sample size of experiment participants affects the statistical analysis that can be conducted on the data and the conclusions that can be drawn (Lenth, 2001). Preliminary power calculations, made using Gpower (v3) software (Faul, Erdfelder, Lang, & Buchner, 2007), were undertaken to guide the choice of sample size for this study. These calculations assumed a small effect size (0.2), based on comparing two groups using an alpha value of 0.05. This alpha value was chosen as it represents the best trade-off between the risk of Type

1 (false positive) and Type 2 (false negative) errors. The minimum number of participants needed to detect within/between-subjects' interactions is 42 (21 per group). The resulting sample size for both groups (autism and WS) is although relatively small, equivalent to published studies of a similar nature investigating differences in visual processing in atypical development (Atkinson et al., 2003; Bertone et al., 2005; Farran et al., 2013; Franklin, Sowden, et al., 2010; Pellicano et al., 2005).

### 2.3.2 Inclusion and exclusion criteria

For each participant group, specific inclusion and exclusion criteria were applied. Inclusion criteria for the autism and WS groups were a clinical diagnosis of the condition and an appropriate chronological age (between 6 and 20 years old). Exclusion criteria for the autism and WS groups included the presence of a comorbid condition (e.g. ADHD), which was confirmed by either the school or parent. Whilst TD participants were excluded if they were diagnosed with a complex or specific neurodevelopmental condition such specific language impairment or dyslexia, again confirmed by either the school or parent. The other main exclusion criterion for all participant groups was the lack of normal trichromatic colour vision (colour-blindness'). Screening for 'colour blindness' was carried out with two standardised tests (see section 2.8 for the testing protocol). Participants who scored in the anomalous colour vision range on these tests were excluded (see section 2.7 Screening for Anomalous Colour Vision for more details).

Of the final recruitment sample of 26 individuals with autism and 27 individuals with WS (with no comorbid conditions), all verified as having normal trichromatic colour vision, a total of 2 participants were removed from the autism group for having a comorbid diagnosis of ADHD. The remaining individuals were each individually matched to a TD participant using the Raven's Colour Progressed Matrices (RCPM). Some TD participants being included in the matching sample for both groups. Descriptive information for the total sample of participants is given in Table 2-1. From this total sample, smaller sub-samples are constructed for each experiment described in further chapters, depending on the completion of the relevant tasks and on the matching criteria. Specific details regarding participant information for each group is outlined in each relevant chapter.

### 2.3.3 Final Sample Criteria

In this thesis both the autism and WS groups will be described in terms of their performance on each task relative to a matched TD control group. Due to the large disparity in both verbal and non-verbal ability within and between autism and WS samples, it was not possible to directly compare these two clinical groups, as the samples did not have equivalent levels of verbal or non-verbal ability at the same chronological age. Instead they will be described in terms of how they perform on the specific task relative to their verbal or non-verbal ability matched TD control group (irrespective of chronological age). Comparison of both autism and WS groups with appropriately matched TD

control groups will enable the investigation of the performance of the clinical sample relative to a sample of individuals matched for verbal or non-verbal mental age. In addition, a developmental trajectory approach (see section 2.4.2) will also be used to answer questions related to deviancy/delay relative to mental age. The inclusion of both the individual case by case matched sample design and developmental trajectory approaches will enable the investigation of colour perception in both clinical groups (autism and WS) relative to TD children, and address issues of deviancy/delay in either clinical group.

To match both autism and WS groups to the TD control group the RCPM will be used (see section 2.4.1). Each participant in the autism and WS groups will be individually matched to a TD participant. A matching criterion of plus/minus two in raw score between two participants will be used. This thesis will also report p-value, effect size and variance ratio when assessing appropriateness of matches, see section 2.4.1 for wider discussion. In addition, matches will also be made on sex, primarily because of the previously documented sex differences in colour preference (Child, Hansen, & Hornbeck, 1968b; Hurlbert & Ling, 2007). For developmental trajectories, all procedures outlined by (Thomas et al., 2009) will be used, whereby regression slopes for task performance against mental age are assessed between groups for each task in this thesis. The use of both the matched group design and developmental trajectories approaches will enable a more comprehensive assessment of colour perception in autism and WS and how (if at all) these differ from TD children.

*Table 2-1 – Participant demographics for all participants that were recruited and passed the inclusion/exclusion criteria.*

| Group | Age in Years (S.D.) | Male | Female | RCPM* (Raw) | Verbal IQ** (Standard Scores) | Performance IQ ***(Standard Scores) | SCQ+ | SDQ++ |
|---|---|---|---|---|---|---|---|---|
| TD n = 77 | 6.47 (1.49) | 30 | 47 | 21.3 (6.58) | 111.93 (13.78) | 104.63 (14.7) | N/A | 3.64 (5.19) |
| WS n = 27 | 12.81 (3.21) | 13 | 14 | 15.89 (4.78) | 69.96 (13.8) | 54.54 (8.94) | N/A | N/A |
| Autism n = 23 | 12.91 (2.3) | 17 | 6 | 26.91 (6.59) | 75.55 (21.13) | 85.05 (18.97) | 26.83 (8.3) | N/A |

* RCPM =Raven's Colour Progressive Matrices (Raven, Raven, & Court, 1962), ** Verbal IQ as measured by the Wechler's Intelligence Scale for Children (WISC, versions 3 and 4) (Wechsler, 1991, 2003), *** Performance IQ as measured by the WISC (versions 3 and 4) (Wechsler, 1991, 2003). + Social Communication Questionnaire (Rutter, Bailey, et al., 2003), ++ Strengths and Difficulties Questionnaire (Goodman, 1997).

## 2.4 Developmental Methods

This section will outline the specific issues that arise when conducting research with developmental condition populations and the types of strategies available to establish appropriate comparisons of how behaviours of individuals with differing developmental conditions can be considered both between conditions and with non-affected individuals. In this thesis, it will enable the typicality of colour perception in autism and WS to be addressed (see next section). The following two sections will look at two of the most common methodologies to compare developmental conditions, matched groups design and developmental trajectories.

### 2.4.1 Matched Groups Design

One of the most common ways to study clinical conditions is using a matched design (Mervis & Klein-Tasman, 2004). In a matched groups design, participants are matched on a selected measure (e.g. mental or chronological age; sex), thereby helping to control for any variability between the individual subjects in the clinical and non-clinical groups that is not related to the experimental measure under study. If there is no significant difference between the two groups on the matching measure, then the groups are considered to be matched. The matching method is a powerful way to assess whether there is a real difference between groups solely in the experimental measure under consideration, while controlling for identified potentially relevant variables. There are two main

methods to use the matched groups design. The first method is to match each individual participant in a clinical group to a single TD participant. The second method is where participants are matched at a group level (i.e. the means of the matching measure do not differ) but that each participant is not individually matched. The subsequent paragraphs will outline the rationale behind the matched groups design.

Traditionally in a matched groups design, depending on the research question, the group with a clinical condition (such as autism or WS) might be compared to more than one TD control groups. For example, it may be more appropriate to select one of the control group to match on chronological age (CA), and choose a second control group matched on mental age (MA) (or both where possible). When there is poorer task performance in the clinical condition than both the CA and MA groups, the clinical condition is considered to have atypical function on the task. However when task performance is poorer than the CA group only, but not the MA group, then the difference is interpreted as a developmental delay or deviance in the clinical group (Hodapp, Burack, & Zigler, 1995; Leonard, 2014).

To assess whether two groups are matched, on the selected matching measure (e.g. CA or MA), an independent samples t-test is conducted. If the test is not significant then this implies that the groups are matched. Importantly, though, there is no universally accepted standard for either the criteria for a match or what constitutes a "good match" (Brock, 2013), or that the two groups are matched at the group level. In particular, it has recently been suggested that the reliance on p-values alone may not be enough in deciding whether two groups are matched (Kover & Atwood, 2013). For example, non-significant result does not necessarily mean that there is not a difference between the groups on the matching measure. This is because the independent samples t-test tests for a difference for a significant difference between the means of a group, it will not take into the underlying distribution between of the two groups on the matching measure. Therefore, it is important also to consider variation in performance on the matching measure (e.g., on the IQ test used to assess mental age). Kover and Atwood (2013) argue that using the p-value alone to interpret whether a group is matched does not account for the sample size and variation. For example, it is possible for two groups not to differ significantly (via the t-test) in their performance on the matching measure with a small sample of participants, but for the two groups to become significantly different from one another when the number of participants is increased, with the same group mean and distribution on the matching measure. This is because small samples are more likely to generate type 2 errors (false negatives) due to the reduced sampling size (see (Brock, 2013; Kover & Atwood, 2013) for examples) and increased distribution of the matching measure (e.g. age or ability). Given that a well-controlled comparison group is a central tenet of research with clinical groups, it is important to consider this potential confound when matching between groups. Not doing so may lead to an inappropriate

comparison between clinical and control groups. In turn, this confusion may lead to increased risk of both type 1 and type 2 errors when interpreting experimental task performance between clinical and control groups if the two groups are not equivalent on the matching measure. Kover and Attwood (2013) suggest that it is important, therefore, also to report effect size (e.g. Cohen's D) and variance ratio between groups on the matching measure test, since these variables are not influenced by sample size. Reporting of the effect size, such as Cohen's D will therefore give additional clarity on the level of similarity of the matching measure between the two groups. However, there is also no 'accepted' threshold for either variance ratio or effect size, other than that both should be as low as possible (i.e. little or no difference); Kover and Attwood (2013) explicitly propose, for example, that groups should be considered adequately matched only when both the effect size and the variance ratio fall within expected standards, relative to the values expected for the experimental measure under study. Individually matching between groups rather than matching at the group level will further reduce the risk of a type 2 error when accepting whether the groups are matched, since the variance ration and effect size will be small (Kover & Atwood, 2013).

The choice of the matching measure has also been shown to affect the outcome of a study. Barbeau and colleagues (2013) found that there was a significant difference in inspection time between an autism and typical control group when matched on full-scale IQ WISC scores as a measure of cognitive ability, but this significant difference disappeared when the Standard Progressive Matrices (SPM) (Raven, Raven, & De Lemos, 1958) was used instead of the WISC as a matching measure for cognitive ability (Barbeau, Soulières, Dawson, Zeffiro, & Mottron, 2013). This finding highlights the importance of the matching measure, whereby there can be different overall results between the clinical condition and control groups when different IQ tests are used to match the groups, even though the participants in the clinical condition group have remained the same. Thus the appropriate choice of both the matching criterion, and the measure chosen to investigate the chosen criterion, are both crucial, as an inappropriate selection may cause either type 1 or type 2 errors.

The selection of an appropriate matching measure is crucial to the matched design approach. It is especially important for investigations of performance by individuals with autism and WS where there is both a wide spectrum of ability and often an uneven cognitive profile (Bellugi, Lichtenberger, Jones, Lai, & St George, 2000; Dawson, Soulières, Gernsbacher, & Mottron, 2007; Mottron, 2004; Soulières, Dawson, Gernsbacher, & Mottron, 2011). A wide variety of different tests have previously been used in the literature to match overall ability and/or different aspects of cognitive ability for individuals with autism or WS groups to TD controls (M. A. Martens et al., 2008; Mottron, 2004). The choice of matching measure is also dependent on the research topic of the study. One measure that has been selected to provide an assessment of overall ability and performance skills used is the

Raven's Colour Progressive Matrices (RCPM) (Raven et al., 1962) for younger children and Raven's Standard Progressive Matrices (SPM) (Raven et al., 1958) for adult participants. In both the RCPM and SPM the participant is required to identify a missing segment that completes a pattern. Each possible answer reflects how the individual is solving the pattern. In addition to the correct answer, there are errors which can be coded into four categories: 1) Difference (no relation to pattern); 2) Inadequate Individuations (failure to combine local features into global construct); 3) Repetition of Pattern (copy of existing pattern); and 4) Incomplete Correlation (partially correct identification of correct pattern). The RCPM and SPM are predominantly measures of non-verbal ability but also map onto problem solving and fluid intelligence because they measure abstract reasoning (Carpenter, Just, & Shell, 1990; Dawson et al., 2007).

The British Picture Vocabulary Scale (BPVS) (Dunn & Dunn, 2009) has also been frequently used to give a measure of receptive vocabulary in children from age two to sixteen years. The test itself requires the participant to identify the correct picture from four possibilities in response to a word spoken by the experimenter. This allows for the BPVS to be used with non-verbal participants to gain an estimate of their vocabulary, although this may also over estimate their verbal ability (Glenn & Cunningham, 2005). Another more comprehensive measure of cognitive functions that has been used commonly for matching is the Wechsler Intelligence Scales for Children (WISC III/IV) (Wechsler, 1991, 2003). The WISC uses a variety of subtests which are combined to give a composite score for verbal, performance (non-verbal) and general abilities. Verbal subtests include Vocabulary (defining words) and Similarities (verbal semantic associations between words). Performance subtests include Block Design (pattern construction with coloured blocks), Picture Concepts (associations between pictures) and Matrix Reasoning (pattern completion similar in nature to RCPM). Different "short form" versions of the test exist, which are still informative for research purposes in giving an estimation of the child's ability (Sattler, 2008), so that completion of all of the different subtests is not required for an assessment of ability. However, in both autism and WS performance across the individual subtests is not uniform (Dawson et al., 2007; Jarrold & Brock, 2004; Soulières et al., 2011), meaning that composite scores may be skewed by either enhanced or reduced performance on one subscale, resulting in a measure that is not an accurate indication of the child's ability.

To avoid the problem of peaks and troughs on test performance, it has been suggested to use a more representative measure to match participants (Farran & Jarrold, 2003; M. A. Martens et al., 2008). This proposal, though, carries with it additional problems for matching between groups. The uneven cognitive profile found in both autism (Hobson,

1992; Ozonoff, Pennington, & Rogers, 1991) and WS (Bellugi et al., 2000; M. A. Martens et al., 2008), mentioned previously, means that individuals with autism are more likely to show better non-verbal ability relative to verbal ability, and vice versa for WS. Thus, it might be difficult to have the same participant from the TD control group matched in both verbal and non-verbal ability to individuals in either group due to the variation across different cognitive domains. Therefore, an *a priori* decision, based on theoretical reasoning as to the contributions of the different types of ability to the experimental measure, need to be made and thus guide the decision about the most relevant aspect of ability to be controlled for by the matching measure. This in turn should inform the choice of the most appropriate matching measure, for example either verbal or non-verbal ability. Consequently, the matching procedure cannot control for all aspects of mental ability. For instance, when the control group is matched on mental age to the clinical group, it is likely, that there will be a difference in chronological age between both the groups. In turn this discrepancy in CA may result in different task performance strategies, i.e. that there are chronological age-related compensation strategies that may have developed in the clinical group. Another problem is that the matched design is developmentally static as it does not assess changes over time or account for differences in the rate of development in skills. For example, there may be faster development of verbal ability compared to non-verbal ability in WS (Jarrold, Baddeley, & Hewes, 1998).

Given the above considerations, this thesis will use RCPM performance as the primary measure of general ability to match both autism and WS participants to appropriate TD participants. The RCPM was chosen because most of the tasks have large non-verbal components. RCPM has also been used previously to compare performance of autism and WS relative to TD on sensory tasks (Farran et al., 2013; M. Martens, S. J. Wilson, & D. C. Reutens, 2008; Mottron, 2004). Because different tasks in the thesis relate to different aspects of colour perception, which in turn map onto different types of general ability, the WISC is also used to give an overall measure of the verbal and non-verbal abilities of the participants, and therefore to enable matching via verbal or non-verbal ability. For example, the assessment of chromatic discrimination (low level sensory) and colour preference (affective response to colour) best map onto non-verbal abilities, for which a non-verbal matching measure should ideally be used, whereas colour naming (assessment of colour cognition, and particularly colour lexical categorisation) is a predominantly verbal task, and therefore a verbal ability match could be used. However, by their nature, there is still non-verbal aspects to these tasks. Therefore, since RCPM is a good estimate of general ability it will also be used in these chapters. The thesis uses both the four- and five-subtest short form versions of the WISC-IV: for verbal ability, the subtests of Vocabulary and Similarities are used. Performance ability is estimated using Block Design and Picture Concepts. Lastly, Matrix Reasoning is used as an additional subtest, in the five-subtest short form. The WISC was chosen over the BPVS as it is less likely to overestimate verbal ability.

It should be highlighted however that even if there is no difference in performance on the experimental measure between the control and clinical group, there might still be a difference in the underlying mechanisms, i.e. the same performance level may be reached via a different method. Also the matched groups design does not explain whether there is deviancy or delay in task performance, only whether there is a difference between groups at that time point (Leonard, 2014). Therefore, it is crucial to understand the developmental context of abilities in both typically and atypically individuals. The following section on developmental trajectories methodology will outline how this analysis method can be used to overcome this limitation of the matched groups design.

## 2.5 The Psychophysics Method

The next section will outline what the psychophysics method is and why psychophysics was used to study colour perception in this thesis. Psychophysics is the quantitative study of perception. It aims to define an objective relationship between physical phenomena and the individual's sensation of those physical phenomena, i.e. to quantify subjective perceptual experience. This method was pioneered by Gustav Fechner, who argued that the brain and body both inhabit the same world and studying the links between the two can reveal the mechanisms between mental sensation and the material world (Fechner, 1832). Since then the psychophysical method has been widely adopted by researchers wishing to study perception. To date the predominant method used to study sensory processing in autism and WS has been questionnaires, usually completed by the parent (see Chapter 1 sections 1.1.1 and 1.2.1, and Chapter 6 for wider discussions). However, these rely upon the secondary judgements made by an informant about observable sensory behaviours of the subject and may not accurately represent the sensory processing of the subject themselves. Some previous studies have used psychophysical methods to study sensory processing in autism and WS across different modalities, e.g. (Atkinson et al., 1997; Bertone & Faubert, 2006; Bonnel, Mottron, Peretz, Trudel, & Gallun, 2003; Franklin, Sowden, et al., 2010; Reiss et al., 2005a), for further discussion see Chapter One. This section will give an overview of some of the most common methods used in psychophysics.

There are multiple methods employed in psychophysics to quantify subjective perception as a function of objective changes in stimulus strength or other properties (Farell & Pelli, 1999; Gescheider, 2013; Pelli & Farell, 1995). Two classic methods are: 1) The adjustment method, in which the participant is required to change stimuli to meet a criterion set by the researcher (e.g. adjusting the colour of a patch to appear grey); and 2) The judgement or choice method, in which the participant decides about the presented stimuli (e.g. are these stimuli the same or different). In

judgement tasks the stimuli can be combined with other methods such as staircase procedure to systematically assess sensory sensitivities. A threshold is defined as the minimal perceptual quantity or just noticeable difference (JND). The participant's response can be plotted against stimulus intensity to create a psychometric function which can then be used to assess performance. When applying either the adjustment or choice methods for use with children or clinical conditions the same principles still apply as outlined above. The low cognitive demands mean that psychophysical tests can used for young children and clinical groups whose ability is lower. Furthermore, the choice method used to establish thresholds means that difficulty is equated regardless of chronological or mental age or the presence of a developmental condition, i.e. judgements about whether you can see or hear a stimulus is the same across participants. To make psychophysical tasks more appropriate they need to be framed in a child appropriate way, this also helps make the task become more enjoyable for the child. For example, this may be following arrows to find a treasure or using fish to describe motion coherence study.

This thesis will use psychophysics methods in two chapters; the chromatic contrast discrimination (see Chapter 3) and the computerised colour preference (see Chapter 4) tasks which employ different psychophysical methods. The chromatic contrast discrimination uses an adaptive staircase combined with a forced choice judgement task to identify chromatic discrimination thresholds. The colour preference task uses a combination of forced choice method with method of constant stimuli to establish different patterns of colour preference. The next section considers these methods in more detail and why such methods were chosen for the current research.

### 2.5.1 Forced Choice Method

The forced-choice method requires the participant to make a judgement about the property of a viewed stimuli and select a response from a fixed number of alternative options (e.g. identifying the direction of an arrow as pointing to either left, right, up or down) (Treutwein, 1995; Wichmann & Hill, 2001). By making the participant decide about the presented stimuli, the risk of bias in accuracy of their response is reduced in the forced choice method, although a participant could still give a biased response, the risk of a bias in accuracy is reduced because the correct choice is counterbalanced among the possible options. This enables performance to be assessed against chance. Another advantage is that the task demands are low, making this more applicable for use with children or individuals with a developmental condition. The forced choice method used in this thesis for both the chromatic contrast discrimination threshold experiment (Chapter 3 Experiment 2) and the colour preference tasks (Chapter 4). In the chromatic discrimination experiment a single arrow stimulus will be presented and then participants will be forced to choose between two possible options but once

the initial stimulus is no longer present on the screen, i.e. a blank screen whilst the participant is asked to decide whether the target arrow was pointing to the right or left. In the colour preference experiment the forced choice method is combined with the method of constant stimuli. Two coloured patches will be presented, one above the other. Since the goal of this experiment is for participants to make a judgement about the two presented stimuli.

### 2.5.2 Staircase Method

One method used to establish a threshold is the staircase method (Cornsweet, 1962). The staircase method is an adaptive method because the current stimulus on a trial is determined by the participant's preceding responses. In this method, the participant is presented with a series of stimuli that either increase or decrease in their signal strength along a given dimension. When the participant's responses change (e.g. from correct to incorrect, or yes to no) the direction of stimulus change reverses (e.g. from increasing strength to decreasing strength). This means that a reversal indicates where a participant can make reliable judgements on the presented stimuli. This procedure continues until enough reversals have occurred for a threshold estimate to be made. There are no fixed rules on how many reversals constitute the "best" threshold (García-Pérez, 1998; Klein, 2001). An increased number of reversals reduces bias in the results but this is not necessarily optimal for use with children or developmental conditions. Virtual simulations of staircase data has shown that the number of reversals did not affect the convergence point (García-Pérez, 2001), but also that number of optimal reversals may vary depending on the number of trials in the staircase, and the step-size (García-Pérez, 1998).The step-size between adjacent stimuli in the staircase may be set prior to beginning the experiment. The commonplace tactic is for these step sizes to change throughout the experiment, going from a large step size and then gradually reducing in size as the participant nears their threshold (Levitt, 1971). Different methods have been proposed for how best to determine these changes in step size (Macmillan & Creelman, 2004), using information from responses across of the entire run (i.e. the performance up to the current trial), as in ,e.g., the PEST method (Taylor & Creelman, 1967), or also including *a priori* information (e.g. both PEST and QUEST (Taylor & Creelman, 1967; Watson & Pelli, 1983)).

Likewise, there are multiple stopping rules for a staircase procedure, e.g. completion of a predetermined set of trials, occurrence of a fixed number of reversals or the attainment of a minimum step size or confidence interval. The threshold is then calculated from the responses data; again, there are multiple ways to do this calculation. These include using the final value of stimulus strength, or averaging stimulus strength over a certain number of reversals or across a certain number of trials. Different procedures use different stopping rules and adaptive processes. When considering the use of the staircase method with children and clinical conditions there needs to be consideration of the length of the experiment. This means a trade-off between the number of trials,

reversals needed to establish a reliable threshold and the length of time to complete the experiment. The adaptive staircase methods used in this thesis, for the chromatic discrimination measure, is a 1up-2down procedure, and are described in more detail in Chapter 3.

## 2.6 Colorimetric specification of colour stimuli

Colorimetry is the study of measuring colour; through assessment of light wavelengths and spectra and how this combines with the human visual system. This section will outline the basic procedures behind colorimetry, it is important to outline these procedures before moving onto explanations of cone-contrast and Hue Saturation Lightness colour space models that were used in this thesis. From the outset, it is important to state that this colour matching functions have all been identified using adult participants. It is possible that there may be developmental changes in the cone sensitivities from childhood to adulthood. The lack of colour matching functions from young children or adolescents reflect a limitation of the field and were beyond the scope of this thesis to rectify this.

The main aim of colorimetry is to provide or assess a quantitative description of colour appearance which enables a match to the colour to be obtained for the human visual system (Fairchild, 2013). It does this by accounting for all three major factors involved when perceiving colour: the light source, the reflective surface and the human visual system. Colorimetry allows for accurate display of colours on the display surface with respect to the physiology of the human visual system. Thus, colorimetry involves the measurement of the spectral power distribution of the reflected light and the calculation of the responses of human retinal receptors to this light. There is a need to have a system in place that allows for consistent notation of colour stimuli. In 1931, the *Commission Internationale de l'Eclairage* (CIE) specified a set of colour matching functions to define the appearance of a void colour in terms of three standardised descriptors. The CIE XYZ tristimulus values are derived from colour matching functions for a two-degree stimulus obtained from matching experiments on several sets of observers with normal trichromatic colour vision. The three equations (one for each tristimulus value) are denoted below where $E$ represents the spectral power radiance distribution, $\bar{x}, \bar{y}, \bar{z}$ are the three colour matching functions, which were derived from linear combinations of matching functions for monochromatic primaries. The tristimulus values is obtained by integrating the spectral power distribution of the light with the colour matching function, over wavelength. The colour matching functions are by necessity linear combinations of the three cone (L,M,S) spectral sensitivities.

$$X = \int E(\lambda)\, \bar{x}\,(\lambda) d\lambda$$

$$Y = \int E(\lambda) \bar{y}\,(\lambda) d\lambda$$

$$Z \quad = \int E(\lambda)\, \bar{z}\,(\lambda) d\lambda$$

The $\bar{y}$ colour matching function was derived to exactly match the luminous efficiency function $V(\lambda)$, and therefore $Y$ is the photometric luminance of the stimulus.

$$\bar{y}(\lambda) = V\,(\lambda)$$

To separate luminance from chromaticity in the tristimulus coordinate system, the CIE *xy (*1931) chromaticity coordinates are calculated by normalizing the tristimulus values by the sum of the tristimulus values. Since *z* can be explained by *x* and *y*, this allows for chromaticity to be described in a Cartesian coordinate system using just *x* and *y*.

$$x = \frac{\mathrm{X}}{\mathrm{X} + \mathrm{Y} + \mathrm{Z}}$$

$$y = \frac{Y}{X + Y + Z}$$

$$z = \frac{Z}{X + Y + Z} = \; 1 - x - y$$

A void colour may then be described by its luminance $Y$ and its chromaticity coordinates $(x, y)$. Whilst the CIE (1931) system is widely used as a standardised colorimetric space, although it is based on a relatively small number of observers, and is neither perceptually uniform nor directly phrased in physiological terms (for wider discussion see (Wyszecki & Stiles, 1982). The next section will give an example of how this can be done for a physiological cone-opponent space.

### 2.6.1 Physiological cone-contrast space

This thesis uses a version of a physiological cone contrast space as specified by Eskew et al. (1999, see Figure 2.1). A cone-contrast spaces is a 3-dimensional space in which each vector corresponds to the cardinal colour axes (red-green, blue-yellow and luminance) directions with respect to the reference surface's white point (i.e. a 3-dimensional space whereby the centre is the white point (Eskew, McLellan, Guilianini, Gegenfurtner, & Sharpe, 1999). Values are then generated using cone fundamentals to generate predicted values based upon weightings of cone fundamentals. The two chromatic axes are modulated within an isoluminant plane, thus controlling for any effects of luminance on the two chromatic axes. Specifically, modulation within a single axis, the colours of the central stimulus are systematically varied in their difference from the background (i.e. their "background contrast"), without changing their background contrast of along the other modulation

axes. For example modulation along the Red-Green axis alters the background contrast of the relative excitation between the L and M cones (L-M), while keeping the background contrast of the S cones constant. While modulation of the Blue-Yellow axis alters the background contrast of the S-cone activation relative M and L cone excitations (S-(L+M)). The formula of the relative weightings for each cone are listed below:

$$Luminance = (0.78 * \Delta L + 0.37 * \Delta M)$$

$$Red\ Green = (0.7 * \Delta L - 0.72 * \Delta M + 0.02 * \Delta S)$$

$$Blue\ Yellow = -(0.55 * \Delta L + 0.25\ \Delta M - 0.8 \Delta S)$$

Where the origin of this colour space is given by the cone excitations for a neutral background (i.e. grey colour) sample in the scene (L0, M0 and S0), ΔL = (L - L0)/L0, ΔM = (M - M0)/M0 and ΔS = (S - S0)/S0, where L, M, and S are the cone excitations of the specified point. LUM is the luminance axis, RG is the "red-green" axis and BY is the "blue-yellow" axis (modified from (Eskew et al., 1999)). In this space, a point may be defined using the cylindrical coordinates radius, azimuth and height (r, a, h), where the radius may be related to saturation changes, the azimuth, i.e. the clockwise angle formed with the RG axis, corresponds to hue, and the height corresponds to luminance. This cone contrast space is the colour space that was used in chromatic discrimination experiment in Chapter 4. The Eskew colour space was chosen because it enables chromatic discrimination to be assessed regarding specific cone-opponent mechanisms.

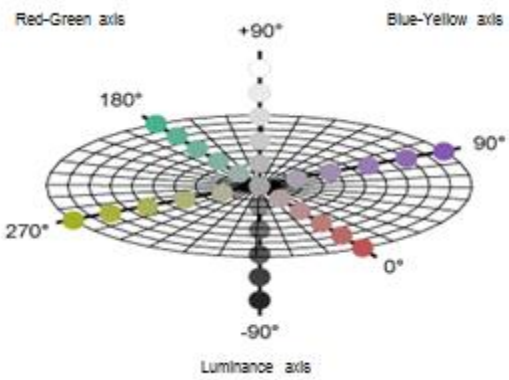



*Figure 2-1 - Example of a Cone Contrast space.* Figure taken from Stockman & Brainard (2009). Along the red-green axis, colours change with respect to the background colour (at origin) only in the difference between the L and M cone excitations. Along the yellow-blue axis, colours change with respect to the origin only in the difference between the S cone and sum of L an M cone excitations.

### 2.6.2 Hue Saturation Lightness (HSL) Colour Space

The computer graphics model of HSL (Joblove & Greenberg, 1978) is defined based on the primaries found within a monitor (i.e. Red, Green and Blue). To denote that hue (primary colour category) is defined by the angle around the central vertical axis (measured in radians). Saturation is the distance

(0-1 horizontal vector) from the centre point and lightness is length of the cylinder (0-1 vertical vector). The motivation for this colour space was to compensate for the limitations of using only RGB and reduce the computational speed when wanting to transform colours in a similar way that the human visual system does (i.e. making judgements about brightness or colourfulness). The HSL space that is used in this thesis follows a similar principle, however instead of using primaries from RGB, primaries are calculated from the perceptually uniform CIE L*U*V* colour space (see (Wyszecki & Stiles, 1982) for more details). The HSL colour space was chosen for us in the colour preference experiment (see Chapter 4) because it allows aspects relating to hue, lightness and saturation to be manipulated in a systematic way allowing for each aspect to be assessed (see figure 2.2).

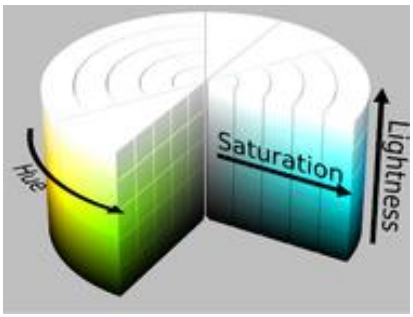



*Figure 2-2 - HSL colour space.* The hue changes depending on the angle, whilst lightness is represented along the y-axis and finally saturation variations across the horizontal vector from the centre point

## 2.7 Screening for Anomalous Colour Vision

Human colour vision is constrained by the spectral sensitivities of each distinct cone type (see Chapter 1 section 2.1 for more details). These peak sensitivities can vary between individuals the spectral sensitivities of the cone class can lead to slight changes in colour perception (Jordan & Mollon, 1995; Neitz & Jacobs, 1986; Schmidt, Neitz, & Neitz, 2014). This is most pronounced for individuals who are colour blind there are abnormalities in their colour vision physiology from changes of the genetic encoding of the cone opsins, i.e. the spectral sensitivities of the cones are shifted or that cones are absent (Neitz & Neitz, 2000). Different types of colour blindness exist depending on which cones are atypical. Achromatopsia (complete absence of colour vision) is rare. Whereas colour blindness due to abnormal sensitivity of one class of photopigment is more prevalent common (Birch, 2012), although this may vary with ethnicity. There are three types of colour blindness; Protanopia (absence of red cone), Deuteranopia (absence of green cone) and Tritanope (absence of blue cone). Milder versions of each Protanopia and Deuteranopia where the affected cone is not absent but its peak spectral sensitivity is shifted towards the intact functioning cone resulting in fewer colours being visible. Individuals with colour blindness see different colours due to deficiency in their colour vision, as such it is important to screen for such individuals before they take part in the study.

Individuals were screened for anomalous colour vision using two tests, the Ishihara Test (Ishihara, 1917) and the Neitz Color Vision Test (Neitz & Summerfelt, 2001). The role of the participant is to correctly identify either a number (Ishihara) or shape (Neitz Color Vision Test). In both tests, the target is a global structure composed from local differently coloured circles. Individual test items are isoluminant with respect to each other, to reduce luminance cues. The Ishihara test is used to identify red-green colour blindness, whilst the Neitz Color Vision Test can also identify tritanopes. The low task demands and uncomplicated stimuli in both tasks make them possible to be adapted for use with both young TD children and children with clinical conditions, e.g. (Birch & Platts, 1993). For example, if there is a participant who is not able to reliably name shapes or numbers then it is possible for the participant to trace the coloured outline of the shape.

## 2.8 General Test Protocol

Throughout data collection there was a general test protocol that was followed. This protocol varied depending on whether the participant took part at school, at the university or home visit. Where participants were tested in schools, they completed three to four short sessions lasting between twenty and thirty minutes. In each session participants completed a different experimental task, i.e. in one session participants completed either; the threshold task (Chapter 3 Experiment 2), colour preference task (Chapter 4), Colour naming (Chapter 5) and the Farnsworth-Munsell 100-Hue Test (Chapter 3 Experiment 1), and IQ assessments (Both WISC/WPPSI and RCPM). The order of sessions was counterbalanced across participants. For home visits, tasks were completed over two sessions. In one session participants completed the RCPM, chromatic threshold and colour preference tasks. The second session compromised of the Farnsworth-Munsell 100-Hue test (Chapter 3 Experiment 1), colour naming (Chapter 5) and WISC IQ assessment. When participants visited the unive,rsity all tasks were completed in one visit. Short breaks for participants were interspersed throughout all sessions. Parental questionnaires on visual and behavioural responses to colour were completed during or after the testing session (see Chapter 6). All TD participants took part at school, whilst parental questionnaires were completed at home. The majority of WS participants were tested at home. Finally, two thirds of the autism participants were assessed in school, whilst parental questionnaires were completed at home. The remaining third of the autism sample were either tested at home or at the university. For the chromatic discrimination and colour preference studies the same viewing box setup was used. This can be seen in figure 2-3.

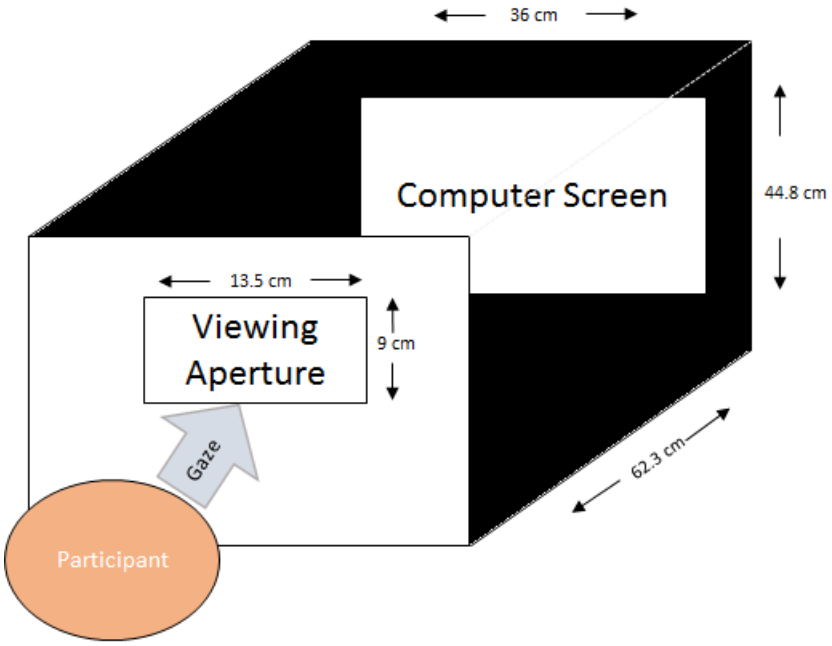



*Figure 2-3 – Illustration of Viewing Box.* The participant was sat in front of the viewing box and placed their head on a chin rest. The participant looked through a viewing aperture to a monitor positioned against the back of an enclosed box. The viewing box was also set up in a dimly lit room. When this was not possible and blanket was placed over the participant to reduce the external light going into the viewing box.

## 2.9 Conclusion

There are numerous methodological issues that need to be addressed when conducting research with developmental conditions. The methods used enabled the specific aims of this thesis to be addressed. The subsequent experimental chapters will apply the methods outlined in this chapter to study low-level perceptual (through psychophysical tasks) and cognitive processing of colour in autism and WS. Furthermore, using the same participants across all experimental tasks will enable relationships between different domains of colour perception to be appraised.

# Chapter 3 – Chromatic Discrimination

## 3.1 Overview

One central aim of this thesis is to examine sensory processing of colour in individuals with autism and Williams Syndrome (WS). This chapter will assess chromatic discrimination in autism and WS. The chapter will include the use of two assessment methods: the Farnsworth Munsell 100-Hue Test, a manual sorting task; and a new psychophysical test of chromatic discrimination designed for use in this thesis. The use of two different chromatic discrimination tasks to measure performance also provides an opportunity to assess the suitability of different tasks to measure visual processing in children with and without developmental conditions. This current chapter also investigates the importance of test selection for use with children and those with developmental conditions.

## 3.2 Introduction

### 3.2.1 Chromatic Discrimination Tests

There are numerous different standardised and customised tests that have been used to assess chromatic discrimination both clinically and by visual scientists, see (Dain, 2004; Paramei & Bimler, 2015) for a complete review of all chromatic discrimination tests. This section will review some of the relevant tests that have been used to study chromatic discrimination in typical development. The tests reviewed in this section do not include screening measures of colour blindness, e.g. nagel anomaloscope or Farnsworth Lantern Test, since these tests do not directly measure chromatic discrimination per se, but instead measure coarse cone activation (see Chapter 1 section 2.1.1 and Chapter 2 section 2.6). Instead the tests reviewed here aim to directly measure chromatic discrimination using standardised testing procedures and seek to develop norms of chromatic discrimination.

The Farnsworth-Munsell 100-Hue Test (Farnsworth, 1957) (FM100) has been widely used by clinicians and visual scientists as a measure of chromatic discrimination ability (Dain, 2004; Paramei & Bimler, 2015) in both typically developing individuals with normal colour vision (Kinnear & Sahraie, 2002) and adults with congenital or acquired colour vision deficits (Gunther, Neitz, & Neitz, 2006; Heywood et al., 1992; Ménage, Papakostopoulos, Dean Hart, Papakostopoulos, & Gogolitsyn, 1993; Victor, Maiesem, Shapley, Sidtis, & Gazzaniga, 1989). The FM100 test involves arranging a set of individual coloured caps of similar lightness and saturation in order between the hues of two fixed caps (e.g. blue and green), so that a smooth colour gradient is formed, with the hue differences between neighbouring caps as small as possible. The FM100 has been used with a wide range of ages, from early childhood (5 years) to elderly populations, and has the advantage that two research groups have attempted to establish age-expected norms (Kinnear & Sahraie, 2002; Knoblauch et al., 1987). It is also useful in identifying

congenital and acquired retinal diseases and as a measure of lens yellowing during normal aging (Beirne, McIlreavy, & Zlatkova, 2008). The FM100 has also been used with children with developmental conditions such as ADHD and Down's Syndrome (Banaschewski et al., 2006; Krinsky-McHale et al., 2014) as well as autism (Franklin, Sowden, et al., 2010) and WS (Farran et al., 2013). However, the FM100 is not without problems; in essence it is a seriation task. Seriation tasks are associated with developmental "milestones" and although present in younger children, seriation ability also increases in their competency with age (Birch, 2001; Dain & Ling, 2009; Karpf, Goss, & Small, 1974; Mareschal & Shultz, 1999). Furthermore, the FM100 has been shown to be significantly related to NVIQ in both typically developing children, adults and children with developmental conditions (Cranwell, Pearce, Loveridge, & Hurlbert, 2015; Hurlbert et al., 2011). This suggests that it may be difficult to dissociate the extent to which performance on the FM100 is due to chromatic discrimination and NVIQ. This finding has implication for the use of the FM100 norms given by Kinnear & Sahraie (2002). Firstly, their norms only account for FM100 performance related to chronological age and more specifically this is only for coarse categories across a whole year. Given the cognitive development within one chronological year during childhood further segmentation of age categories are needed. For example, it is not appropriate to treat an old five year old as categorically different from a young six year old. Another limitation of these norms is the low participant numbers (less than ten) for some ages. Given that there is a wide range of cognitive ability at any given chronological age and the absence of reported measures of ability of participants by Kinnear & Sahraie, it is unclear whether these norms are accurate for the given age. Nonetheless, even with their weaknesses, these are the only reported chronological age norms for the FM100.

One of the main aims of this thesis is to assess sensory processing of colour. However, given the variable IQ profile found in ASC (see Chapter 1 section 1.1) and lower IQ in WS (see Chapter 1 section 1.2), it is important to have a direct measure of chromatic discrimination that is not confounded by other factors such as NVIQ. The FM100 is also a relatively time-consuming test that requires attention and a degree of visuo-motor competence, and is subject to learning and practice effects (Schroeder, Kreutz, Meyer, & Erb, 2007). For these reasons it is important to consider whether other tests of colour discrimination might be more useful or appropriate for the children recruited in this study (Foote, Neitz, & Neitz, 2014).

Various computerised tests also exist, primarily used in basic science or clinical research rather than as standard diagnostic procedures. These are psychophysical tests which manipulate various colour signals in a systematic way (see Chapter 2 section 2.6 for more details). For example The Cambridge Colour Test (CCT) (Mollon & Reffin, 2000) is a test that uses pseudo-isochromatic displays similar to the Ishihara Test. However instead of identifying a number, participants are required to identify the open side of a stylised "Landolt C". The "C" is made up from various sized

circles, against a background of more circles. Each circle's luminance varies to control for different luminance noise effects across the stimulus presentation (Regan, Reffin, & Mollon, 1994a), and the circles' colours are controlled so that the "C" will be indistinguishable from the background to people with certain CVDs. In the CCT, participants make judgements about the direction open side of a Landolt C. The decision increases in difficulty via staircase procedure. Chromatic discrimination thresholds are calculated for confusion lines along protan, deutan and tritan axes as defined using the CIE (1976) *u'v'* colour space. Normative data exist for the CCT from 20 years of age upwards (Paramei, 2012; Paramei & Oakley, 2014). At present, there are no normative data for participants below the age of 20 years. The CCT has been successfully used as an assessment of chromatic discrimination in adults and is successful in identifying colour deficient individuals (Mollon & Reffin, 2000). It has also been used to assess chromatic discrimination in adults with other diseases such as multiple sclerosis (Moura et al., 2008), diabetes (Feitosa-Santana et al., 2010), Alzheimer's Disease (Regan, Freudenthaler, Kolle, Mollon, & Paulus, 1998) or toxic work environments (Klinger et al., 2001; Ventura et al., 2005). Goulart and colleagues (2008) adapted the CCT for use with children. In this adaptation the stimuli is changed to a static square located where the open end of the "Landolt C" would have been (Goulart et al., 2008). This version was trialled on twenty-five children aged between two and seven years old (mean age 4.25 years). The results showed an increase in chromatic discrimination ability with chronological age. However, for children aged between seven and 11, performance was better than expected when compared to previous data. It is unclear whether this better performance reflects wider inter-individual variation or increased competency at the task due to intellectual development. Since IQ measurements were not taken it is not possible to discount the latter. Furthermore, the CCT may not be an appropriate test of chromatic discrimination in children or children with developmental conditions. The essence of the CCT (and the child friendly version) is to construct a global structure from different local circle elements. There are developmental changes in the competency in using global information and this may not develop until later in typical development (12 years) (Kramer, Ellenberg, Leonard, & Share, 1996), which may lead to poorer performance is not necessarily related to poorer chromatic discrimination.

Per the Weak Central Coherence account of autism (see Chapter 1 section 4.1), individuals with autism are more likely to process information using a more local strategy (Happé & Frith, 2006; Mottron et al., 2006; Plaisted et al., 1999). Furthermore there may be difficulty in switching between global and local elements within a display (Rinehart, Bradshaw, Moss, Brereton, & Tonge, 2001). This overt focus on local elements again may lead to a poorer performance on the CCT that is not the result of chromatic discrimination for individuals with autism. Similar biases in attention towards local processing have also been suggested in WS using a variety of tasks such as drawing, and copying patterns where individuals with WS can copy local elements of a picture but struggle integrating this

into a global structure (Bellugi, Sabo, & Vaid, 1988; Bertrand, Mervis, & Isenberg, 1997; Farran, Jarrold, & Gathercole, 2003; Porter & Coltheart, 2006). Again, this suggests that the CCT may not be an appropriate test to measure chromatic discrimination in WS. In addition there is a similar suggestion that making judgements on global shapes is difficult for individuals with intellectual disability, suggesting that alternative methods which do not have such cognitive demands are more desirable in order for the individual to have adequate task comprehension and measure of chromatic discrimination (Barnhardt, Block, Deemer, Calder, & DeLand, 2006).

Another computerised test that has been widely used is the Colour Assessment and Diagnosis (CAD) test. The CAD test requires participants to identify the direction of a moving colour defined square against an equiluminant background with a random luminance masking technique (Barbur, Harlow, & Plant, 1994; Birch, Barbur, & Harlow, 1992; Rodriguez-Carmona, O'Neill-Biba, & Barbur, 2012). Chromatic discrimination is measured across sixteen directions within CIE (1931) space. From these sixteen interleaved directions, separate thresholds are calculated for "Blue-Yellow" and "Red-Green" thresholds. Normed data exist from over 300 different normal trichromat adults. The CAD test has been successfully applied in occupational settings such as aviation, fire service, and transport services (City University, 2014; Rodriguez-Carmona et al., 2012). The CAD test however also has potential problems for use with children and children with developmental conditions. The test is not just a measure of chromatic discrimination but relies heavily on the ability to detect motion. Motion coherence thresholds have been shown to be higher in both autism and WS ((Atkinson et al., 2003; Atkinson et al., 2006; Bertone et al., 2003), see also Chapter 1 section 4). Motion coherence thresholds also decrease with chronological age in typically developing children (Parrish, Giaschi, Boden, & Dougherty, 2005). The combination of these issues mean that the CAD test may not be appropriate for use with children or individuals with either autism or WS.

This section has described three of the most common measures of chromatic discrimination, all of which have some degree of standardisation at least from laboratory studies. The FM100 is a manual sorting task, whilst the CCT and CAD test are computerised psychophysical tests. These tests have been widely used, and in the case of the FM100 have even been used with children and adolescents with autism or WS. However, the appropriateness of these tests as a direct measure of chromatic discrimination for use with children and children with developmental conditions is questionable. This consideration has motivated the development of a new psychophysical task for this thesis's and related studies that is an independent measure of chromatic discrimination.

### 3.2.3 Chromatic Discrimination across the Lifespan

The FM100, CCT and CAD test and other laboratory measurements of chromatic discrimination ability, e.g. (Birch et al., 1992; Regan et al., 1994a), show that chromatic discrimination ability

changes over the life span. During infancy, chromatic discrimination develops at a slower rate than luminance discrimination which is constrained by the later physiological development of the parvocellular pathway compared to the magnocellular pathway (Dobkins, Anderson, & Lia, 1999; Kelly, Borchert, & Teller, 1997; Morrone, Burr, & Fiorentini, 1993; Teller & Palmer, 1996). Throughout childhood and adolescence chromatic discrimination ability continues to increase with sensitivity peaking in late adolescence/early adulthood (Barbur & Rodriguez-Carmona, In Press; Kinnear & Sahraie, 2002; Paramei & Oakley, 2014). It then decreases throughout the remaining adulthood (from 20s onwards) as the lens density decreases and yellows (Kinnear & Sahraie, 2002; Knoblauch et al., 1987; Knoblauch, Vital-Durand, & Barbur, 2001; Paramei & Oakley, 2014). There is some suggestion of different degradation of chromatic axes. Paramei and Oakley (2014) used the CCT in adults up to 80 years old. They found that the discrimination on the Tritan axis (Blue-Yellow axis) decreased at a faster rate (from 60 years) than either the deutan (Green) or protan (Red) axes. Performance on both manual and psychophysical tasks also supports this notion that discrimination for the Red-Green axis is better than the Blue-Yellow axis discrimination (Kinnear & Sahraie, 2002; Knoblauch et al., 2001; Paramei, 2012; Paramei & Oakley, 2014).

Using the FM100, there is also evidence of improvement in chromatic discrimination performance with age up to early adulthood, found in studies that establish age-dependent norms (Kinnear & Sahraie, 2002; Knoblauch et al., 1987) although this improvement probably reflects both a general increase in ability to complete seriation tasks in addition to the underlying increase in chromatic discrimination ability (Birch, 2001; Karpf et al., 1974).

In addition to variation of chromatic discrimination ability with age, there is also wide inter-individual variation. This inter-individual variation is seen on both manual and computerised chromatic discrimination tasks (Kinnear & Sahraie, 2002; Knoblauch et al., 2001; Paramei, 2012; Paramei & Oakley, 2014) and individual differences increase with chronological age, being most pronounced after 60 years (Paramei, 2012).

### 3.2.4 Discrimination in Atypical Development

In recent years, chromatic discrimination has been increasingly studied in developmental conditions including autism (Franklin, Sowden, et al., 2008; Franklin, Sowden, et al., 2010; P. Heaton et al., 2008; Hurlbert et al., 2011), ADHD (Banaschewski et al., 2006; Kim, Chen, & Tannock, 2014), and Williams Syndrome (Farran et al., 2013), as the extent of atypical sensory processing across all visual domains has become more evident in these disorders. It is therefore increasingly important to ensure that the tools, used to assess sensory processing, and colour perception, are both sensitive and specific in isolating sensory processing only. This requirement

is complicated by the hypothesised relationship between sensory processing and intelligence (Galton, 1883; Spearman, 1904). As per the original hypothesis of Galton and Spearman (Deary, 1994b; Galton, 1883; Spearman, 1904), higher intelligence is associated with better sensory discrimination abilities in the typical population. The support for this hypothesis has been mixed (Acton & Schroeder, 2001; Deary, 1994a, 1994b; Li et al., 1998; Melnick et al., 2013), with low correlations found between general intelligence and some measures of sensory discrimination (including colour perception), and other more recent evidence (Melnick et al., 2013) demonstrating a strong link between IQ and performance on a visual motion discrimination task. Nonetheless, the putative relationship makes it vital to ensure that tests of sensory processing are not confounded by direct contributions of general ability to performance. The extent to which this relationship between sensory and intelligence is present in autism is unknown. As far as the author is aware only one study has investigated this links: Meilleur and colleagues (2014) compared performance on low level visual (luminance contrast) and auditory (pitch discrimination) tasks and high level visual (pattern construction) tasks in adults with and without autism. Using multiple linear regression they identified that there were differences in the covariation in performance on the tasks between the autism and control groups, where there was covariation between in task performance between both low-level auditory and visual (Meilleur et al., 2014).

Alongside the work described in this thesis, there have been three prior direct studies of chromatic discrimination in autism. Heaton and colleagues (2008) used a three-alternative forced choice task where participants had to identify the “most different” colour within the display. Coloured stimuli varied in either 1, 2 or 3 Munsell steps for red, blue, green and yellow regions of Munsell colour space. They found that young adolescents with autism were significantly less accurate than chronological age matched typically developing controls. However, this study did not match for mental ability between the autism and TD group, instead mental age ability was compared to a moderate learning disability group, meaning that a direct comparison for mental ability with respect to typically development cannot be made. Furthermore, the task used was not a standard task for measuring chromatic discrimination and the stimuli did not map onto the physiological processes that underlie chromatic discrimination. The second study of chromatic discrimination in autism was conducted by Franklin and colleagues (2010). They used two different tasks to assess chromatic discrimination, the FM100 and a custom developed psychophysical task designed specifically for their study. On both tasks, they found that discrimination was poorer in an adolescent high functioning autism group compared to mental and chronological age matched TD control group. They found that the reduced chromatic discrimination in the autism group was a generalised deficit rather than specific to a single colour axis. The final study by Koh and colleagues (2010) used a psychophysical task to assess luminance and red-green contrast sensitivity in high functioning adolescents with

autism and a typically developing group matched on chronological and mental age, and a separate chronological and mental age group that was at a high risk for autism. They found that there was no difference between the autism and typically developing group for contrast sensitivity thresholds for the luminance and red-green gratings. However both the autism and typically developing had significantly poorer contrast sensitivity thresholds compared to the high risk group (Koh, Milne, & Dobkins, 2010). It is unclear why the high-risk group would have better chromatic discrimination than both the TD and the autism group. This finding goes against a similarly designed study in infants who were at either high or low risk for developing autism by McCleery and Colleagues (2007). McCleery and Colleagues (2007) found enhanced luminance, but no difference for red-green, contrast sensitivity thresholds in infants at high risk for developing autism (McCleery et al., 2007). Koh and colleagues (2010) reconcile these differences in findings by proposing that chromatic discrimination (and as such parvocellular activity) could potentially serve as a "preventative" factor against developing autism as the parvocellular pathway develops slower than the magnocellular pathway (Bosworth & Dobkins, 2009; Dobkins et al., 1999; Dobkins, Bosworth, & McCleery, 2009). Despite this suggestion it is unclear the extent to which chromatic discrimination or wider parvocellular serves as a preventative function as there is a lack of visual perception studies assessing visual function in individuals at high risk for developing autism, making this possibility difficult to assess (see also Chapter 1 section 3.1 for discussion of magnocellular and parvocellular functions in autism). The findings by Koh and colleagues (2010) on the surface appear to differ from Franklin and colleagues (2010), yet methodological differences between the studies may also reconcile the two studies. Both studies find that there is no difference between luminance and red-green thresholds. The study by Franklin and colleagues only find a group difference in chromatic discrimination between autism and TD with the addition of the blue-yellow axis, which was not measured by Koh and colleagues. Although Franklin and colleagues found no group differences for any individual colour axis they do all have distinct functional pathways (see Chapter 1 section 2.1). Furthermore, since Franklin and colleagues did not assess all colour axes for every participant. Thus, to date there has been no study which assesses luminance, red-green and blue-yellow axes within the same individual who has autism.

To date there has been just one study of chromatic discrimination in WS conducted by Farran and colleagues (2013) who used the FM100 to assess chromatic discrimination in adolescents compared to mental and chronological age matched controls. The WS group showed chromatic discrimination in line with the mental age matched control group but were found to have significantly poorer discrimination than their chronological age matched controls.

In the two studies that have used the FM100, and one additional measure of chromatic discrimination (in autism), both autism and WS groups have been shown to have poorer chromatic discrimination compared to typically developing controls of the same chronological age. However, in comparing the results of the two studies, one might conclude that there is a dissociation between groups regarding performance compared to their mental age, in which high functioning individuals with autism have been reported to perform worse than their mental age, and conversely WS individuals have been reported to perform at a level comparable to their mental age, despite similar mean chronological ages (young adolescents) between the WS and autism samples in the two studies.

### 3.2.5 Aims

This chapter assesses the chromatic discrimination ability on two different tasks (the FM100 and a computer-based chromatic discrimination threshold task) is assessed in adolescents with either Autism and Williams Syndrome relative to mental age controls. This will investigation of the typicality of the sensory processing of colour in autism and WS (Aim 1, see Chapter 1 section 7). A secondary aim is to determine the extent to which the relationship between non-verbal general ability and performance on the FM100 and psychophysical tasks varies with development, between typical and atypical development. A third aim was to investigate the extent to which performance on different chromatic discrimination tasks is related to intellectual ability. Data collected during this thesis have been published demonstrating the dissociation between intellectual ability and performance on chromatic discrimination tasks (see Cranwell et al., 2015).

## 3.3 Methods

### 3.3.1 Participants

Ninety-four participants took part in the study, split across four different participant groups on the basis the typicality of their development (autism/Williams Syndrome and a TD control group for each developmental condition (see table 3-1 for details).

All participants completed both the FM100 and the CCDT. Matches between participant groups were made using the RCPM as outlined in Chapter 2. An additional matching requirement was used for participants in this chapter. This was to match on which experimental set up for the CCDT was used. This was predominately the case for the autism group where some participants completed the CCDT using either the 8bit or 10bit setup. Seven participants in the autism group (and their appropriate matches) completed the 10bit version of the CCDT. There was one participant with WS who completed the 8bit CCDT version and was matched to an appropriate TD who also did the 8bit CCDT version. The remaining twenty-six participants in the WS group all completed the 10bit CCDT version and were matched to TD participants who also completed the 10bit CCDT version.

A number of participants were removed from the analysis due to FM100 Total Error Scores (TES) of over 500, implying poor task comprehension (Kinnear & Sahraie, 2002); These were three participants in the autism group (and their matches) and seven participants with WS (and their matches). Two participants with WS did not complete the FM100 due to time constraints. There was no significant difference in mean RCPM scores between the autism and control group, $t(28) = 1.04$, $p=0.31$, effect size = 0.19, Cohen's D = 0.38. There was also no significant difference in mean RCPM scores between the WS group and their control group, $t(35) = 0.13$ $p=0.9$, effect size = 0.02, Cohen's D = 0.04. Table 3-1 reports indices for the remaining participants, whose FM100 results were included in the analysis.

Some of the participants that are reported in this chapter were also included in the paper by Cranwell and colleagues (2015). The overlap of participants between those reported in this chapter and the paper were; one participant with WS (8bit CCDT & FM100), thirteen participants with Autism (8bit CCDT & FM100), two participants with autism (removed from both analyses due to FM100 TES above 500) and fifteen TD participants (11 8bit CCDT, 4 10bit CCDT & FM100).

*Table 3- 1- Chapter 3 participant demographics.* Chronological age is shown in months. Verbal and Non-Verbal IQ are shown as standardised scores. RCPM values are raw scores. In all groups, IQ was assessed using either the WISC Fourth Edition or WPPSI Third Edition and RCPM. Standard deviations are shown in brackets.

| Group | Chronological Age | Verbal IQ | Non-Verbal IQ | RCPM |
|---|---|---|---|---|
| TD (Autism Control) (n=20) | 80.63 (9.21) | 113.94 (14.21) | 107.83 (11.37) | 24.8 (3.73) |
| Autism (n=20) | 155.32 (28.76) | 74.33 (21.15) | 84.33 (19.45) | 26.73 (6.14) |
| TD (WS control) (N=27) | 67.71 (13.54) | 103.85 (12.62) | 96.85 (16.71) | 16.57 (4.56) |
| Williams Syndrome (n=27) | 153.69 (38.53) | 70.38 (13.39) | 54.85 (9.09) | 16.38 (4.52) |

### 3.3.2 Farnsworth-Munsell 100-Hue Test (FM100)

The Farnsworth-Munsell 100-Hue test (Farnsworth, 1957) is a measure of chromatic discrimination. It consists of 85 coloured caps split across four trays. The caps vary only in hue, with lightness and saturation kept constant. Each tray has 21 removable intermediate caps (except for the first tray where there are 22 caps) whose hues vary smoothly between those of the two fixed caps at either end. Standard administration procedures were followed: For each tray, the intermediate caps are removed from the tray and placed in a random arrangement while the participant looked away.

The participant was then asked to view and place the intermediate caps in the correct order in the tray between the two fixed caps, with as little difference in hue between neighbouring caps as possible. Standard prompts of, “Which colour is most like the one at the end?”, were used to ensure that the task was understood correctly. The trays were completed in different orders between participants. The order in which the participant placed the caps was recorded by the experimenter. The task was completed under simulated daylight illumination of colour temperature 6500K (D65) produced by a VeriVide D65 “Artificial Daylight” lamp.

Standard scoring procedures were followed. Error scores for each tray position are calculated from the differences between its chosen cap and the two neighbouring caps, generating a baseline score of 2 for each cap when in perfect order. Error scores for caps at the end of each tray were calculated using the neighbouring cap in the same tray and the first cap of the next tray, so that all caps are considered on a continuum around the colour circle. The Total Error Score (TES) is computed by first subtracting the baseline score from each tray position error score and then summing all 85 individual error scores. Specific anomalies of colour vision are revealed by specific error patterns (clustering of cap transposition errors along the protan, deutan or tritan axes).

### 3.3.3 Chromatic Contrast Discrimination Threshold Test (CCDT)

#### *3.3.3.1 Overview*

The Chromatic Contrast Discrimination Threshold (CCDT) was designed to isolate and assess discrimination within each of the cardinal chromatic mechanisms independently (Cranwell et al., 2015), similarly to the class of contemporary chromatic discrimination tests which include the Colour Assessment and Diagnosis (CAD) test (Birch et al., 1992) and the Cambridge Colour Test (CCT) (Regan et al., 1994a). It was developed for use in this and related studies of colour perception in children, with the requirements that the task should be engaging for children, portable, relatively quick to run, and easily reproduced without specialist equipment. Unlike the CAD and CCT, the CCDT requires only the detection of a form, and as such does not suffer from possible confounds of motion detection or local/global identification.

The thresholds measured by the CCDT are comparable to those from the CAD and CCT, although exact comparisons cannot be made because of differences in the specific shape discrimination task used and the background chromaticity and luminance. The CCDT differs from the Farnsworth-Munsell 100-Hue test in that it directly measures thresholds for chromatic discrimination around a point of neutral chromaticity, whereas the FM100 does not measure thresholds but instead requires the observers to detect (and then seriate) chromatic differences between colours at a fixed distance from neutral chromaticity. These chromatic differences have been selected to be near

threshold for normal observers (note that, in general, discrimination thresholds for hues of roughly equal luminance and saturation will differ from thresholds relative to a neutral chromaticity). Despite the differences between the two types of test, other studies suggest that the age dependence for both is similar, supporting the assumption that both rely on the same basic chromatic processing mechanisms.

*3.3.3.2 Apparatus and Setup*

Colour stimuli were displayed on a computer screen placed at the back of a black viewing box (36cm x 44.8cm x 62.3cm). Participants rested their heads on a chin rest placed centrally at the front of the box and viewed the screen through an aperture (13.5cm x 9cm) placed 21cm along the box length, from 62cm. One of two different computer setups was used to control the experiment, depending on the group: for participants completing the 8bit version, the experiment ran on a Dell Inspiron Laptop with stimuli displayed on its 14-inch screen; while for the 10bit version group the experiment ran on a custom built portable desktop tower, with standard components, running Windows 7 64-Bit edition with a PNY 600 10-bit graphics board with the stimuli displayed on a 10bit 23-inch Proart LCD monitor PA 238Q using a display port adapter. The same experimental programme (the Chromatic Contrast Discrimination Threshold test, or CCDT) was used for both setups, written in Matlab (v7.6.0, 2012b, The MathWorks, 2008, 2012), with graphics display functions from Psychtoolbox (Brainard, 1997) and colorimetric conversion functions from *kccv* (a set of Matlab routines based on standard formulae (Wolf, 2011) tailored for 8- or 10-bit displays appropriately); the 10-bit display used the NVIDIA QUADRO performance drivers. Spectral emission properties of both screens were characterised using a PR-650 spectroradiometer and colorimetric calibration tables were checked regularly using a Minolta CS-100 chromameter and updated when necessary to ensure colorimetric accuracy of the displayed stimuli.

*3.3.3.3 Stimuli*

On each trial, a single coloured arrow (visual angle = 1.83$^{o}$), pointing either leftwards or rightwards, was presented. The vertical position of the arrow was randomly jittered from trial to trial 5.51$^{o}$ above and below the central fixation point (visual angle = 0.92$^{o}$), on an achromatic grey background (CIE 1931 coordinates: x=0.36, y=0.37; Y=20.46 cd/m$^2$ for the 8-bit display; x=0.314, y=0.339; Y=64.8 cd/m$^2$ for the 10-bit display). The arrow colour was systematically varied in increments along only one of the three cone-opponent-contrast axes (Eskew et al., 1999) ( L-M or 'Red-Green'; S-(L+M) or 'Blue-Yellow'; L+M or 'luminance'). The just-noticeable difference in arrow colour with respect to the background was calculated in ΔE units in a perceptually uniform colour space (CIE L*u*v* space).

*3.3.3.4 Design*

A standard, computer-controlled staircase protocol was used to vary the colour difference between the arrow and background on each trial, stepping through differences on each half of the red-green, blue-yellow and luminance cone-opponent-contrast axes separately, beginning with supra-threshold difference values and moving in progressively smaller increments to difference values that are just reliably detected by the observer (for discussion of validation of such methods see chapter 2 section 2.5). A one-up/two-down procedure was used, in which the participant must be correct twice consecutively to go down the staircase (i.e. testing smaller colour differences) whereas an incorrect answer will take the participant up the staircase (i.e. testing larger colour differences). The details of step sizes are provided in the appendices. Each colour axis was tested in a separate block of trials, with a maximum of 100 trials per colour axis (50 for each colour direction of the axis; e.g. 50 each for "bluer" and "yellower") and a maximum number of 30 reversals per half-axis. Thresholds were calculated from the mean of the last four reversals.The contrast of the arrow stimulus with respect to the background was calculated in cone-opponent-contrast coordinates following the formulae given in Eskew et al. (1999). The origin of this coordinate space is given by the cone excitations to a reference white surface ($L_0$, $M_0$ and $S_0$), in this case, the uniform grey background, using the Smith-Pokorny (1975) cone fundamentals (Smith & Pokorny, 1975). Cone excitation values for the arrow are defined with respect to this origin by the ratios: $\Delta L = (L - L_0)/L_0$, $\Delta M = (M - M_0)/M_0$ and $\Delta S = (S - S_0)/S_0$, where L, M, and S are the cone excitations to the arrow stimulus. The cone-opponent contrast coordinates of the arrow stimulus are then calculated for each of three axes (see Chapter 2 Section 2.6.1 for more details on Eskew colour space). There were two independent staircases for each half-axis randomly interleaved in the one block. Each trial began with a 500ms centrally positioned white fixation dot, followed by the target which appeared for 150ms. The participant had to respond whether the arrow pointed to either the left of right, by pressing the corresponding mouse button. There was no time limit on the response, and the next trial began immediately after the response had been given.

This new task uses existing a 1-up/2-down staircase method that is well validated. Similar staircase techniques have been used in the study of similar chronological and mental age typically and atypically developing populations (Atkinson et al., 1997; Heaton, Williams, Cummins, & Happé, 2008; Spencer et al., 2000) (see Chapter 2 section 2.4.1). The use of colorimetry to ensures that stimuli accurately reflect cone-opponent activity. As outlined in Chapter 2 section 2.5, the approach taken here is in line with similar studies of chromatic discrimination with adults and infants (Dobkins et al., 1999; Knoblauch et al., 2001). The step sizes and number of reversals were chosen during prototype testing on adult participants. Furthermore, adult thresholds across all three cardinal axes were comparable with similar chronological age scores on other existing chromatic discrimination tests

(CCT and CAD). One of the motivations for the development of the CCDT was to apply such staircase and colorimetry to children. As such, early inspection of child participants' staircases confirmed that reversals and step sizes were appropriate.

#### *3.3.3.5 Procedure*

Participants sat in front of the viewing box. Prior to the start of the experiment, the researcher checked that the participant understood the difference between left and right. A short practice set of highly visible arrows (with supra-threshold luminance contrast) was administered to check that the participant understood the experiment. After the practice set the actual experiment began. In a game-like format, participants were presented with a choice for each condition (each depicted by a different storybook image) and performed each condition only once. The order of condition was not counterbalanced but there was no significant difference of order for thresholds on chromatic axes any of the groups (lowest p=0.11). Each colour-axis condition ran until either the maximum number of trials or maximum number of staircase reversals was reached. Once each condition had finished the participant was given a short break before continuing to the next condition. All conditions were either completed in one session or over two sessions.

## 3.4 Results

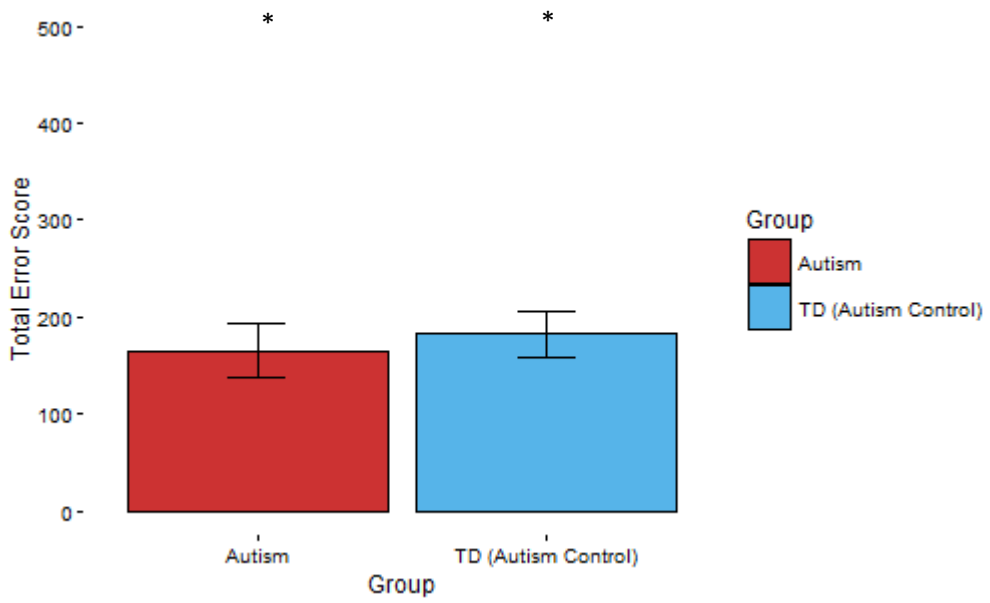
### 3.4.1 Farnsworth-Munsell 100-Hue Test Results

#### *3.4.1.1 Error Scores*

The error patterns of all included participants revealed no specific anomalies of colour vision. An independent samples T-test was conducted on the total error score for each developmental condition and their control group. There was no significant difference between the autism group and their control group, $t(30) = 0.51$, $p=0.61$ (Figure 3.1). A repeated measures ANOVA was conducted on partial error scores (PES), with a within-subjects factor of colour axis (Red-Green PES/Blue-Yellow PES) and a between-subjects factor of group (Autism/Control). There was a significant main effect of axis, $F(1,30) = 7.79$, $p < 0.01$, where the Red-Green PES were significantly lower than Blue-Yellow thresholds for both groups, $t(31) = 2.7$, $p < 0.05$. There was also no significant group by colour axis interaction, $F(1,30)=3.17$, $p = 0.085$. Results for the Williams Syndrome group mirrored those found in the autism group. There was also no difference between the Williams Syndrome group and their control group, $t(35) = 1.38$, $p=0.18$ (Figure 3.1). The repeated measures ANOVA also revealed a significant main effect of colour axis, $F(1,35) = 23.67$, $p < 0.001$, where the Red-Green PES was significantly lower than the Blue-Yellow PES, $t(36) = 5.22$, $p < 0.001$. The interaction between group and colour axis was not significant, $p=0.57$.

*3.4.1.2 Relation to previously reported age-norms*

Scores on the FM100 were compared with previously reported age expected norms based on small, presumptively typically developing populations Kinnear & Sahraie (2002). Difference scores were calculated between the actual score and expected chronological age score, where a positive score indicates better than expected performance and negative scores indicate worse than expected performance. T-tests were conducted for each group against a test value of 0. The autism group performed significantly worse than expected on their TES scores, $t$ (16) = 2.56, $p < 0.05$. The autism TD control group performed significantly better than expected on their TES scores $t$ (18) = 3.43, $p < 0.005$. The WS group performed significantly worse than expected on TES scores, $t$ (18) = 9.62, $p < 0.001$. The TES difference score was not significantly different from zero for the WS control group, $t$ (18) = 1.21, $p = 0.24$.

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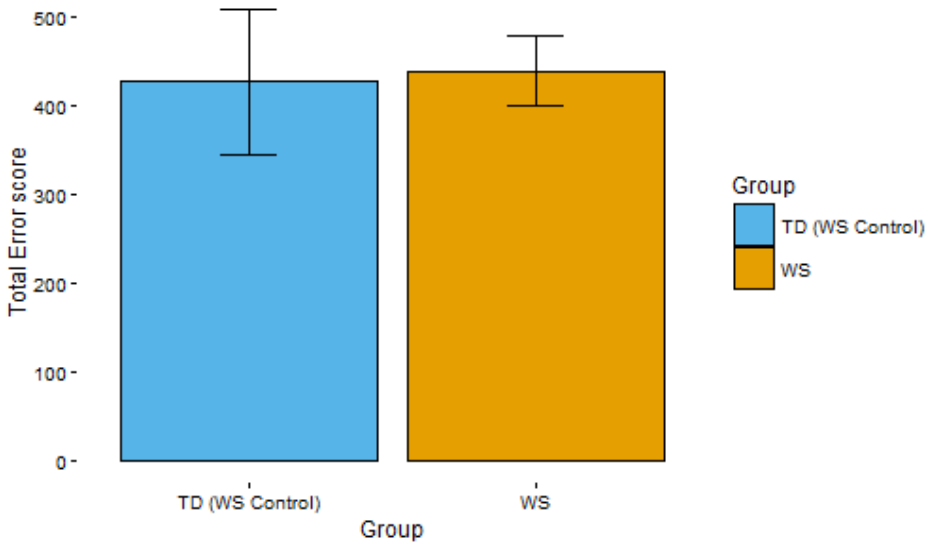
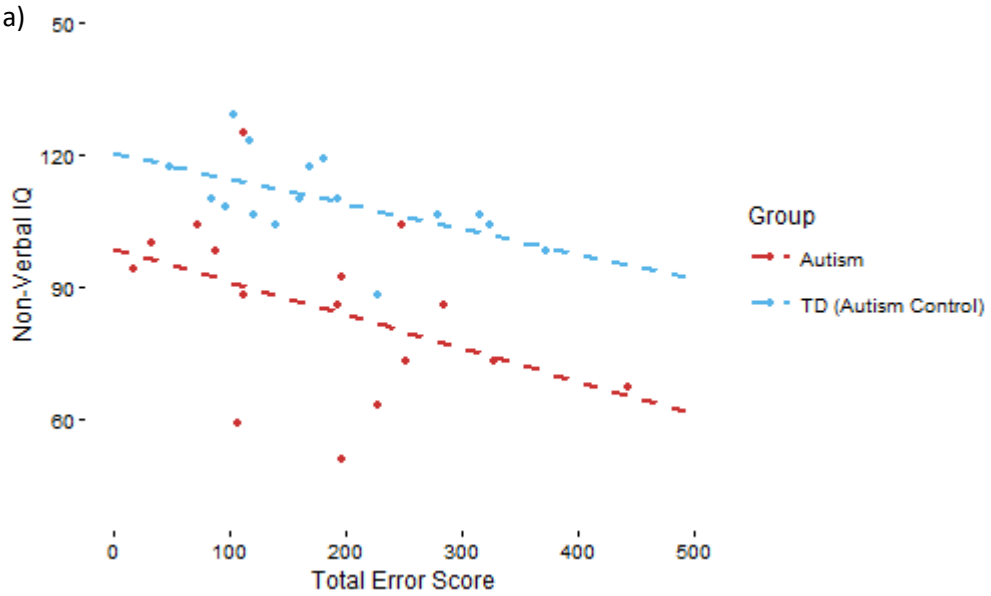



*Figure 3-1 - Total Error Scores (TES) for the FM100.* Higher TES reflects poorer performance on the test. Errors bars are +/- one standard error. The * denotes a significant difference from the age-appropriate norms reported by Kinnear & Sahraie (2002).

### *3.4.1.3 Correlation of IQ with TES (Figure 3.2)*

Correlations were calculated for each group between TES and standardised scores on VIQ and NVIQ subscales of respective IQ tests. There was a significant negative correlation between VIQ and TES in the autism group, $r$=-0.49, p<0.05. There was no significant correlation between VIQ and TES for the TD control group, $r$=-0.19, $p$=0.44. There were significant negative correlations between NVIQ and TES for both the autism, $r$=-0.53, $p$<0.05, and TD groups, $r$=-0.55, $p$<0.05. In the WS group there was no significant correlations between TES and either NVIQ, $r$=-0.18, $p$=0.18, or VIQ, $r$=-0.15, $p$=0.55. In the TD control group, there was a significant negative correlation between NVIQ and TES, $r$=-0.67, $p$<0.001, but no significant correlation between VIQ and TES, $r$=-0.2, $p$=0.45.

a)






b)

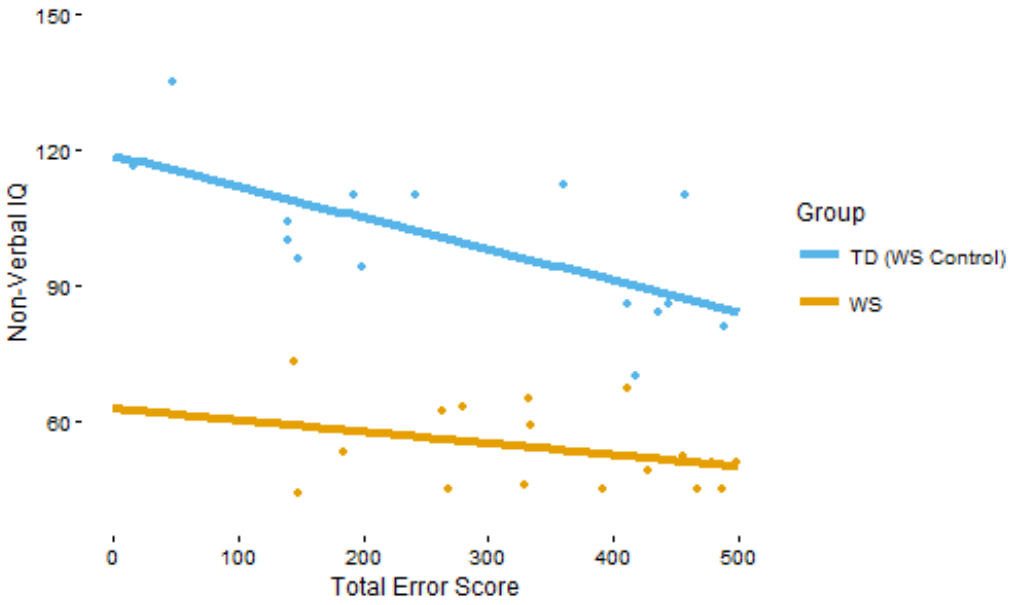



*Figure 3-2 – NVIQ scores plotted against FM100 Total Error Scores.* Lines are least-squares best fits for each group. In both graphs, TD groups are represented by filled triangles and dashed lines. (a) Autism: The TD group are denoted by blue circles and dashed linesand the Autism group by red circles and dashed lines. (b) WS: The TD group are denoted by filled blue circles and the WS group by filled orange circles and lines.

### 3.4.2 CCDT Results

#### *3.4.2.1 Discrimination thresholds on chromatic and luminance axes*

Individual staircases for each colour half-axis were analysed for convergence and excluded from further analysis if either of two conditions were met: (a) no reversals in the final 20% of trials or (b) final stimulus contrast less than or equal to zero, relative to background chromaticity (note that negative contrasts represent a positive contrast along the opposite half-axis; such stimuli occurred very rarely when the staircase attempted to step down at very low stimulus contrasts on the tested half-axis). If the staircase for one half-axis did not meet these conditions, the staircase in the other half-axis was also discarded from analysis. For any one axis (Red-Green (R-G), Blue-Yellow (B-Y), or luminance (LUM)). All staircases from the 10bit version were included.

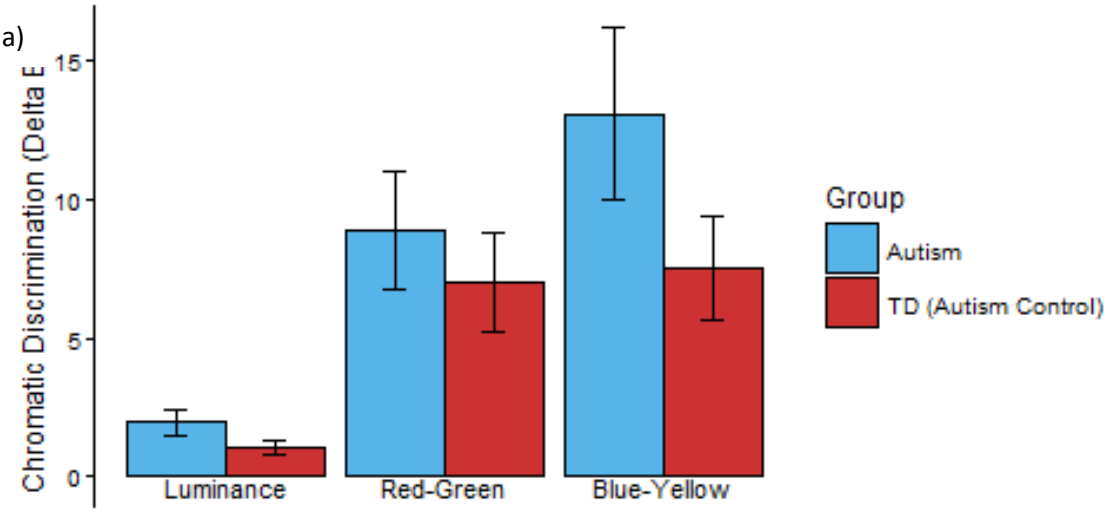
Threshold contrasts for each of the six half-axes were calculated in $\Delta E_{uv}$ units, as described in the methods section. The two thresholds for the two directions for each condition were then averaged together to give an overall threshold for each cardinal axis (e.g. blue and yellow individual thresholds were averaged together for a B-Y axis threshold). The threshold distributions for each colour axis, $\Delta E_{uv}$ thresholds were converted to a logarithmic scale to normalise the data.

To compare thresholds between colour axes in the autism and their TD control group, participants were also matched on whether their thresholds were collected from the 8-bit display

(n=11) and 10-bit display (n=7) systems. A two-way ANOVA with participant group (TD/Autism) and colour axis as fixed factors and threshold ($\Delta E_{uv}$) as dependent factor revealed a significant main effect for colour axis, $F(2,87)=80.88$, $p<0.01$. Post-hoc t-tests revealed that thresholds were significantly lower for the luminance than both R-G and B-Y colour axes for both TD (highest $p<0.001$) and autism groups (highest $p<0.001$). There was no significant difference between R-G and B-Y thresholds across all participants or for either participant group (lowest $p=0.2$). A significant main effect was also observed for group, $F(1,87)=4.93$, $p<0.05$. Post-hoc tests were conducted for each colour axis, which further revealed significant differences between groups on the B-Y axis, $t(29)=2.01$, $p<0.05$, with thresholds significantly higher for the autism group relative to the TD group, but not for luminance ($p=0.13$) or R-G axes ($p=0.1$).

In the WS analysis, a two-way repeated measures ANOVA was conducted, with a within-subjects factor of colour axis (Luminance/Red-Green/Blue-Yellow) and a between subject factor of group (WS/TD). There was a significant main effect of colour axis, $F(2,98)=188.64$, $p<0.001$. Post-hoc t-tests revealed that thresholds on the luminance axis were significantly lower than both the Red-Green, $t(50)=11.89$, $p<0.001$ and Blue-Yellow colour axes, $t(50)=24.6$, $p<0.001$. Furthermore, thresholds one the Red-Green axis were significantly lower the Blue-Yellow axis, $t(50)=5.58$, $p<0.001$. There was no significant main effect of group or group by colour axis interaction, lowest $p=0.7$.

a)




b)

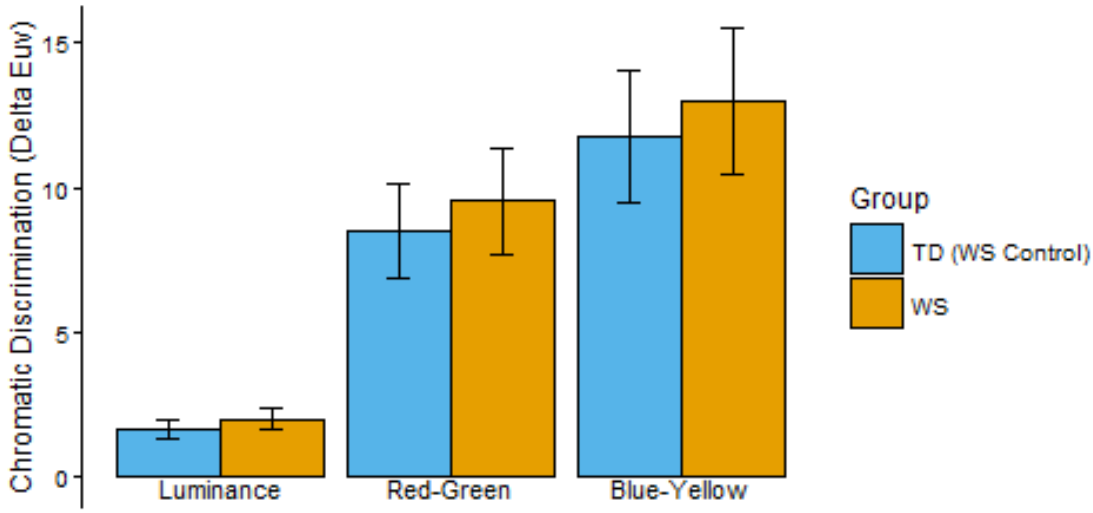



*Figure 3-3 – Chromatic Contrast Discrimination Threshold discrimination thresholds for the luminance, red-green and blue-yellow colour axes.* a) autism CCDT thresholds, b) Williams Syndrome CCDT thresholds For all three groups (TD, Autism and WS) are indicated by different colours, as shown in the legend. Error bars indicate standard errors.

#### *3.4.2.2 Correlations of Thresholds with IQ*

Correlation analyses were conducted to investigate reveal relationships between VIQ and NVIQ and discrimination thresholds on the three colour axes, using adjusted p-values to control for multiple comparisons for the different sets of analyses. There were no significant correlations between either VIQ or NVIQ in the autism (lowest $p$ = 0.12) or TD control group (lowest $p$ = 0.4). There were also no significant correlations found in either the WS (lowest $p$=0.18) or their TD control group (lowest $p$=0.42)

### 3.4.3 Cross Task Performance between FM100 and CCDT

Correlation analyses were conducted to examine the relationship between performance on the FM100 and CCDT tests. Performance was again split via participant group. There was no significant correlation for both TD groups between average CCDT threshold and TES, $r$=0.41, $p$=0.115. There was a significant correlation in the autism group, $r$=0.661, $p$<0.005. There was no significant correlation between tasks for the WS group, $r$=0.106, $p$=0.674, or their TD control group, $r$=0.405, $p$=0.086.

### 3.4.4 Correlation with Chronological Age

For the autism group, there was no significant correlation between chronological age and performance on the FM100 ($p$=0.122) or for the CCDT chromatic ($p$=0.124) or luminance ($p$=0.132) thresholds. The same pattern was also observed in the TD control groups for both FM100 ($p$=0.097) and chromatic ($p$=0.624) and luminance ($p$=0.126) thresholds on the CCDT. In the WS group, there were no significant correlations between chronological age and performance on either the FM100 ($p$=0.449) or on the CCDT ($p$=0.406). This was also the case for individual colour axes on each test

(lowest $p$=0.269). Both TD control groups there were significant correlations between chronological age and performance on the FM100, $r$=-0.627, $p$<0.005, and CCDT average chromatic threshold, $r$=-0.456, $p$<0.05, but not for luminance thresholds, $r$=-0.167, $p$=0.495. There was a significant correlation with both Red-Green PES, $r$=-0.712, $p$<0.001 and Blue-Yellow PES, $r$=-0.688, $p$<0.001. In the CCDT there was a dissociation between colour axes, where the Red-Green colour axis did not significantly correlate with chronological age, $r$=-0.184, $p$=0.45. However, there was a significant correlation between chronological age and Blue-Yellow colour axis, $r$=-0.642, $p$<0.005.

### 3.4.5 Drivers of Task Performance on FM100

To further assess the role of possible factors on FM100 performance, a multiple regression was carried out to estimate the extent to which performance on the FM100 might be predicted by NVIQ, chronological age and an independent measure of chromatic discrimination. This relationship was identified by Cranwell and colleagues (2015) and is further explored specifically for individuals with autism and Williams syndrome. For this purpose, the independent measure of chromatic discrimination was calculated as the average of B-Y and R-G thresholds from the CCDT test, which provided an overall measure of chromatic discrimination analogous to the TES on the FM100. Four predictors were included in the model: NVIQ, chronological age, development typicality (TD/Autism or WS) and chromatic discrimination threshold. The analysis included only participants who completed both the FM100 and the CCDT test, and only participants who had a TES of lower than 500. Predictors were entered into the regression model using the backward entry method, appropriate in the absence of an *a priori* theory for which predictors would explain the most variance in FM100 performance. Separate regression models were generated for autism and WS groups with their respective controls. The number of predictors was valid to detect a large effects for this sample size based on previously recommended values (Miles & Shevlin, 2001).

In the autism group a total of thirty-four participants were included. The regression analysis generated two models. Model 1 included all the variables, while Model 2 included all the variables except group (Autism/TD) since this factor explained the least amount of variance in FM100 performance (see Table 3-2). Both Model 1 and Model 2 significantly explained a large amount of variation in performance. Model 1 explained 52.5% of the variance, $F(4, 32) = 7.75$, $p < 0.001$, whilst Model 2 explained 50.4% of the variance, $F(3, 32) = 9.82$, $p < 0.001$.

Thirty-seven participants were included in the WS group. Again, there were two models generated. Model 1 included all the variables, while Model 2 included all the variables except chronological age since this factor explained the least amount of variance (See Table 3-3). Overall, both models

significantly explained substantial variance in the FM100 performance. Model 1 explained 43.1% of FM100 TES variance, $F(4, 32) = 5.31$, $p < 0.005$, While Model 2 explained 42.3% of FM100 TES variance, $F(3, 32) = 7.1$, $p < 0.001$.

*Table 3-2 - Backward stepwise regression model for the contributions to FM100 performance of the autism and TD groups.* The distinct factors for the autism analysis were: NVIQ, chromatic discrimination, chronological age and Group (Autism/TD). Model 1 includes all the predictor variables; Model 2 omits only Group (Autism/TD). The symbols *, **, ***,**** denote significance at *p*<0.05, *p*<0.01, *p*<0.005 and *p*<0.001 respectively

| | Model 1 | | | Model 2 | | |
|---|---|---|---|---|---|---|
| Variable | *B* | *SE B* | β | *B* | *SE B* | B |
| NVIQ | -3.16 | 0.928 | -0.575*** | -2.91 | 0.906 | -0.531*** |
| Chromatic Discrimination | 258.068 | 81.492 | 0.5*** | 215.082 | 72.253 | 0.417** |
| Chronological Age | -0.892 | 0.647 | -0.365 | -1.476 | 0.387 | -0.603*** |
| Group (Autism/TD) | 69.851 | 62.235 | 0.333 | | | |
| $R^2$ | | 0.525 | | | 0.504 | |
| F-value | | 7.745 | | | 9.819 | |

*Table 3-3- Backward stepwise regression model for the contributions to FM100 performance of the WS and TD groups.* The distinct factors for the WS analysis were: NVIQ, chromatic discrimination, chronological age and Group (WS/TD). Model 1 includes all the predictor variables; Model 2 omits only chronological age. The symbols *, **, ***, **** denote significance at p<0.05, p<0.01, p<0.005 and p<0.001 respectively.

| | Model 1 | | | Model 2 | | |
|---|---|---|---|---|---|---|
| Variable | *B* | *SE B* | β | *B* | *SE B* | B |
| NVIQ | -5.694 | 1.636 | -1.072*** | -3.743 | 0.859 | -0.76** |
| Chromatic Discrimination | 10.216 | 6.02 | 0.249 | 11.132 | 5.784 | 0.272 |

| | | | | | | |
|---|---|---|---|---|---|---|
| Chronological Age | -0.483 | 0.77 | -0.183 | | | |
| Group (WS/TD) | 151.49 | 122.69 | 0.546 | 209.09 | 80.82 | 0.753* |
| $R^2$ | | 0.431 | | | 0.423 | |
| F-value | | 5.31 | | | 7.1 | |

## 3.5 Discussion

The major finding in this study is that there is dissociation in chromatic discrimination between autism and WS. The autism group had significantly worse chromatic discrimination than non-verbal mental age controls (RCPM) and this was driven by poorer discrimination for the "blue-yellow" axis. There was no difference between the WS and TD groups. There were also differences in results between the FM100 and the psychophysical task. Non-verbal general ability differentially affects participants' performance on two different chromatic discrimination tasks. The results from the FM100 task show that performance was significantly associated with non-verbal ability in all groups except the WS group. Furthermore, this association is stronger in the autism group. The results from experiment 2 demonstrate, conversely, that this association between general ability and colour perception does not hold for the computer-based chromatic discrimination threshold test (CCDT) for either autism, WS or TD groups.

The results in this chapter show poorer chromatic discrimination for individuals with autism but not Williams syndrome relative to mental age matched typically developing controls. This finding is in line with results of the previous study that chromatic discrimination in Williams syndrome is in line with mental age (Farran et al., 2013). This result is expanded in this study using an additional psychophysical task. This finding is in line with similar previous research on chromatic discrimination in individuals with autism compared to mental and chronological age matched controls (Cranwell et al., 2015; Franklin, Sowden, et al., 2008; P. Heaton et al., 2008; Koh et al., 2010). However, the nature of this reduced chromatic discrimination was different from previous studies. Here there was a reduction, specifically in the "Blue-Yellow" axis. Previous research has found poorer general chromatic discrimination in autism (Franklin, Sowden, et al., 2010; P. Heaton et al., 2008). However, it is important to note the differences in tasks used to measure chromatic discrimination in these studies. Heaton and colleagues (2008) used a forced choice task compared to the manual sorting and psychophysical tasks used in this chapter. They did not use a psychophysical task instead using an alternative forced choice task using pairs varying

in different Munsell colour steps. However, there were very few pairs of stimuli (only 24 trials). In addition, by using Munsell colour space, the stimuli do map onto the underlying physiological responses of the different cones. Despite this Heaton and colleagues (2008) still find reduced chromatic discrimination in autism. However, it is not possible to compare this finding with the different colour axes with this study because the stimuli used by Heaton and colleagues do no isolate either the “red-green” or “blue-yellow” chromatic axes.

Franklin and colleagues (2010) used tasks that were more like the tasks used here. They assessed the performance of high functioning adolescents with autism compared to mental age and chronological age matched typically developing controls on the FM100 and a similar psychophysics task. On both tasks, there was significantly less accurate performance by the autism group, but there was no specific colour axis deficit found on either task. There are some key differences between this study and the current one which may explain the difference in results. The Franklin study tested a narrow age range of high functioning individuals with autism whilst the current study tests a much wider range of both age and ability. However, there is currently no evidence available that evaluates performance on sensory psychophysical tasks and whether this performance is influenced by diagnostic or other factors such autism severity, high or low functioning. Although not directly analogous there is some evidence that sensory sensitivities are partially related to certain demographic characteristics. For example, a reduction with chronological age and increased ability levels (see Introduction section 3.2.3 for further discussion).

Other differences can be found between the psychophysics tasks. The staircase procedure used by Franklin et al was based upon QUEST, whilst the task in this chapter uses a 1up/2down procedure. Little work has assessed the efficacy of different staircase procedures for use with children. One study has compared different psychophysical methods (Method of Constant Stimuli, 1up/2down, QUEST) on a speed discrimination task (Manning, Jones, Dekker, & Pellicano, 2015). They suggest that plotting the psychometric function may be more effective at revealing “true” performance on a psychophysical task, due to increased attentional capabilities in older participants compared to children where there are fewer attentional resources. Yet by comparison in the CCDT there are few trials and thresholds were not correlated with non-verbal ability. Furthermore, any differences in staircase methods between the CCDT and Franklin et al (2010) tasks are also unlikely to cause differences in the results for a particular colour axis. The stimuli were also different between the two studies. The Franklin task presented participants with a disc that was defined by a chromatic diagonal line. These stimuli were much larger (visual angle = 8°) compared to the one in the CCDT (visual angle = 2°). Different colour matching functions are differentially specified for either 2° or 10° and colour matching functions and subsequent calculated cone weightings are only valid for the visual angle under which the colour matching function was identified (Brainard & Stockman, 2010).

The appropriate visual field was used for this study but was not reported in the Franklin study. Regarding the chromatic properties of the stimuli there were different colour spaces was used between the studies, where the Franklin study uses Macleod-Boynton colour space (MacLeod & Boynton, 1979) and the CCDT uses Eskew (Eskew et al., 1999). This colour space is defined as a set of Cartesian coordinates which reflect the excitations of the different cone types within a plane of constant luminance. These cone excitations are based on the Smith and Pokorny cone fundamentals (Smith & Pokorny, 1975), see also Chapter two section 2.6.1 for further details on how colour spaces are constructed. The colour space used in the CCDT is the Eskew colour space (Eskew et al., 1999). Whilst also having coordinates that correspond to the three different cone activations, they use a different set of their own cone fundamentals. These colour spaces have slightly different weightings for the different cone classes. However, the thresholds in both studies are converted to Delta E, suggesting that this may not be an issue. Despite this there were differences between the studies in the starting positions and backgrounds. The starting position for the red-green axis was more orangish, and the blue-yellow position was more yellowish than the starting position compared to the CCDT stimuli starting positions. This difference should not necessarily relate to differential performance of the autism on chromatic axes between the studies. The differences in the colour of the backgrounds may have differential influence chromatic adaptation for the presented coloured stimuli. Another difference which participants completed different chromatic axes. Participants in the Franklin study completed the luminance axis and one chromatic axis, whilst participants in this study completed all three colour axes. This division of participants by Franklin make their results unclear. Firstly, the reported match statistics are at the task level and group level and are not stated for the separate analyses for each chromatic axis, indeed there are different numbers of participants between the groups for each colour axis. It is possible that this difference in numbers is reflected in the different results, qualitative inspection of the results show that blue-yellow thresholds are higher than TD participants. Nonetheless despite these differences it is difficult to ascertain their impact, therefore the only way to reconcile the differences between the study would be to assess the same set of participants on both tasks with appropriate matching criteria.

The origin of the differential reduction in performance along the blue-yellow axis in the ASC group is difficult to ascertain from this study. Processing of chromatic signals along the blue-yellow axis is anatomically and functionally distinct from red-green axis processing, and begins with the S cones in the retina (see Chapter 1 section 2.1), which feed into specialised cells in further layers of the retina: S-ON signals (changes towards “blue”) are segregated from S-OFF signals (changes towards “yellow”),

and distinct types of bipolar and small bistratified retinal ganglion cells carry these distinct signals from the bistratified ganglion cells which project differentially to koniocellular layers in LGN, depending on either S-ON or S-OFF response (Dacey & Lee, 1994; Martin & Lee, 2014). Less is known about the cortical pathway of the koniocellular layers after the LGN. S-cone specific responses have been found in V1 which is less specific to spatiotemporal variations compared to parvocellular and magnocellular responsive cells (Conway, 2014). Furthermore an addition pathway, in non-human primates, has also been proposed between koniocellular layers in the LGN and MT (Sincich, Park, Wohlgemuth, & Horton, 2004), though it remains to be seen whether such a pathway in humans.

Some evidence exists for differences between ASC and TD in visual processing generally at these distinct stages. For example, one study used electroretinograms (ERG) to study retinal responses to blue and red monochromatic light in adults with and without autism (Ritvo et al., 1988). There was a reduction in b-wave amplitudes, a measure of bipolar cell activity, in the autism group. Given that part of the retinal colour signal is processed through the bipolar cells, yet S-cone activity is reduced in the bipolar cells compared to the midget cells (Martin & Lee, 2014). It should be noted though, that the study by Ritvo and colleagues (1988) measures the response to monochromatic lights that are not spectrally tuned towards the peak sensitivities of each cone. Unfortunately, there are no published studies that look at specific colour axis processing in the retina of individuals with autism. Due to the sparse evidence in this area of autism research it is not possible to conclude whether the evident reduction in sensitivity on the “blue-yellow” axis originates within the retina. Another possibility is that there is a difference in processing within the parvocellular and koniocellular layers within the LGN. As outlined in Chapter 1 section 2.1 chromatic signals are processed predominately within the parvocellular layers. It has been recently suggested that the s-cone signals are processed in the koniocellular layer (Casagrande, 1994; Hendry & Reid, 2000; Tailby, Solomon, & Lennie, 2008). The results found here could be taken as preliminary evidence for possible koniocellular dysfunction in autism, although the location of this deficit within the physiology of the koniocellular pathway cannot be ascertained. Nor whether the different structure and connectivity within primary visual areas in individuals with autism is the cause (see Chapter 1 section 4.2). It is impossible to say which of these interpretations are true from the methods used within this study. Dysfunction of the koniocellular pathway has also been found in a wide range of conditions such diabetes (Feitosa-Santana et al., 2010), Alzheimer’s Disease (Regan et al., 1998), as well blue-yellow deficits discrimination identified in ADHD . However, yet this is the first demonstration of a specific koniocellular deficit in autism. Therefore, future research may explore possible abnormalities in the koniocellular pathway in autism and in particular utilising neuroimaging methods to determine where this deficit occurs.

The results in this thesis are in line with previous reports of poorer chromatic discrimination in individuals with autism (Franklin, Sowden, et al., 2010; P. Heaton et al., 2008). However current accounts of visual functioning of autism have not included consideration of chromatic discrimination. It has been proposed that individuals with autism have enhanced perceptual functioning (Mottron et al., 2006) or a focus on details at the expense of the whole (Happé & Frith, 2006). To date this research has not considered aspects of colour vision. Furthermore, this reduction in chromatic discrimination does not align with processing of other low level sensory information within the ventral stream, reported in the literature. Thresholds for orientation of oblique lines have been shown to be lower (better performance) in adults with autism compared to chronological age controls (Bertone et al., 2005; Dicksinson, Bruyns-Haylett, Jones, & Milne, 2015). It is possible that there may be a dissociation of ventral stream functions in autism, where chromatic discrimination is reduced but orientation discrimination is enhanced. This may reflect the different underlying physiological processing of orientation and chromatic information in primary visual areas.

The WS results replicate and extend the single previous study to have assessed chromatic discrimination. Farran and colleagues (2013) used the FM100 to assess chromatic discrimination in a similar chronological and mental age group with WS relative to separate mental and chronological age matches. There was no difference between the WS and mental age control group, but the WS group had significantly reduced chromatic discrimination relative to the chronological age group. The current study using a similar WS group in terms of ability and age range, replicated Farran and colleagues (2013) result, where there was no difference between WS and a mental age control group. There is no chronological age control group in this study, however the WS group performed significantly poorer with respect to the Kinnear and Sahraie (2002) age expected norms. This suggests that this difference would also be present with the current WS sample. Elsewhere the relationship between FM100 performance with NVIQ, and limitations of the Kinnear Sahraie FM100 norms (for further discussion see below) have been shown with a different subset of participants (Cranwell et al., 2015). Therefore, there is a need for other tests to be used to assess chromatic discrimination, particularly those tests where there are complex decisions made about the stimuli (see introduction of this chapter for more details). The results on the psychophysics chromatic discrimination task (CCDT) expand on those reported by Farran and colleagues (2013), showing that there is no difference between WS and mental age controls in their chromatic discrimination when using a task that has been shown to be independent of ability. The findings here combined with those observed by Farran and colleagues (2013) suggest that chromatic discrimination is intact in WS relative to their mental age.

The findings are in line with the wider visual functioning profile in WS. A dorsal stream deficit has been proposed for WS, for which numerous studies have shown elevated

motion coherence thresholds (Atkinson et al., 2003; Atkinson et al., 2006; Atkinson et al., 1997). Colour meanwhile is processed by the ventral stream (see Chapter 1 Section 2). Other low level ventral stream functions have been shown to be intact with respect to mental age in WS. For example form identification and orientation discrimination have been shown to be relatively intact (Atkinson et al., 1997). However, it is has also been suggested that the development of orientation may follow a different developmental trajectory compared to typical development (Farran, 2006; Palomares, Englund, & Ahlers, 2011; Palomares, Landau, & Egeth, 2009). Although there was no measure of dorsal stream taken in this study, the results here do provide further evidence for relative typicality in ventral stream function in WS with respect to mental age. Specifically, that chromatic discrimination represents a developmental delay with respect to their chronological age.

### 3.5.1 Appropriateness of Chromatic Discrimination Tasks

A wide range of chromatic discrimination tests are available; but it is unclear whether they are appropriate for use with children or children with developmental conditions. The results of this study support previous findings that cognitive factors (such as non-verbal ability) unrelated to chromatic discrimination ability may well influence performance on the FM100 task (Cranwell et al., 2015; Dain & Ling, 2009; Hurlbert et al., 2011). The FM100 performance-NVIQ correlation slopes are steeper and more significant in the autism group compared to the TD group. Furthermore, the results of the regression model suggest that non-verbal ability is a significant predictor of performance in addition to age and the ability to discriminate between colours. When considering why the FM100 is associated with general cognitive ability but the chromatic discrimination threshold test is not, the relative task demands are important. Successful performance on the FM100 requires attentional and visuospatial abilities in addition to chromatic discrimination ability. Spatial comparisons are required between the selected and non-selected caps. Attention switching between the local field – in making a comparison between two adjacent caps, and the global field - in overseeing the entire colour gradient - is also essential for good performance. These task demands may be influenced by different factors in each group. That is not to say that the FM100 is not a measure chromatic discrimination, but that it is not a pure measurement of chromatic discrimination and measures non-verbal ability. There was also a significant correlation between scores on the FM100 and CCDT thresholds for the autism group. Although there was no correlation in any of the other groups this can be explained by general task performance. In the Williams syndrome analysis error scores were close to ceiling in both Williams syndrome and TD groups suggesting that the participants in these groups found the FM100 particularly difficult to complete. Furthermore, the Autism regression analyses performance on the CCDT was a significant predictor, although non-verbal ability predicted a greater amount variance than CCDT thresholds.

In typically developing children competency of global processing does not develop until late into childhood (Kramer et al., 1996). Individuals with ASC are more likely to process visual information locally rather than globally (Plaisted et al., 1999) and to have difficulty switching between local and global processing (Rinehart et al., 2001). Non-verbal ability may also differentially affect performance between groups. Better performance in the older TD groups may reflect more mature global processing competency than in the younger TD groups. Poorer performance in the ASC groups may be the result of difficulty both in sustaining and switching of attention between local and global fields of the FM100.

These results have implications for the FM100 norms that have previously been reported (Kinnear & Sahraie, 2002). The findings from the Kinnear & Sahraie study implicitly assume that performance is unrelated to IQ and that task demands are consistent between different ages. In the current study, both TD child groups are above average in non-verbal ability and perform significantly better than expected from the Kinnear and Sahraie (2002) norms. Given that the number of participants in both studies is similar for each respective age group, this comparison calls into question the reliability of the norms reported for these age groups. Further for the younger age group there is the additional concern about the small sample size (e.g. 9 participants for age 5 years) (Kinnear & Sahraie, 2002). These results, in combination with the findings from a preliminary study by Hurlbert and colleagues (Hurlbert et al., 2011) indicate that caution must be exercised when using FM100 norms, for all ages, but especially for younger children or clinical populations where the NVIQ is lower than average with respect to chronological age, given that the relationship between FM100 performance and NVIQ appears to be stronger than previously recorded.

In comparison to the FM100, the CCDT has fewer task demands. This test of chromatic discrimination requires attention on a trial-by-trial basis only to identify the direction of an arrow. Like other standardised tests in whose class the CCDT falls (e.g. the CCT (Regan et al., 1994a)and the CAD (Birch et al., 1992) ), the CCDT measures discrimination thresholds along isolated chromatic directions away from a fixed adaptation point. Although other colour discrimination tasks have been adapted for use with children (e.g. the CCT; (Goulart et al., 2008)), to our knowledge these have yet to be demonstrated as independent of general ability. Other standardised chromatic discrimination threshold tests call on more complex aspects of visual processing which may introduce additional confounds when used in children: the CCT (Regan, Reffin, & Mollon, 1994b), for example, requires participants to identify a global shape composed from local elements while the CAD (or City Colour Vision Test(Barbur et al., 1994)) requires participants to discriminate the direction of a moving stimulus(Barbur et al., 1994; Birch et al., 1992). The former thus presents a potential

confound in distinguishing between deficits in local chromatic discrimination vs global shape processing, while the latter may be unable to dissociate between deficits in motion direction discrimination vs chromatic discrimination, which are known to develop at different rates in children (Knoblauch et al., 2001; Parrish et al., 2005) The CCDT task used in this study may provide a more direct measure of chromatic discrimination by being a simple shape identification task, which requires only a coarse binary judgement of left versus right, does not depend on numeracy or literacy skills, and does not involve a trade-off between local and global processing, involving the discrimination of only a single large shape against a uniform background. Performance is more likely to be independent of developmental stage or cognitive ability, allowing for age variations in chromatic discrimination to be more accurately captured. Because participants continue the task only until they reach their own individual threshold, task difficulty also remains constant between individuals even though other factors such as chronological age or chromatic discrimination ability may differ between participants. Because of these shared properties, we would expect CCDT performance to show the same pattern of age dependence as that demonstrated for the CCT and CAD (Barbur & Rodriguez-Carmona, In Press) , and our ongoing studies support this expectation. Although we have assessed the relationship between CCDT performance and only in the younger age groups and therefore demonstrated this independence from cognitive ability only for those groups, we would expect the independence to hold for the adult groups also, particularly given their better chromatic discrimination ability, higher absolute IQ, and increased attentional capacity relative to children (McAvinue et al., 2012).

## 3.6 Conclusion

In summary, there is dissociation in chromatic discrimination ability between autism and WS relative to typical development, where the autism group showed poorer chromatic discrimination while the WS group showed no difference when compared to mental age TD individuals. The results also highlighted the importance of using a developmentally appropriate test. The dissociation between autism and WS performance was only revealed when using a task that was independent of general ability. The appropriate use of a psychophysical task which has fewer task demands and is of equal difficulty across all ages will give a more accurate measure of colour discrimination and ultimately visual function in children, in both typically developing and children with developmental conditions.

# Chapter 4 – Colour Preference

## 4.1 Overview

The previous chapter investigated low-level sensory processing using two different tests of chromatic discrimination. This chapter reports assessments of a higher-level behavioural response to colour, the affective response, measured via a colour preference task. From a basic science point of view, there are two reasons to explore colour preference in autism and Williams Syndrome: Firstly, to determine whether simple tests of colour preference may capture, reflect or predict extreme behavioural responses to colours in daily life; (2) To determine whether colour preference responses are typical and therefore whether affective responses to visual stimuli are typical in these populations, and, whether they are age-appropriate. Although the origins of colour preference are not fully understood, expression of colour preference requires some degree of chromatic discrimination, and neural linking of the emotional system with visual stimuli as well as abstraction of cultural influences. Therefore, colour preference may also indicate typicality between low-level sensory processing and cultural norms. Understanding the origins and patterns of colour preferences in autism and Williams Syndrome, relative to TD controls, is also important from the applied science viewpoint because colour is used widely in both primary and secondary interventions (e.g. sensory rooms) and in clinical or teaching guidelines, yet there is relatively little evidence to support current colour choices in these areas.

## 4.2 Introduction

Colour preference is an expression of the simplest affective response to colours liking or disliking colours. Colour preferences may have a wide effect on an individual's behaviour, for example, in choosing clothing or room decorations. A wide range of research has been conducted on colour preference, encompassing studies from psychology, advertising and interior design, comprehensive coverage of which is beyond the scope of this chapter. The focus of this brief literature review will relate instead to factors which modulate individual differences in colour preference. Examples of these include sex or culture (for reviews see (Hurlbert & Owen, 2015; Palmer, Schloss, & Sammartino, 2013)), developmental changes in colour preference, and anecdotal accounts and focus group studies of responses to colour in individuals with autism or Williams Syndrome. Models and theories that attempt to predict preference patterns or explain the origins of colour preference will also briefly be introduced (Hurlbert & Ling, 2007; Ou, Luo, Woodcock, & Wright, 2004a; Schloss & Palmer, 2010).

### 4.2.1 Individual Differences in colour preference

Despite evidence for consistent underlying patterns in colour preference that occur along different dimensions, there are significant inter-individual variations in colour preference. This section will

outline research findings on the modulating factors that contribute to these inter-individual variations.

#### *4.2.1.1 Colour Preference variations in hue, saturation and lightness*

Early studies of colour preference gave mixed and contradictory results due to uncontrolled or unreported colour stimuli (McManus, Jones, & Cottrell, 1981). Recent studies using controlled colour stimuli have identified that despite inter-individual differences in colour preference, at the wider population level there are systematic differences in colour preference which depend on the hue, saturation and lightness properties of the stimuli (see Chapter 2 section 2.6.2 definitions of these properties) (Camgöz, Yener, & Güvenç, 2002; Hurlbert & Ling, 2007; McManus et al., 1981; Ou et al., 2004a; Palmer & Schloss, 2010).

In Western adults (UK and American) at the population level, there is a relative peak in preference for blue and green hues and relative trough in preference for yellows and orange (Camgöz et al., 2002; Hurlbert & Ling, 2007; Palmer & Schloss, 2010). Furthermore, individual colour preference was predicted by either 'blue-yellow' (S-(L+M)) or 'red-green' (L-M) cone contrasts weightings of the colour preference stimuli (see Chapter 1 section 2.1 for definitions of cone-contrast encodings of colour). Variation along the 'blue-yellow' axis accounted for the most variation of colour preference (Hurlbert & Ling, 2007; Palmer & Schloss, 2010). There tends to be greater preference for highly saturated colours compared to low saturated colours regardless of hue (Camgöz et al., 2002; McManus et al., 1981; Ou, Luo, Woodcock, & Wright, 2004b; Palmer & Schloss, 2010; Taylor & Franklin, 2012). The same is also true for lightness, with a tendency for increased preference for lighter colours (Guilford & Smith, 1959; McManus et al., 1981; Taylor & Franklin, 2012). Lightness preferences, though, are clearly modulated by individual factors such as sex and culture, and by interactions between hue and lightness or saturation, suggesting that each hue has a specific "peak preference" for differing saturation and lightness levels (Guilford & Smith, 1959; Ou et al., 2004b; Palmer & Schloss, 2010; Taylor & Franklin, 2012). For example, yellow and olive or orange and brown can be separated by differing levels of lightness.

#### *4.2.1.2 Cultural variations in colour preferences*

The previous section described studies on colour preference in Western countries. However, there are differences in colour preferences between cultures. Hulbert and Ling (2007) found that the hue preference curve for Chinese adults was shifted towards redder hues, compared with UK adults who preferred bluer and greener hues. Further hue specific differences have been found for other Asian countries, such as Japan and Indonesia relative to western countries (Fushikida, Schloss, Yokosawa, & Palmer, 2009; Saito, 1996; Sorokowski, Sorokowska, & Witzel, 2014). Differences in colour preferences have been found between industrialised and non-industrialised countries. For example, the Himba tribe, a rural tribe found in Namibia, differ in their colour preference patterns relative to

UK adults. The Himba tribe show colour preferences that are less dependent on hue and more dependent on saturation (or, specifically, chroma) (Taylor, Clifford, & Franklin, 2013).

Although the aim of this thesis is not to explore cross- cultural differences in preference, these differences are important to acknowledge, as they illustrate the extent that to which colour preference can, at least in part, be explained at the group level by the culture itself. Strong concordance of an individual's colour preference behaviour with the culturally specific pattern would also suggest that the individual must be susceptible to cultural influence and must be able to "extract" the relevant valence ratings for different colours within their culture. Given the social difficulties that characterise both autism and Williams syndrome (see Chapter 1 sections 1.2 and 1.3 respectively), it might be that they are less susceptible to cultural influence and show different qualitative patterns in their colour preference behaviour.

#### *4.2.1.3 Developmental Changes in Colour Preference*

Colour preference also changes with chronological age. The previous two sections have discussed colour preference results in adults; this section will focus on studies of colour preference with infant and child participants. Infant colour preference studies use habituation paradigms to present two different colours side by side. Total looking time and fixations are then used to infer colour preference in the infants (Adams, 1987; Bornstein, 1975; Franklin, Bevis, Ling, & Hurlbert, 2010; Franklin, Pitchford, et al., 2008; Taylor, Schloss, Palmer, & Franklin, 2013; Teller, Civan, & Bronson-Castain, 2004; Zemach, Chang, & Teller, 2007). The infants used in these studies are from 3 months old since infant vision becomes trichromatic at this time (e.g. (Knoblauch et al., 2001). When considering variations along the hue dimension, infants showed increased preference around blue hues and decreased preference for yellows and increased preference for saturated colours (Bornstein, 1975; Franklin, Pitchford, et al., 2008; Teller et al., 2004; Zemach et al., 2007). Although this pattern is not always present (Adams, 1987), it should be noted that the stimuli used in these studies vary in their lightness and saturation. To directly compare infant and adult colour preference patterns the same stimulus set needs to be used. Franklin and colleagues (2010) used the same stimuli that were used by Hurlbert and Ling (2007). They found that unlike adult hue preferences which vary most along the 'blue-yellow' axis, infant colour preferences varied more along the 'red-green' axis, with higher preferences for redder hues were more preferred. There are also differences in colour preference between adults and infants for the interaction between lightness and hue. Taylor and colleagues (2013) compared infant and adult colour preferences for stimuli that varied in hue and lightness, finding that infants showed increased preference for light red and dark yellow, and decreased preference for light blue and dark green. Curiously, adults in this study showed the opposite pattern in colour preference for the same stimuli, suggesting that there are age related changes in hue preference.

By comparison, far fewer systematic studies have been conducted on colour preferences in young children and adolescents. Zentner (2001) reports that in young children (3-4 years) red, pink and dark blue were most preferred, with no difference in the qualitative colour preferences between males and females (Zentner, 2001). However, the number of different coloured stimuli used by Zentner (2001) was very small and did not systematically vary in either hue, saturation or lightness, making it difficult to determine which attributes contributed to the reported preferences. Child and colleagues (1968) found that preference for saturation also changes with age, with the preference for higher saturation colours shown by young children (aged 6-9 years) decreasing into adolescence (12-13 years and 17-18 years). A similar pattern was observed for lightness, where high lightness colours preferred more than low lightness colours (Child, Hansen, & Hornbeck, 1968a). Specifically examining hue preference, at constant lightness and saturation, Ling and Hurlbert (2011) found that hue preferences changed with sex and age, with the most pronounced preference patterns and the most pronounced sex differences in preference seen in adolescence compared with younger age groups.

Other studies have used varying tasks to identify colour preferences in young children, for example, free drawing at 5-6 years (Iijima, Arisaka, Minamoto, & Arai, 2001) ,colour selection from array at 3-12 years (Burkitt, Barrett, & Davis, 2003; Chiu et al., 2006) or coloured toy selection at 3-6 years (Burkitt et al., 2003; Chiu et al., 2006; Iijima et al., 2001; Picariello, Greenberg, & Pillemer, 1990)) ; all report sex differences, with females preferring to use or select pink and purple, and males preferring blue, red or brown (Burkitt et al., 2003; Iijima et al., 2001; Picariello et al., 1990). However, these studies do not use controlled colour stimuli, and it is possible that differences in saturation and lightness between the stimuli are driving colour preference instead of hue. Further changes in colour preference are seen throughout adulthood into old age. For example, blues become less popular with age, whilst green and red become more popular (Dittmar, 2001). Hue preference patterns specifically show reductions in peak preferences in the blue region as well as less pronounced sex differences at older ages (> 60 years) (Ling & Hurlbert, 2011).

*4.2.1.4 Sex Differences in colour preference.*

Differences in colour preference between males and females are popularly assumed to exist, but not always evident in empirical studies, possibly partly because of differences in methodologies as well as other contributing factors. Although there are reports of no evidence of sex differences in infant studies of colour preference (Bornstein, 1975; Franklin, Bevis, et al., 2010; Franklin, Pitchford, et al., 2008; Taylor, Schloss, et al., 2013; Teller et al., 2004; Zemach et al., 2007), there have been reported sex differences in colour preference for children, adolescents and older adults. These occur in two different forms: difference in the preference values for specific colour regions, and differences in the overall preference variation or stability across colours and/or time. For example, there is recurring evidence for a female preference for reddish colours, in adults (Hurlbert & Ling, 2007; McManus et

al., 1981; Saito, 1996), as well as in children (Burkitt et al., 2003; Chiu et al., 2006; LoBue & DeLoache, 2011). Nonetheless, extrapolating to general statements of the existence and/or development of sex differences in colour preference remains difficult because of variations in methodology between specific studies. For example, Chiu and colleagues (2006), used a colour-selection task in which children (aged 3-12 years) chose their favourite colour from an array of 144 colours varying in hue, lightness and saturation, which were subsequently indexed with the colour name provided by the child. There was a significant sex difference in the first choice of colour, with females showing significantly higher preference for colours named "pink" or "purple" than boys, whilst boys showed significantly higher preference for "reds", and both sexes showing the same preference for "blues". In the free drawing task of Iijima and colleagues (2001), girls aged 5-6 years used significantly more "pink" colours than did boys, whereas boys used significantly more "blues" (Iijima et al., 2001). These sex-specific colours preferences in young children have been attributed to the influence of stereotypically coloured toys and clothing (Jadva, Hines, & Golombok, 2010; LoBue & DeLoache, 2011). Other studies have demonstrated that children as young as 3 years of age choose their favourite toys based on colour according to their own sex-stereotype (Picariello et al., 1990).

The colour stimuli used in the above studies are not characterised in terms of their colorimetric properties, preventing replication or systematic comparisons. The lack of sex differences found in some studies (Camgöz et al., 2002; Child et al., 1968a; Ou et al., 2004b; Zentner, 2001), may similarly be due to unreported variations in stimulus attributes of hue, saturation and lightness. Investigation of hue preference using stimuli controlled for lightness and saturation reveals that sex differences in hue preference continue into adolescence and early adulthood, with significant differences in preference in the purple and red-purple hue region (female > male) and green and green-yellow region (male > female) (Ling & Hurlbert 2011). Preferences for blue hues are equal in both sexes (Ling & Hurlbert, 2011). These results agree with a larger study of young UK adults, in which males preferred hues with negative weightings on their L-M cone-contrast component (colours with greenish contrast against the background; see Models of Colour Preference section below) and females preferred hues with positive weightings on the same component ("redder" colours) (Hurlbert & Ling, 2007). However in older adults (50+ years) the sex differences in hue preference are no longer significant (Bonnardel, Harper, Duffie, & Bimler, 2006; Ling & Hurlbert, 2011).

There is also no sex differences in colour preference in young adults (Camgöz et al., 2002; Ou et al., 2004b). This mismatch between studies may in part be due to variations in statistical power, with some studies lacking sufficient participant numbers to demonstrate differences in preference across the hue dimension, where there are usually the most variation of coloured stimuli.

Sex differences in preference for saturation and lightness dimensions of colour have also been found. Young female children (aged 6-9 years) preferred lighter colours compared to males of a similar age,

and this increased preference for lighter colours increased with age into adolescence (Child et al., 1968a). Child and colleagues (1968) also found that females were also more likely to prefer saturated colours relative to their male counterparts, although these sex differences also been found to change with age. There are reductions in the relative differences when comparing early adulthood (twenties) to older individuals (50+ years) (Bonnardel et al., 2006; Ling & Hurlbert, 2011).

#### *4.2.1.5 Summary of Individual Differences in Colour Preferences*

The preceding sections have highlighted that colour preferences are modulated by different factors, such as age, sex and culture, which may also interact with each other in influencing preferences for hue, saturation and lightness. For example, although sex differences in infancy have not been reported, by the time that the individual has gone into childhood or adolescence then sex differences with respect to hue, lightness and saturation preference do appear. Finally, the sex differences in colour preference reduce in late adulthood. This section has highlighted different modulating factors on colour preference, however it does not necessarily explain why such differences occur. The various modulating factors of colour preference show how it will be important to control for these factors as much as possible in this study.

### 4.2.2 Models of colour preference

The above section demonstrates that there are broad variations in colour preferences across individuals, influenced by sex, culture and age. Despite these variations, underlying consistencies in preference patterns are reported in experimental and observational studies. Although various studies have investigated the effects of various factors on colour preference, e.g. (Eysenck, 1941; Hurlbert & Ling, 2007; McManus et al., 1981; Ou et al., 2004a; Palmer & Schloss, 2010), see (Hurlbert & Owen, 2015; Palmer et al., 2013) for reviews), in contrast there are fewer investigations into why colour preference occurs. Instead the emphasis has been on "*what"* colours individuals like and dislike, but not *"why"* certain colours are liked and disliked. The basis of several current theories and models is that the preference for certain colours is driven by the behavioural significance of those colours in nature, and by the significance of the objects associated with those colours. Hurlbert and Ling (2007) suggest that colour preferences are driven by evolutionary pressures. For example, foraging for ripe food or searching for a suitable mate are associated with colours. If these behavioural drivers are evolutionarily embedded, then the neural encoding of colours themselves may reflect the encoding of preferences. Thus, the extent to which preferences are predicted by low-level neural encoding mechanisms of colour may correspond to the extent of universality in colour preferences. Alternatively, if colour preferences are instead largely the result of transfer from object preferences developed during the individual's lifespan rather than develop across evolutionary time frames, there may be much greater individual variability in preference than predicted by sensory encoding of colour. This second theory argues that emotion or object associations influence colour

preference (Palmer & Schloss, 2010). This section will summarise the main aspects of these two theories.

*4.2.2.1 Cone-contrast components as predictors of colour preference:*

Hurlbert and Ling (2007) observed that underlying the individual variations in hue preference in young adult populations was a regularity which was captured by a small number of components, the first two of which closely corresponded to the L-M ("red-green") and S-(L+M) (blue-yellow) cone-opponent contrast components of the stimuli. Running a regression model to explain the variation in colour preference for hue variations they found that the cone-contrast components explained 70% of the variation in colour preference in a cross-cultural population of 208 participants. They also found differences in the relative weightings of the components, with male preferences predicted by more negative weights along the "red-green" axis (greener hues) compared with females, and both sexes preferring positive weights along the "blue-yellow" axis (bluer hues) (see section 4.2.1.4 for discussion of sex differences). The authors speculated that the sex difference may relate to the differing uses and relevance of colours in the sex-specific roles in a hunter-gatherer society with females' preference for redder colours possibly aiding the identification of ripe berries or skin colour changes. Some support for the embeddedness of this encoding comes from an infant study, where the cone-contrasts predicted 40% of the variation in the infants' looking time towards preferred colours (Franklin, Bevis, et al., 2010). Whilst there is a high amount of variance explained when stimuli vary only in hue, this amount noticeably drops to around 40% when lightness and saturation are also included in the stimuli set (Ling & Hurlbert, 2009; Palmer & Schloss, 2010). The logical extension of the theory is that to some extent colour preference would be universal. Although systematic tests of hue preference alone (without variations in lightness and saturation) have not been performed in other cultures, some studies suggest that other encoding factors may better predict preference variations in other cultures. For example, Taylor and colleagues (2013) assessed colour preference in the Himba tribe using a stimuli sets varying in hue, lightness and saturation, and found that although only 22% of the variance was explained by cone-contrasts alone, and only the "blue-yellow" axis predicted colour preference in males only, the male colour preference curve showed little dependence on hue at all, and instead was predicted largely by the chroma and lightness of the stimuli (57% of variance in a separate regression). The data suggest that the preference measure in the Himba people represents a measure of sensory impact than affective response to colour per se (see Hurlbert and Owen 2015 for discussion). As found in other studies (Ling & Hurlbert, 2009; Palmer & Schloss, 2010; Schloss & Palmer, 2009) less variance (21%) than for a hue-varying-only set was explained by the cone-contrast model in the English adult sample in this study with the "red-green" axis a significant predictor for males but not females, and only the "blue-

yellow" axis only a significant predictor in the females. Given the small sample size and varying stimulus set properties, the results cannot be directly compared with earlier work.

Whilst the cone-contrast model explains a large portion of variance in colour preference for hue-varying stimuli sets for a single lightness level, it is not an adequate model for stimuli sets that vary across lightness due to the additional physiological components involved in the sensory encoding of the colours that vary across different lightness levels. Despite this there are some consistencies for hue at different saturation and lightness variations. For example, increased preference for bluish hues is consistent across differing demographic factors (Hurlbert & Ling, 2007; Hurlbert & Owen, 2015; Schloss & Palmer, 2010). However, for certain hues there is a noticeable change that comes with changes in saturation or lightness. For example yellowish hues have lower preference ratings for lower lightness saturation levels (Palmer & Schloss, 2010). These changes in colour preference also shift with changes in the categorisation of the colour from yellow to brown, suggesting that cone-contrasts alone may not account for colour preference for hues that have categorical variations across lightness and saturation. The next section will outline an alternative account for colour preference.

#### 4.2.2.2 The Ecological Valence Theory (EVT)

Unlike the cone-contrast model, the Ecological Valence Theory (EVT) proposed by Palmer and Schloss (2010) argues that the relative extent of colour preference depends on the individual's preference for objects that are primarily associated with that colour rather than physiological basis. This colour-object associations are largely ones learned during an individual lifetime. The rationale is that individuals will be more likely to have a positive affect for colour if it is associated with an object that is beneficial to them. For example, the general preference for blue recurrently found in colour preference studies over the past century may be due to its association with clean water or clear skies, while a general dislike for brown might arise from its association with faeces or rotting food. This theory accounts for different modulating factors on colour preference, i.e. variations in colour preference across sex, culture and age vary because these factors may cause differences in typical object-colour associations. Palmer and Schloss (2010) capture this by calculating a weighted affective valence estimate (WAVE) for each coloured stimulus. This process involves participants generating names of objects associated with a colour, and then providing valence ratings for both the objects and the colours. An averaged valence rating can then be calculated for each colour based upon object valence ratings. The WAVE for each colour is then put into a regression model. Palmer and Schloss (2010) found that the WAVE calculated from responses in one set of observers explained 80% of the variance in colour preference measured in another set of observers from the same local community. Over the same set of 24 colour stimuli varying in hue, saturation and lightness, the cone-

contrast model explained 37% of the variance, whereas for the subset of stimuli in which saturation and lightness were nearest to being held constant, the cone-contrast component model predicted 64% of the variance, like the results of Hurlbert and Ling (2007). Further support for this model comes from British and Japanese adult samples (Fushikida et al., 2009; Taylor & Franklin, 2012). Further evidence for the individual malleability of colour preference based on learned associations comes from priming experiments, in which subsequent colour preferences were related to the valence of the preceding priming image (e.g. blood vs. strawberries for red or mould vs. kiwi fruit for green), (Strauss, Schloss, & Palmer, 2013) and the finding that individuals who display a strong association with their college sports teams show an increased colour preference for the colours of the team (Schloss, Poggesi, & Palmer, 2011). The EVT would also explain the association between stereotypically coloured toys for each sex (Jadva et al., 2010; LoBue & DeLoache, 2011). For example, sex specific toys would be associated by children with their corresponding colours. According to the EVT these "toy associated" colours would then be preferred more or less depending on how the child felt about that specific toy. Therefore, since boys will usually prefer to play with toys that are designed for boys they will "*generally*" prefer the colours of these toys.

In examining colour preference in autism and Williams Syndrome, it is important to consider both the contributions of low-level colour encoding mechanisms, given the potential difference in fundamental discrimination abilities along these dimensions, as well as the contribution of colour-object associations predicted by the EVT. As the previous chapter reported, chromatic discrimination is significantly poorer in individuals with autism compared to mental age controls, and it is not known to what extent this will affect preference for colours that are predominantly weighted on this axis. It is also possible that individuals with autism and Williams Syndrome form both strong positive and negative object associations with colours, the influence of which may introduce greater individual variation into preference patterns, despite underlying regularity. For example, a female child's aversion to a green room may partly result from an underlying tendency of females to dislike greenish contrasts but be further exaggerated by the association of that green with a negative object. The next section will outline some of the papers that find colour affected behaviours, e.g. obsessions and aversions, in autism and Williams syndrome.

### 4.2.3 Colour preference in Autism and Williams syndrome

To date there have been no published systematic studies of colour preference in autism or Williams Syndrome. However, for autism, there are published accounts of individual's colour-affected behaviours, in both case studies and other anecdotal and qualitative reports. In these, colour seeking/rewarding behaviour may be inferred as indicating increased preference for a colour, whilst colour aversive behaviour may be inferred as decreased preference for a colour. These accounts of

colour affected behaviour are for high-functioning individuals with autism or parents of children with autism. To quote one example,

"*He could distinguish colours at the age of 2. He shows certain preferences: He likes everything yellow and prefers to draw with a yellow pencil. Some colours disturb him"* (Bogdashina).

Other descriptions of extreme behavioural responses to colour include (but are not limited to) aversion to particularly high contrast colours, emotional responses to certain colours, and fascination with shiny/coloured objects (Bogdashina, 2011; Lawson, 1998; A. K. Ludlow et al., 2014; Williams, 1994). Franklin and colleagues (2014) report a case study of an adolescent with autism who displayed colour obsessive behaviour. Patient J.G. exhibited a strong preference for blue or purple colours, refusing to wear clothes that were not these colours or to travel in cars that were not blue. In this case, J.G.'s colour preferences appeared to strongly influence his behaviour. After the introduction of blue-tinted glasses his general behaviour improved. Although the story of J.G. and other related reports are useful in highlighting the potential impact of colour attachments on behaviour, these studies do not give a coherent analysis of explanation for colour affected behaviour. It is unclear whether or which of the distinct components of colour (hue, saturation, lightness) individually or in combination give rise to these behaviours.

Implicit assumptions about the colour preferences of children with autism are also found in guidelines for the use of colour in environmental settings, laid down by clinicians and/or interior designers. For example, the NICE guideline 142 for adults with autism explicitly advises to "*avoid patterns*" for walls and furnishings and instead to "*use low-arousal colours such as cream"* (NICE, 2012) *p13*. For children with autism, similar advice is given to take care about what colours to use in walls and furnishings (NICE, 2011). Guidelines from interior designers are similar, typically based on consultation with the various stakeholders (e.g. interior designers, teachers, clinicians, parents and the children themselves), and typically concluding that unsaturated colours are preferred by children with autism. For example, one unpublished study, using stimuli from the Natural Colour System, concluded adolescents with autism preferred unsaturated colours (Beevers, 2010). Unfortunately, the methods used in that study are unclear, and therefore the validity of the conclusion is also unclear. Other interior design studies report conflicting recommendations, some advocating warmer hues (e.g. reds) and others cooler hues (e.g. blues) for optimal comfort – presumptively preference matching – in children with autism (Beevers, 2010; Cotton & Geraty, 1984; Gaines & Curry, 2011). Nonetheless the existence of such studies and reports demonstrate the awareness that colour preferences exist and may elicit certain colour-related behaviours. Overall, there seems to be a consensus amongst clinicians and interior designers that individuals with autism will prefer "calm and non-stimulating" colours, but with much ambiguity and little experimental evidence. The NICE guidelines and recommendations from interior designers are also at odds with the design of sensory

based interventions (e.g. sensory rooms) and sensory toys, in which the colours used are *more stimulating*. Therefore, without a direct study of colour preference in individuals with autism, the picture remains unclear.

Similarly, there are no systematic studies of colour preference in individuals with Williams Syndrome, and even fewer clinical guidelines or interior design recommendations. One set of guidelines for school teachers states that colour may be a source of distraction for individuals with Williams Syndrome (Udwin et al.). In addition, the use of muted colours as described above has been recommended as general policy that has been recommended for wider application in special education schools (Beevers, 2010; Cotton & Geraty, 1984; Park, 2009).

### 4.2.4 Study Rationale and Hypothesis

The purpose of this study was to systematically investigate colour preference in children and adolescents with either autism or Williams syndrome relative to typically developing mental age controls. Assessment of colour preference allows various aspects to be addressed. Differences in colour preference may be due to an inability to extract relevant cultural norms for the valence of colours. Given the dissociations in social functioning between autism and Williams syndrome, it is hypothesised that the autism group may show preference differences due to difficulties in abstracting social information, whilst the Williams syndrome group may show typical colour preference patterns. Conversely, differences in colour preferences may also reflect differential object-colour associations. Due to time constraints, it was not possible to directly assess object-colour associations using the Palmer & Schloss (2010) method. Instead, the results reported in this chapter are linked to responses in bespoke parental questionnaire about colour affected behaviours (see Chapter 6). Colour preference assessment also allows for links between sensory processing and higher behavioural functions to be established, where the relative preference for certain colours may depend on the chromatic discrimination along colour axes.

## 4.3 Methods

### 4.3.1 Participants

In total, eighty-eight participants took part in the study: twenty-six children with Williams Syndrome, eighteen with autism, and 44 typically developing individuals who were individually matched on RCPM performance to each of the test group individuals. Due to the documented sex differences in colour preference patterns, participants were also matched on sex. There were no significant differences between autism, $t$ (34) = 0.355, $p$ = 0.725, *Cohen's d* = 0.118, or Williams Syndrome, $t$ (50) = 0.31, $p$ = 0.976, *Cohen's d* = 0.08, groups relative to their respective TD control groups on RCPM scores. The participant demographics for the studies reported in this chapter can be seen in table 4-1.

*Table 4-2- Participant demographics for Chapter 4. Chronological age is reported in years. Scores on the RCPM are raw scores. VIQ is the standard score on either the WISC-III, WISC-IV or WPPSI. Standard deviations are reported in brackets.*

| Group | Chronological Age (Years) | Females | Males | RCPM | VIQ | NVIQ |
|---|---|---|---|---|---|---|
| Autism | 13.43 | 6 | 12 | 27.06 | 77 | 83.93 |
| TD (Autism control) | 7.54 | 6 | 12 | 26.28 | 118.22 | 114 |
| Williams Syndrome | 12.81 | 13 | 13 | 15.69 | 70.68 | 54.5 |
| TD (Williams Syndrome control) | 5.57 | 13 | 13 | 15.65 | 105.28 | 95.38 |

### 4.3.2 Apparatus

Colour stimuli were displayed using apparatus and set up described in Chapter 4. One of two different experimental setups were used to display the experiment. Twelve of TD (autism control) and ten of autism participants were assessed using the 8bit setup. Five of TD (autism control) and seven of autism participants were assessed using the 10bit setup. One Williams Syndrome and one TD (Williams Syndrome control) participant were assessed using the 8bit setup. The remaining Williams Syndrome participants and matching TD (Williams Syndrome control) participants were assessed using the 10bit setup. experimental programme (the Colour Preference test) was used for both setups, written in Matlab (v7.6.0, 2012b, The MathWorks, 2008, 2012), with graphics display functions from Psychtoolbox (Brainard, 1997) and colorimetric conversion functions from *kccv* (a set of Matlab routines based on standard formulae (Wolf, 2011) tailored for 8- or 10-bit displays appropriately); the 10-bit display used and the NVIDIA QUADRO performance drivers. Spectral emission properties of both screens were characterised using a PR-650 spectroradiometer and colorimetric calibration tables were checked regularly using a Minolta CS-100 chromameter and updated when necessary to ensure colorimetric accuracy of the displayed stimuli.

### 4.3.3 Stimuli

On each trial, two coloured rectangles (visual angle = 2.38°) were presented on an achromatic background (CIE 1931 coordinates: x=0.31, y=0.32, Y=50 for the 8-bit display; x=0.3127, y=0.329, Y=60 for the 10-bit display). The rectangle colours were drawn from a total of nineteen colours which varied systematically within HSL space (see Chapter 2 section 2.6.2). Seven of these colours varied only in hue, at a constant midlevel saturation and midlevel lightness. Four of these hues (approximately matching blue, green, red and brown) were combined with either high or low

lightness (at mid saturation) or high saturation (at mid lightness) to create three additional groups of colours. See table 4-2 for the complete set of HSL values and figure 4-1 for illustrative mock ups of stimuli. The HSL values of the Colour Checker Chart samples can be found in Appendix 2.

*Table 4-3 - The colour coordinates of colour preference stimuli. The* H*-value represents the hue angle,* S*-value the saturation level and* L*-value is the lightness level as defined within HSL colour space. The manipulation column shows the type of variation that the colour stimuli is.*

| | | 8bit | | | 10bit | | |
|---|---|---|---|---|---|---|---|
| | Colour Group | H | S | L | H | S | L |
| 1 | Hue-varying | 0.783 | 0.5 | 80 | 0.3 | 0.5 | 80 |
| 2 | Hue-varying | 2.543 | 0.5 | 80 | 1.2 | 0.5 | 80 |
| 3 | Hue-varying | 3.734 | 0.5 | 80 | 2.0 | 0.5 | 80 |
| 4 | Hue-varying | 4.01 | 0.5 | 80 | 2.76 | 0.5 | 80 |
| 5 | Hue-varying | 5.141 | 0.5 | 80 | 4.0 | 0.5 | 80 |
| 6 | Hue-varying | 5.997 | 0.5 | 80 | 5.0 | 0.5 | 80 |
| 7 | Hue-varying | 6.117 | 0.5 | 80 | 5.8 | 0.5 | 80 |
| 8 | Low Lightness | 0.73 | 0.5 | 50 | 1.2 | 0.5 | 80 |
| 9 | Low Lightness | 2.5 | 0.5 | 50 | 2.76 | 0.5 | 80 |
| 10 | Low Lightness | 3.807 | 0.5 | 50 | 4.0 | 0.5 | 80 |
| 11 | Low Lightness | 6.243 | 0.5 | 50 | 5.8 | 0.5 | 80 |
| 12 | High Lightness | 0.753 | 0.5 | 110 | 1.2 | 0.5 | 110 |
| 13 | High Lightness | 2.5 | 0.5 | 110 | 2.76 | 0.5 | 110 |
| 14 | High Lightness | 3.589 | 0.5 | 110 | 4.0 | 0.5 | 110 |
| 15 | High Lightness | 6.147 | 0.5 | 110 | 5.8 | 0.5 | 110 |
| 16 | High Saturation | 0.73 | 1 | 80 | 1.2 | 0.9 | 80 |
| 17 | High Saturation | 2.5 | 0.8 | 80 | 2.76 | 0.9 | 80 |
| 18 | High Saturation | 3.807 | 0.9 | 80 | 4.0 | 0.9 | 80 |
| 19 | High Saturation | 6.243 | 1 | 80 | 5.8 | 0.8 | 80 |

For participants who completed the 10bit version, an additional colour was put into the stimulus set. This additional stimulus was selected by the participant from the Macbeth Colour Checker Chart (McCamy, Marcus, & Davidson, 1976). This colour was selected either during the naming experiment (see Chapter 5) or prior to the start of the main colour preference experiment.

*Figure 4-1 – Mock ups of Colour Preference stimuli.* a) Hue variants, with saturation and lightness kept constant. b) High Saturation Variants of four hues. The low, mid and high lightness variants are shown by c), d) and e) respectively.

### 4.3.4 Design

The method of constant stimuli was used to present the stimulus to participants. A two-alternative forced choice task was used, in which the participant must choose the colour they like the most from two presented colours on each trial. Participants were shown all possible pairs once each from the fixed stimulus set described above. This meant that there 171 trials in the 8bit version (19 experimental colours), and 190 trials for the 10bit version (19 experimental colours + the individual selected favourite colour).

### 4.3.5 Data Analysis

Preference ratings were calculated for each colour as the proportion of trials in which it was chosen out of the total number of trials in which it was shown. This gave a rating for each individual colour between 0 and 1, with higher values indicating higher preference for the colour. Individual preference ratings were calculated for each colour over all pairwise comparisons for the entire set of 19 (20) colours, yielding the "total" preference curve.

In addition, to assess the dependence of colour preference on hue, lightness and saturation separately, hue preference curves were calculated as within-set proportions for the set of 7 colours at mid-lightness, mid-saturation which varied only in hue, i.e. as the preference ratings across all pairwise comparisons within that set only. To assess lightness preference, preference curves were calculated as within-set proportions for the 4 hues shown at each of three lightnesses (low, mid and high) (12 colours total), the lightness-varying set. These curves yield lightness preference as a function of hue. Likewise, saturation preference curves, as a function of hue, were calculated as within-set proportions for the 4 hues at each of two saturation levels (mid, high) (8 colours total), the saturation-varying set. Average preferences at each lightness or saturation level were calculated by averaging over hues in the relevant lightness/saturation preference curves.

Total preference strength was measured as the standard deviation of the total preference curve. A larger standard deviation indicates greater variability in preferences between colours. Hue preference strength was measured as the standard deviation of the hue preference curve. Lightness preference strength was calculated as the difference in the z-scores for high vs low (or mid) lightness within the lightness-varying set; similarly, saturation preference strength was calculated as the difference in the z-scores for high vs mid saturations within the saturation-varying set. Significant preferences (both like and dislike) for individual colours can also be calculated based on one sample t-tests against a test-value of 0.5. This value was chosen as it indicates where there is no preference for this colour. Preference scores significantly above this test-value indicate the colour that is liked, whilst scores that are significantly below this value indicate that the colour is not liked.

For the analysis of hue preference dependence on physiological colour-encoding components, the colour coordinates were converted to cone-opponent contrast values ("red-green" and "blue-yellow") (Eskew et al., 1999). (see Chapter 2 section 2.6.1).

### 4.3.6 Procedure

Participants completed the colour preference task as part of a wider battery of tasks (see Chapter 2, section 2.8 for more details on general testing protocol). Participants completing the 10bit version were initially shown the Macbeth Colour Checker Chart (McCamy et al., 1976) and were asked to select the colour that they liked the most. This selected "favourite" colour was then added to the main stimulus set, from which all pairwise comparisons were shown. Otherwise, participants on both setups performed the colour preference test in the same way. Prior to the start of the experiment, participants were informed to select the colour that "they liked the most" of the pair shown on each trial. After a 10 second countdown, participants were presented with the first trial. In between trials there was an inter-stimulus-interval of 500ms. The position of each stimulus was randomly assigned to top or bottom of the display on each trial. The total duration of the test was approximately 5-10 minutes for both the 8-bit (171 trials) and 10-bit (190 trials) setups. Where participants were unable to use the mouse (3 Williams syndrome, 2 autism and 4 TD participants), the experimenter moved the mouse and selected colours under direction from the participant.

## 4.4 Results

### 4.4.1 Hue Variants

Colour preferences for the seven colours for mid lightness and mid saturation levels. These can be seen in figures 4-2 and 4-3. Looking qualitatively at the preference curves, it can be seen than for all groups there are clear peaks and troughs in the preference curve. For all groups, males preferred blue and green hues, whilst pink and purples hues were relatively disliked. For females, the opposite

pattern was observed, where generally pink and purple hues were preferred and blue and green hues were relatively disliked. To further probe this a repeated measures ANOVA was conducted with Hue (7 levels: Hue only variants) and between subjects of group (TD and Williams Syndrome or autism) and sex (male and female).

In the autism analysis, there was a main effect of hue, $F$ (6, 288) = 8, $p < 0.001$. Blue, green and red hue variants were preferred over brownish hue variants. There was also a significant interaction between Hue and Sex, $F$ (6, 288) = 10.17, $p < 0.001$ (see figure 4-2 for colour preference plots for the autism and TD groups). The main effect of group ($p = 0.92$), the interaction between Hue and Group ($p = 0.13$) and the three-way interaction between Hue, Sex and Group ($p = 0.14$) were not significant. For the variation in preference across different hues there were no significant differences in the between the autism and TD groups, lowest $p = 0.4$. For detailed breakdown of hue comparisons see Appendix 3.

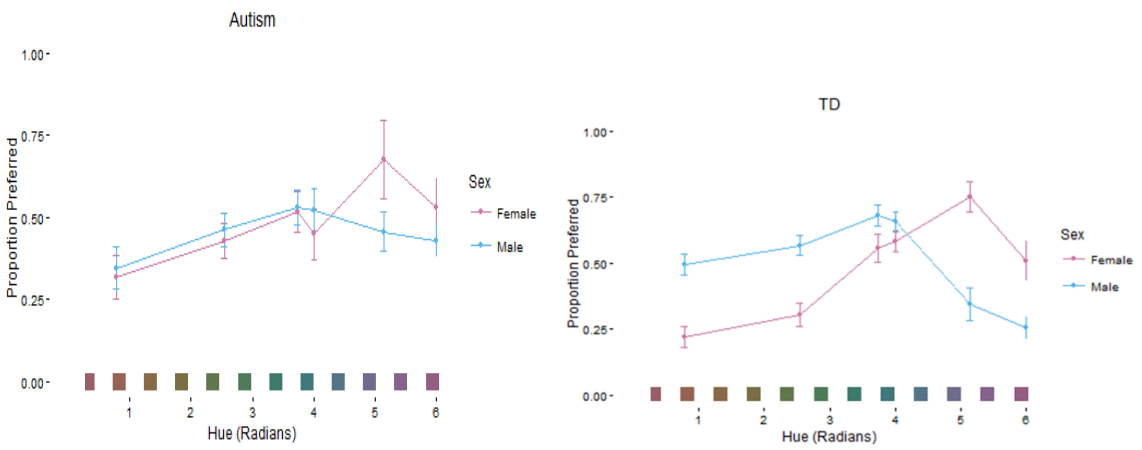



*Figure 4-2 - Colour preference across Hue variants split by male and female participants for the TD and autism groups.* Females are shown via the pink line, whilst males are displayed by the blue line

In the Williams Syndrome analysis. there was a main effect of hue, $F$ (6, 288) = 3.17, $p < 0.05$. This main effect of hue was driven by increased preference for blue and green hues were significantly preferred over brownish hues (see figure 4-3). There was a significant interaction between Hue and Sex, $F$ (6, 288) = 10.89, $p < 0.001$. Females in both groups (although increased in the TD group) preferred blue, red and pink hues over brownish hues, whilst males preferred green and blue hues over brownish hues (see Figure 4-3). There was a main effect of group, $F$ (1, 48) = 7.67, $p < 0.01$. This was driven by relatively higher preference across the 7 hues in the WS group. Further inspection of the hue preference plots (figure 4-3) reveals that this is because the Williams Syndrome group do not show as extreme sex differences in hue preference compared to the TD group. The interaction between Hue and group also approached significance, $F$ (6, 288) = 2.58, $p = 0.06$. Tentative

exploratory post-hoc t-tests found that there were no colours that significantly differed in their preference between groups (lowest $p = 0.084$). The three-way interaction between Hue, group and sex was not significant ($p = 0.09$). Post-hoc results for the main effect of hue can be found in appendix 3.

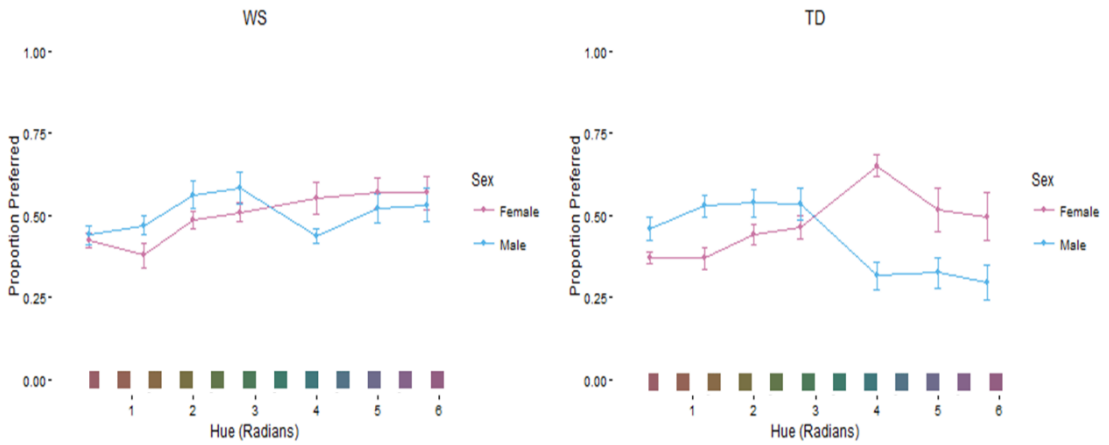



*Figure 4-3 – Colour preference across Hue variants split by male and female participants for the TD and Williams Syndrome groups.*

### 4.4.2 Saturation Variants

To measure the effect of saturation on colour preference, four mid lightness and mid-saturation hues were averaged and compared with the averaged preference of the four corresponding hues at a high saturation (see figure 4-4 and 4-5). A repeated measures ANOVA with saturation (two levels: Medium and High) and hue (four levels: Brown, Green, Blue and Red) as a within subject factor and between subjects factor of group (TD and Williams Syndrome or autism) and sex (male and female).

In the autism analysis, there was a significant main effect of saturation, $F(1, 30) = 43.24$, $p < 0.001$, where highly saturated colours were preferred more than medium saturation colours (see figure 4-4). There was also a main effect of hue, $F(3, 90) = 10.81$, $p < 0.001$. This was driven by increased preference for blue hues. There was also a significant interaction between hue and sex, $F(3, 90) = 7.01$, $p < 0.001$, in which females preferred reddish hues more than males. There were no significant group effects ($p = 0.11$) or significant interactions between saturation and sex or group (lowest $p = 0.07$).

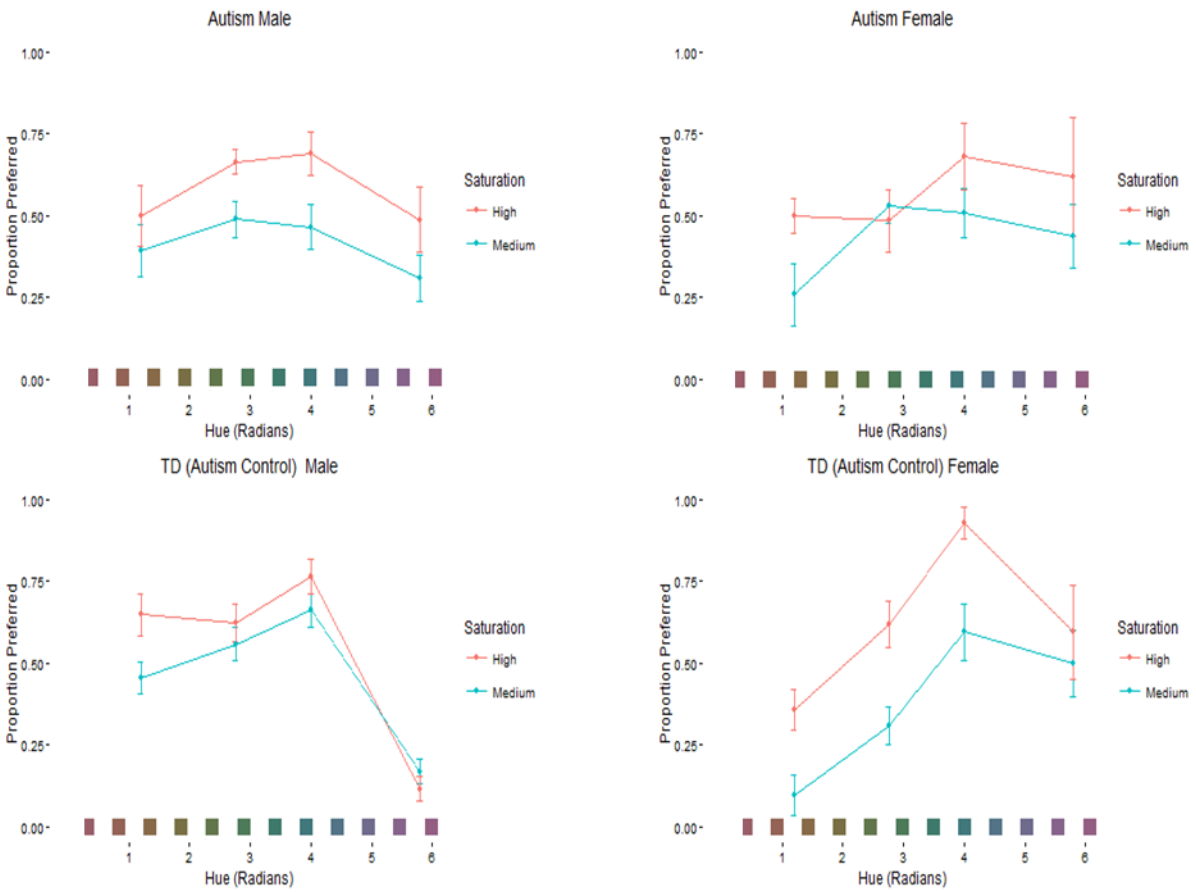



*Figure 4-4 - The preference for different saturations variants across both autism and TD groups.* This shows saturation preferences for male and female participants in both the autism and TD groups.

For the Williams Syndrome analysis (see figure 4-5) the high saturation colours were significantly preferred to mid-level saturation colours, $F$ (1, 47) = 29.048, $p < 0.001$. There was also a significant interaction between Saturation and Sex, $F$ (1, 48) = 9.47, $p < 0.005$. Post hoc t-tests revealed that this driven by an increased preference for highly saturated colours by females compared to males, $t$ (50) = 2.74, $p < 0.01$. There was also a main effect of hue, $F$ (1, 48) = 5.339, $p < 0.005$, and a significant hue by sex interaction, $F$ (1, 141) = 8.143, $p < 0.005$. Both groups preferred blue and red hues most, however females preferred red hues more than males. There were no significant group effects ($p$ = 0.322) or group interactions with saturation level, $p$ = 0.18, or sex and saturation level, $p$ = 0.11.

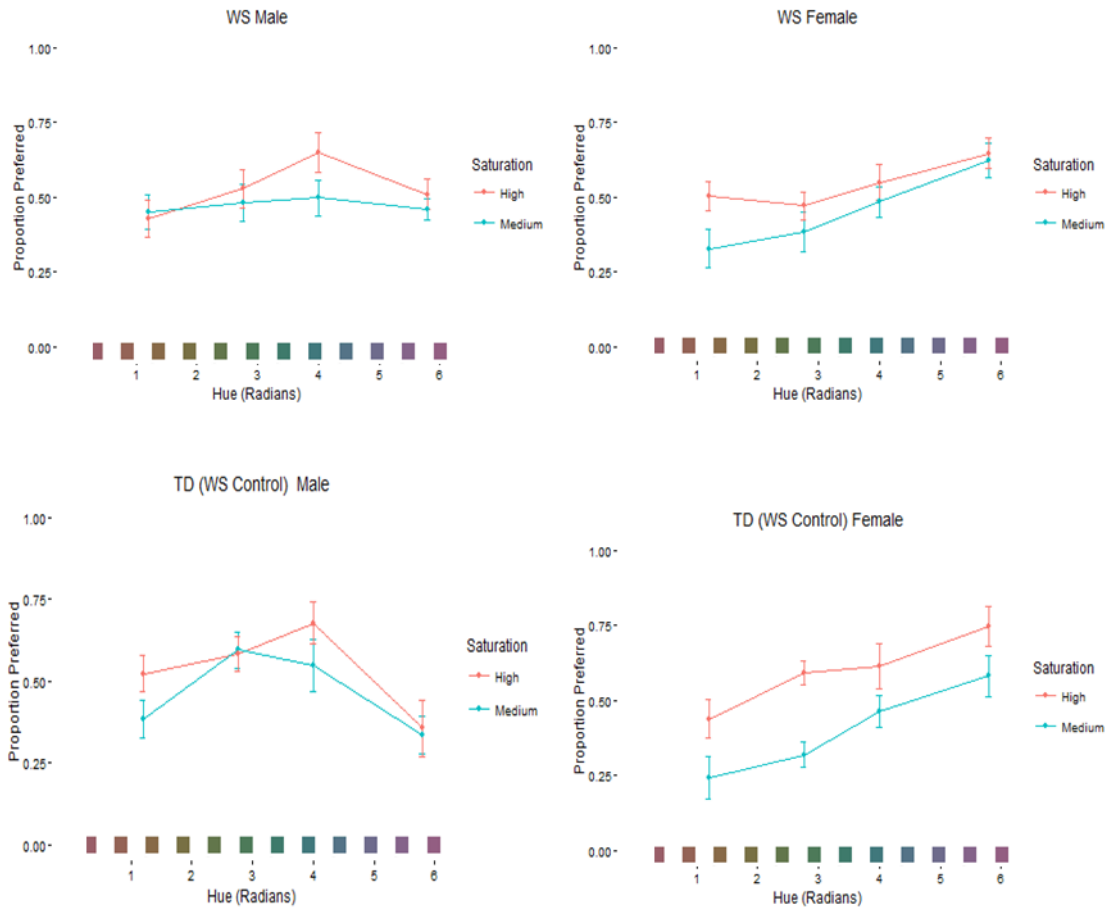



*Figure 4-5 The preference for different saturations variants across both Williams syndrome and TD groups.* This shows saturation preferences for male and female participants in both the Williams syndrome and TD groups.

### 4.4.3 Lightness Variants

Colour preference across different lightness variations was assessed in a similar way to the saturation variants. Where four mid lightness and mid-saturation hues were averaged together and compared with averaged preference of the four corresponding hues for low and high lightness (see figure 4-6 and 4-7). A repeated measures ANOVA with lightness as a within subject factor (three levels: Low, Medium and High) and between subject factors of group (TD and Williams Syndrome or autism) and sex (male and female).

For the Williams Syndrome analysis, there was a main effect of lightness, $F$ (1, 96) = 13.36, $p < 0.001$. This was driven by significantly higher preference for high lightness colours compared to both low lightness colours, $t$ (51) = 3.83, $p < 0.001$, and mid lightness colours, $t$ (51) = 3.83, $p < 0.001$. The group main effect was not significant ($p = 0.322$). There was also a significant interaction between Sex and Lightness, $F$ (1, 96) = 4.9, $p < 0.05$. Post hoc t-tests revealed that this was driven by significant increased preference of high lightness colours compared to both low lightness, $t$ (25) = 5.08, $p <$

0.001, and mid lightness, $t$ (25) = 5.94, $p < 0.001$, colours in females. There were no significant differences for males between any of the lightness variables, lowest $p = 0.3$.

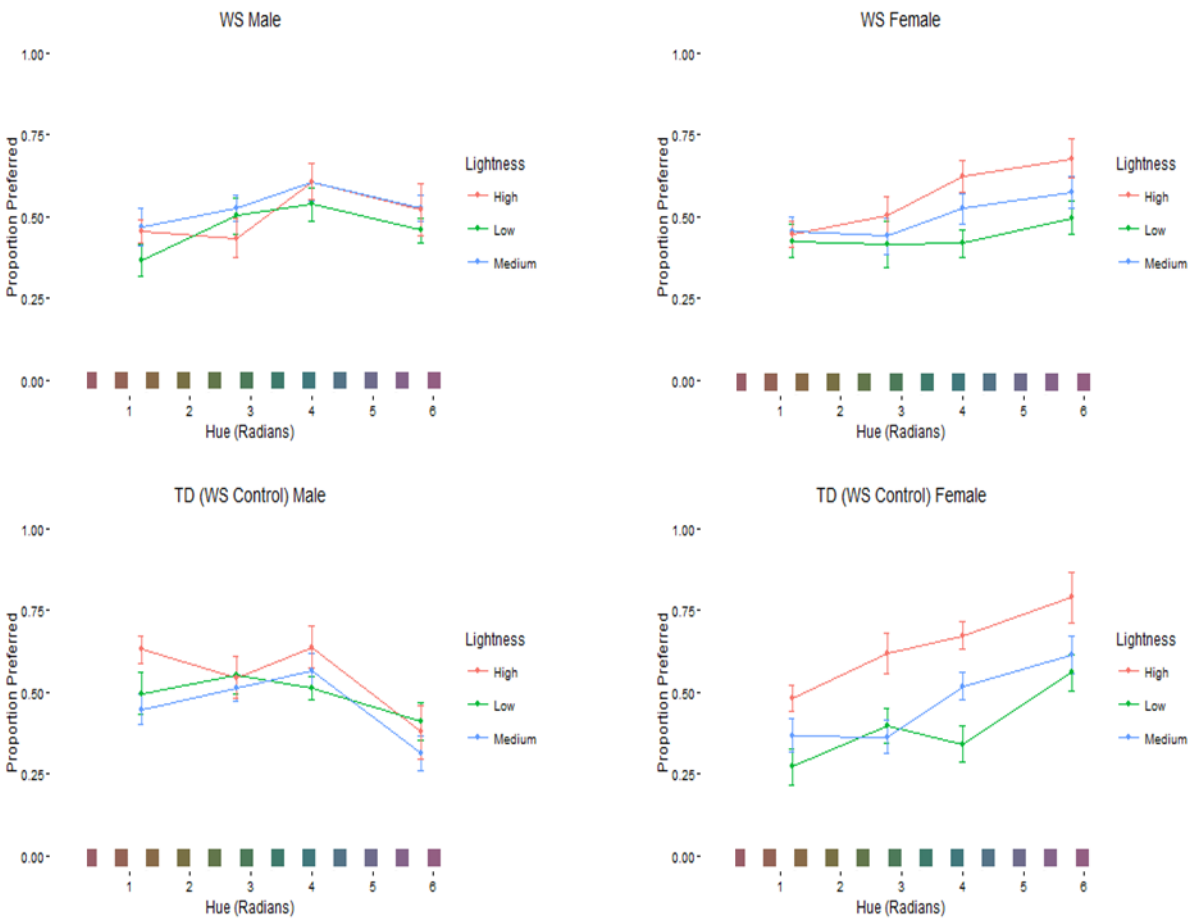



*Figure 4-6 – Colour preference for different lightness variants across both Williams Syndrome and TD groups.*

In the autism analysis (see figure 4-7) there was a significant main effect of lightness, $F$ (2, 60) = 4.61, $p < 0.05$, where high lightness variants were preferred more than medium and low lightness variants. There was also sex by lightness interaction, $F$ (2, 60) = 5.673, $p < 0.05$. This interaction was driven by opposite patterns of lightness preference between sexes, where males preferred low/medium relative to high lightness variants whilst females preferred high to low lightness. The main effect of hue, $F$ (6, 180) = 10.332, $p < 0.001$, and the interaction between hue and sex, $F$ (6, 180) = 5.37, $p < 0.005$, following the same patterns as the saturation and hue analysis. The interaction between hue and lightness was also significant, $F$ (6, 180) = 3.613, $p < 0.01$. This was driven by indifference to hue at low lightness and an increased preference for blue hues at medium and high lightness. There was no main effect of group (p = 0.204) and neither were any other interactions were significant (lowest $p$ = 0.214).

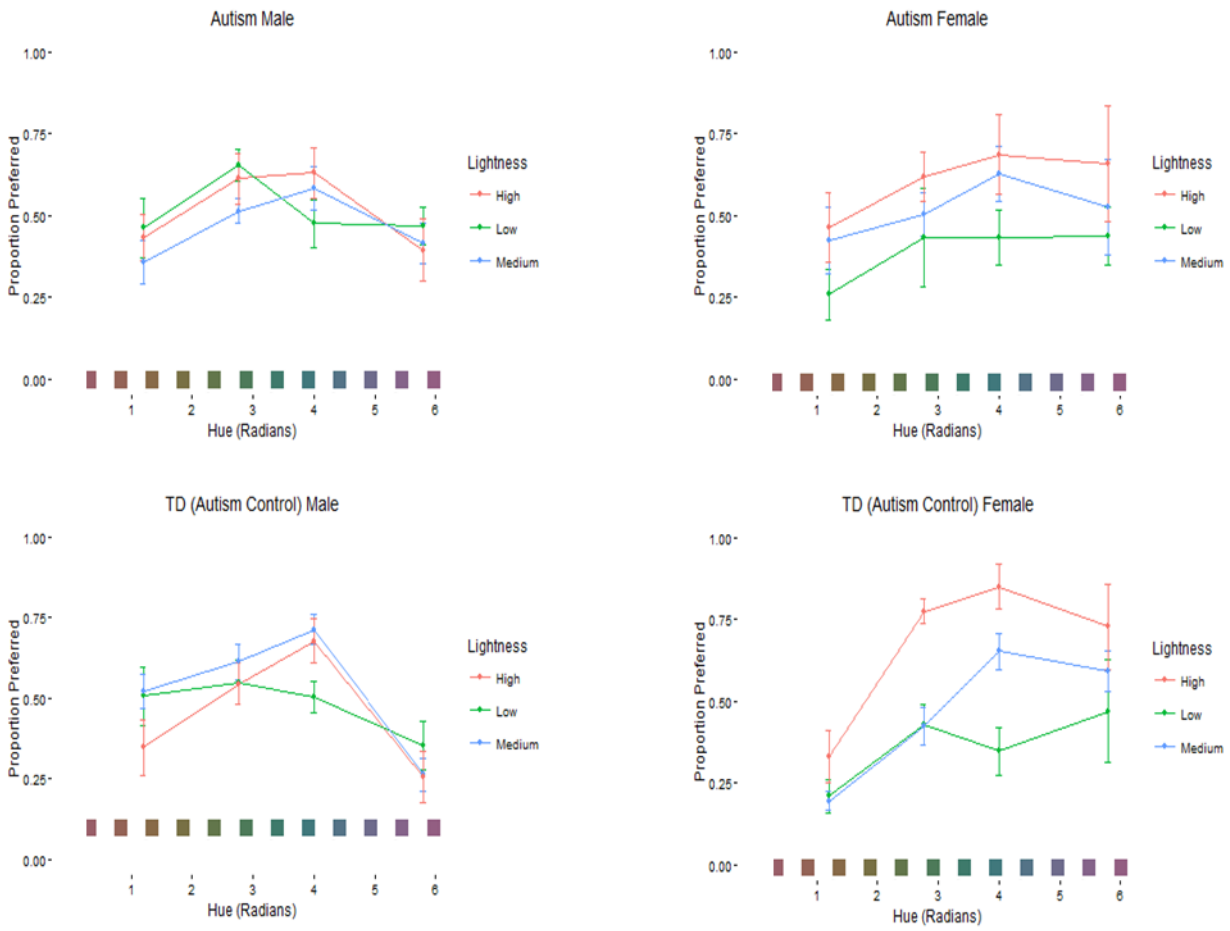



*Figure 4-7 – Colour preference for different lightness variants across both Williams Syndrome and TD groups.*

### 4.4.4 Strength of colour preference

Each participant's individual colour preference curves were inspected and revealed substantial inter-individual variation in both the strength of preference for colours or variants and the overall strength of preference for favourite colours (see Chapter 7 and Appendix 6 for illustrative examples). This variation is quantified by calculating the standard deviation of the preference curve for hue variants (strength of colour preference across hue) and for the total preference curve (overall colour preference strength).

For the autism analysis (see figure 4-8) there was no significant difference between the autism and TD groups in the standard deviation for either total preference curve, $t(32) = 1.206$, $p = 0.236$, or hue preference curve, $t(32) = 0.311$, $p = 0.758$. There was also no difference when analysing colour preference strength by sex (lowest $p = 0.158$).

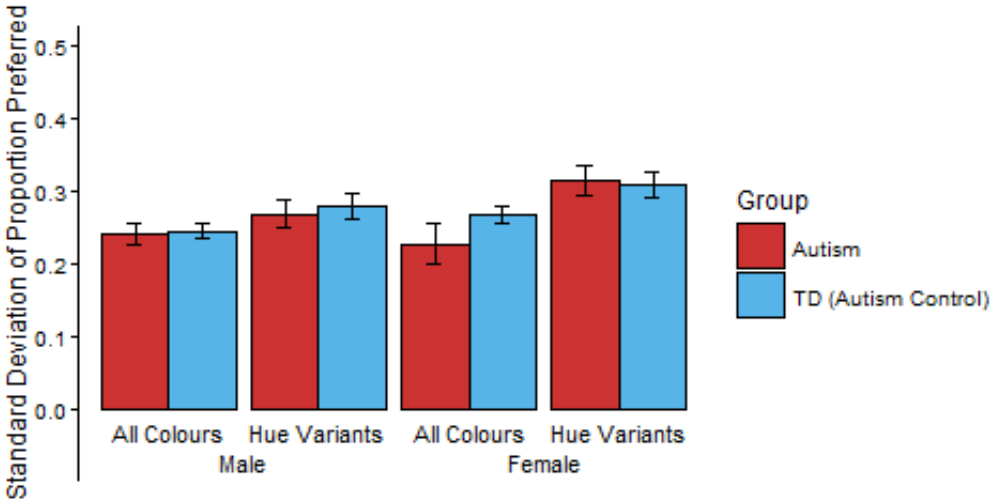



*Figure 4-8 – Strength of colour preference (as measured by standard deviation of proportion preferred) for the autism and TD groups.* Strength of colour preference is shown for all colours and hue variants only. Groups are also divided by sex.

In the Williams Syndrome group (see Figure 4-9) there was a near significant difference in the preference strength for the hue preference curve between the Williams Syndrome and TD group, $t$ (50) = 1.93, $p < 0.06$, but not for the total preference curve, t (50) = 0.354, p = 0.725. The mean standard deviation for the hue preference curve was lower for the Williams Syndrome group, indicating that Williams Syndrome individuals showed less variation in hue preference compared to the TD group. Analysing the curves separately for each sex indicates that the strength difference is significant only in the hue preference strength, and only in the females. These findings begin to quantify the noticeable flatness of the hue preference curves in the Williams Syndrome group seen in Figure 4-3. Individual inspection of colour preference curves for the showed marked inter-individual variation, with some curves showing pronounced dependencies on hue or no discernible colour preference pattern. Possible reasons for this will be covered in more detail in Chapter 7.

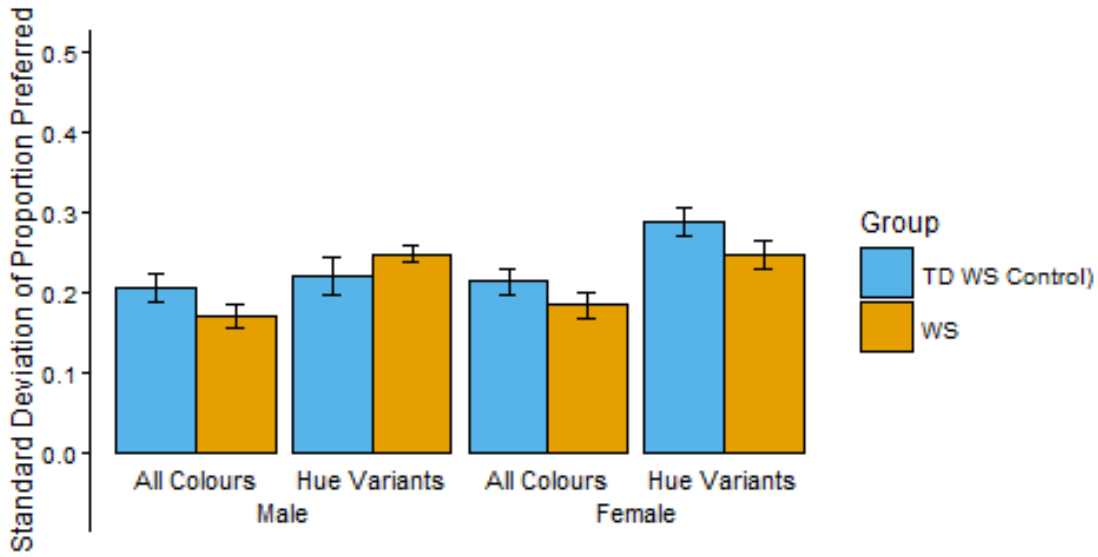



*Figure 4-9 - Strength of colour preference (as measured by standard deviation of proportion preferred) for the Williams syndrome and TD groups.* Strength of colour preference is shown for all colours and hue variants only. Groups are also divided by sex.

#### *4.4.4.1 Variation of Colour Preference across Saturation and Lightness Variants*

This section looks at the variation in preferences for lightness and saturation variants. Difference scores were calculated between each hue variation for either saturation or lightness variation. This was done for the average for each saturation and lightness variant. Additional difference scores were calculated for the saturation variants this was between the high and medium saturation variants and for the lightness variants the difference between the high and low lightness variants were used.

#### 4.4.4.2 Saturation Variants

Difference scores between the averaged high and medium saturation variants. Independent sample t-tests with group as between subject's factor found that there was no difference between the Williams Syndrome and TD groups, either at the group level or when split by sex (lowest $p = 0.18$). For the individual colour analysis, there was no significant main effect of colour or significant interactions (lowest $p = 0.25$).

There was also no difference at the group level between the autism and TD groups, $t$ (33) = 0.285, $p$ = 0.78. When split by sex there was a trend towards a significant effect between males in the autism and TD group, $t$ (21) = 1.98, $p = 0.06$, where the males with autism had showed a greater difference between their preference of highly saturated colours compared to those of medium saturation. There was no significant difference between the female autism and TD groups, $t$ (10) = 1.59, $p$ = 0.143. For each individual colour, there was no significant main effect of colour or group/sex or any significant interaction (lowest $p = 0.338$).

#### 4.4.4.3 Lightness Variants

Difference scores between the averaged high and low lightness variants. Independent sample t-tests with group as between subject factor found that there was no difference between the Williams Syndrome and TD groups, either at the group level or when split by sex (lowest $p = 0.22$). When comparing difference scores for all hue variants there was a main effect of hue, $F$ (3, 144) = 4.671, $p < 0.05$. Post hoc paired sample T-tests showed that this driven by an increased preference for brownish and greenish high lightness variants compared to the reddish variants (lowest $t = 3.047$, highest $p < 0.05$). There was also a significant interaction between sex and hue, $F$ (3, 144) = 8.188, $p < 0.001$. This interaction was driven by larger preference for lighter bluish variant by females, $t$ (50) = 4.13, $p < 0.001$. No other interactions were significant (lowest $p = 0.104$).

In the autism analysis for the average across all hue variants there were no differences either at the group level or when split by sex for each (lowest $p = 0.65$). When considering the comparison between hue variants, there was a main effect of hue, $F$ (3, 96) = 4.968, $p < 0.005$. Post hoc paired sample T-tests revealed that the bluish high lightness variant was preferred significantly more than the all other variants (lowest $t = 2.819$, highest $p < 0.05$). There was no significant main effect of group or sex or significant interactions (lowest $p = 0.114$).

### 4.4.5 Preferences of Individual colours

Individuals who completed the 10-bit version of the colour preference task also selected their favourite colour from the Macbeth Colour Checker Chart (see Figure 4-10). This was done for all participants in the Williams Syndrome group and their control group. There was no significant difference between the Williams Syndrome and TD groups in the proportion that participants selected their chosen favourite colour, $t$ (49) = 0.339, $p = 0.74$. One sample t-tests revealed that the selected favourite colours were significantly preferred in both the Williams Syndrome, $t$ (24) = 6.06, $p < 0.001$, and TD groups, $t$ (25) = 4.67, $p < 0.001$. Participants in the autism group predominately completed the 8-bit version, nonetheless a similar preliminary analysis was conducted on the subgroup of participants who completed the 10-bit version of the colour preference task. There was no significant difference between the autism and TD participants in the proportion that participants selected their chosen favourite colour, $t$ (11) = 1.03, $p = 0.324$. One sample t-tests also found that the chosen favourite colour was significantly preferred in the autism sample who completed the 10-bit versions of the task, $t$ (5) = 5.62, $p < 0.005$, and TD groups, $t$ (6) = 10.81, $p < 0.001$.

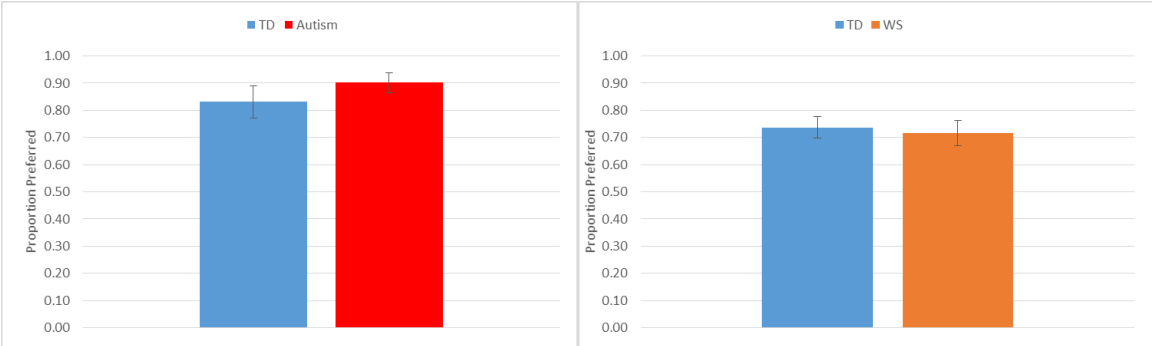



*Figure 4-10– The proportion that the selected "favourite" colour was selected.* Figure 4-10-A – For the autism group. This was only possible for participants who completed the 10-bit version. Figure 4-10-B – Proportion preferred for selected favourite colour the Williams Syndrome group.

### 4.4.6 Cone-opponent contrast components of hue preference

Eskew cone contrast values (Eskew et al., 1999) were calculated for hue variants of the coloured stimuli (separate values were calculated for the 10-bit and 8-bit versions); see Table 4-3 of the 7 colours in the appropriate (8- or 10-bit) hue-varying cone contrast. A regression model was then used for each individual participant to see how much the Eskew cone contrast values (whilst controlling for changes in luminance) of the hue variants explain variation in colour preference. This linear regression gave a coefficient weight for each cone-opponent axis, as well the amount of variance in hue preference that can be explained by colour encoding at the level of the cone-opponent axes (see Chapter 1 section 2.1 for more detail on colour encoding at the neuronal level and Chapter 2 Section 2.6.1 for discussion of Eskew Colour space). A positive coefficient for the "Red-Green" axis reflects a preference for colours with "reddish" contrasts against the background, whilst a negative coefficient reflects a preference for "greenish" contrasts. For the Blue-Yellow axis, a positive coefficient indicates a preference for "bluish" contrasts; a negative coefficient suggests a preference for "yellowish" contrasts. The more a coefficient deviates from 0 the more strongly the hue preference is influenced by that colour axis.

*Table 4-4 - Cone contrasts values for the 8bit and 10bit setups.*

| Colour Stimuli | 10bit Setup | | | 8bit Setup | | |
|---|---|---|---|---|---|---|
| | LUM | RG | BY | LUM | RG | BY |
| Hue Variant 1 | -0.57 | 0.03 | -0.15 | -0.57 | 0.04 | -0.11 |
| Hue Variant 2 | -0.57 | -0.04 | -0.09 | -0.57 | -0.04 | -0.06 |
| Hue Variant 3 | -0.57 | -0.04 | 0.14 | -0.57 | -0.05 | 0.10 |
| Hue Variant 4 | -0.57 | -0.03 | 0.18 | -0.57 | -0.04 | 0.14 |
| Hue Variant 5 | -0.57 | -0.02 | 0.19 | -0.57 | 0.02 | 0.14 |
| Hue Variant 6 | -0.57 | -0.05 | 0.03 | -0.57 | 0.05 | 0.02 |

| | | | | | | |
|---|---|---|---|---|---|---|
| Hue Variant 7 | -0.57 | -0.05 | 0.01 | -0.57 | 0.06 | 0.01 |

*4.4.6.1 Cone-contrast component weights*

4.4.6.1.1 Autism

As expected from previous findings (Hurlbert & Ling, 2007; Ling & Hurlbert, 2011) there were significant differences between males and females for coefficients on both Red-Green, $t$ (34) = 3.968, $p < 0.001$, and the Blue-Yellow, $t$ (34) = 4.641, $p < 0.001$, colour axes, shown in independent sample t-tests with sex as a between-subjects factor over all participants. Males were more likely to weight the R-G axis negatively (preferring greenish contrast hues), whilst females were more likely to prefer hues with a reddish contrast. For the Blue-Yellow axis, females were more likely to prefer colours with a bluish contrast, whilst males did not show this pattern. The one sample t-tests against zero confirmed this dissociation between sex and colour axis coefficients (see Figure 4-11 and Table 4-4): on average, across both groups, female hue preferences are more dependent on blue-yellow contrast, while male hue preferences are more dependent on red-green contrasts.

Due to the sex differences in cone-contrast weights, further analyses within and between autism and TD groups were sex-specific. Bonferroni corrected p-values were used to control for multiple autism comparisons. The within-group sex differences in cone-contrast weights followed the same pattern as the overall sex differences, but this was only significant for the TD group only, shown by independent sample t-tests (see Table 4-4). In addition, there were significant sex differences between in the TD group for both L-M and S cone-contrasts, but there were no significant sex differences for the autism group (lowest $p$ = 0.458).

*Table 4-5 - Results for individual cone contrast weights for autism and TD groups.* A test value of 0 was used to denote the directionality of cone contrast weights

| Group | Sex | L-M Weights | | S-Weights | |
|---|---|---|---|---|---|
| | | t-value | p-value | t-value | p-value |
| Autism | Male | 6.91 | 0.001 | 0.957 | 0.361 |
| Autism | Female | 1.04 | 0.346 | 11.833 | 0.001 |
| TD (Autism Control) | Male | 0.951 | 0.362 | 1.283 | 0.266 |
| TD (Autism Control) | Female | 0.336 | 0.754 | 0.802 | 0.467 |

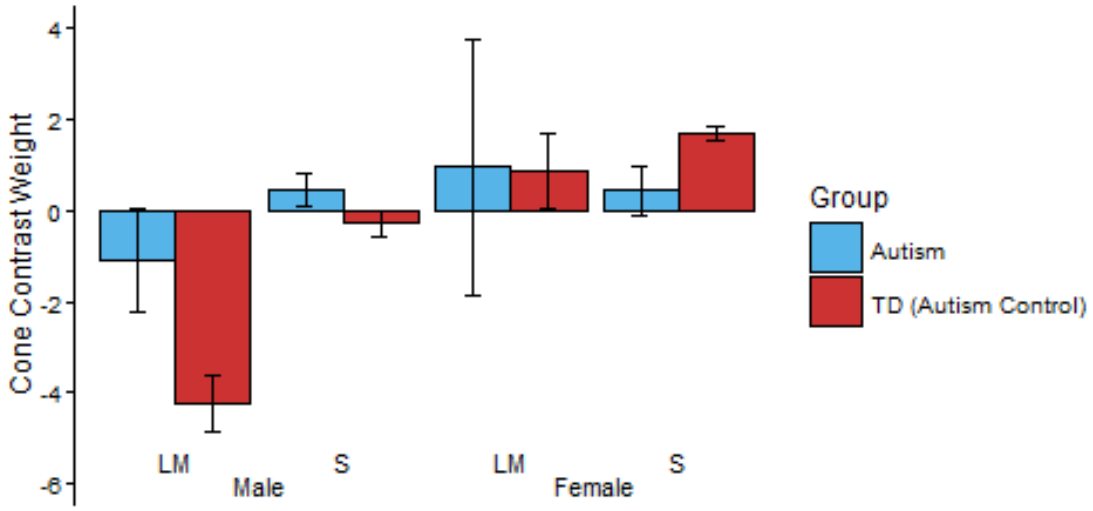



*Figure 4-11 – The cone contrast weights for the autism and TD groups.* Cone contrast weights for each group are further split by sex.

#### 4.4.6.1.2 Williams Syndrome Analysis

The same analysis was conducted between the Williams Syndrome and TD control groups. There were significant differences between males and females for coefficients on both Red-Green, $t$ (50) = 2.617, $p < 0.05$, and the Blue-Yellow, $t$ (50) = 4.493, $p < 0.001$, colour axes, shown in independent sample t-tests with sex as a between-subjects factor over all participants. As in the autism analysis and previous studies, males were more likely to weight the R-G axis negatively (preferring greenish contrast hues), whilst females were more likely to prefer hues with a reddish contrast. For the Blue-Yellow axis, females were more likely to prefer colours with a bluish contrast, whilst males in either group did not show this pattern. The one sample t-tests against zero confirmed this dissociation between sex and colour axis coefficients (see Table 4-5 and Figure 4-12): on average, across both groups, female hue preferences are more dependent on blue-yellow contrast, while male hue preferences are more dependent on red-green contrasts, but this was not a significant deviation from 0 in either the TD or Williams Syndrome male groups. Due to the sex differences in cone-contrast weights, further analyses within and between Williams Syndrome and TD groups were sex-specific. Bonferroni corrected p-values were used to control for multiple comparisons.

The within-group sex differences in cone-contrast weights followed the same pattern as the overall sex differences, for both TD group only, shown by independent sample t-tests. There were no significant sex differences for the Williams Syndrome group.

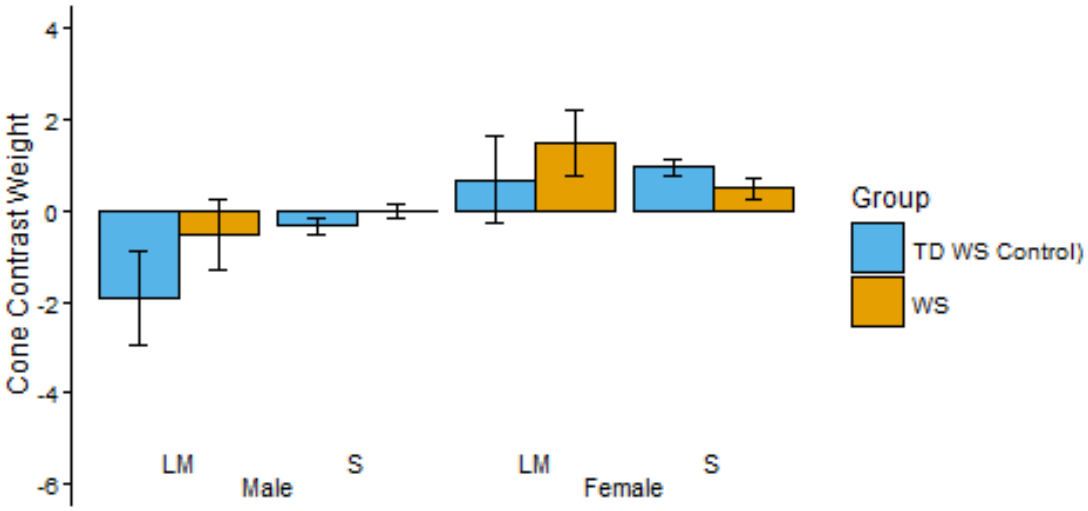



*Figure 4-12 - The cone contrast weights for the Williams Syndrome and TD groups.* Cone contrast weights for each group are further split by sex.

*Table 4-6 – Results for individual cone contrast weights for Williams Syndrome and TD groups.* A test value of 0 was used to denote the directionality of cone contrast weights

| Group | Sex | L-M Weights | | S-Weights | |
|---|---|---|---|---|---|
| | | t-value | p-value | t-value | p-value |
| Williams Syndrome | Male | 0.66 | 0.518 | 0.17 | 0.987 |
| Williams Syndrome | Female | 2.036 | 0.064 | 2.137 | 0.054 |
| TD (Williams Syndrome Control) | Male | 1.868 | 0.086 | 1.922 | 0.079 |
| TD (Williams Syndrome Control) | Female | 0.714 | 0.489 | 4.895 | 0.001 |

### *4.4.6.2 Amount of variance explained.*

Previous studies have shown that the variance in hue preference curves across populations is explained by at most three principal components, and that the first two of these closely match the LM and S cone-opponent contrast components of the hues. For example, in a group of 94 adults, the first two PCs explained 69% of the variance in hue preference curves across the population, while the LM and S components explained 74% of the variance, with the S component contributing approximately 51% and the LM component 22% for all subjects (Hurlbert & Ling, 2007). The

regression coefficients indicate the goodness of fit of the cone-contrast model for each individual subject.

The amount of hue colour preference variance explained by the Eskew cone contrasts was also compared between the groups. Independent sample t-tests were used to compare the overall variance in hue colour preference explained by both Red-Green and Blue-Yellow axes coefficients between participant groups. To supplement this a repeated measures ANOVA with a within subject factor of variance explained by axis (2 levels: Red-Green or Blue-Yellow) and between subject factor of sex (2 levels: Male or Female) and group (autism or Williams Syndrome and TD) was also conducted to assess the relative contribution of both colour axes to explaining variation in colour preference.

#### 4.4.6.2.1 Autism

In the autism analysis, there was no significant difference between the autism and TD groups in the amount of hue variant colour preference which can be explained by the cone contrast values of the stimuli, $t$ (34) = 1.706, $p$ = 0.097 (see Figure 4-13). The repeated measures ANOVA did not find a main effect of colour axis, $F$ (1, 32) = 0.352, $p$ = 0.557, or a significant interaction between colour axis and group, $F$ (1, 32) = 0.801, $p$ = 0.377. There was a significant interaction between colour axis and sex, $F$ (1, 32) = 16.161, $p$ < 0.001. This was driven by significantly higher variation for male's hue colour preference explained by Red-Green cone contrast compared to Blue-Yellow cone contrast, $t$ (21) = 3.409, $p$ < 0.01, a pattern that was not found in the female group, $t$ (21) = 1.653, $p$ = 0.124. There was also a dissociation between the colour axes that most explained colour preference between sexes. For males, the Red-Green axis explained the most variance (although there was only a trend towards a significant difference between sexes, $t$ (34) = 2.239, $p$ = 0.064), whilst for the females the Blue-Yellow axis explained significantly more variance than it did for the male participants, $t$ (34) = 3.343, $p$ < 0.01. There was also a significant three-way interaction between colour axis, sex and group, $F$ (1, 32) = 19.422, $p$ < 0.001. To explore this three -way interaction two way ANOVAs were conducted for each participant group (Within subject factor: Colour Axis, Between subjects' factor: Sex). For the TD group, there was a significant interaction between sex and colour axis, $F$ (1, 16) = 78.557, $p$ < 0.001, where males hue colour preference variance was explained significantly more by the Red-Green contrast coefficient, $t$ (16) = 4.728, $p$ < 0.001, whilst females hue colour preference variance was explained significantly more by the Blue-Yellow contrast coefficient, $t$ (34) = 7.932, $p$ < 0.001. There was no significant main effect of colour axis ($p$ = 0.755). For the autism analysis, there was no significant main effect of colour axis or interaction between colour axis and sex (lowest $p$ = 0.415).

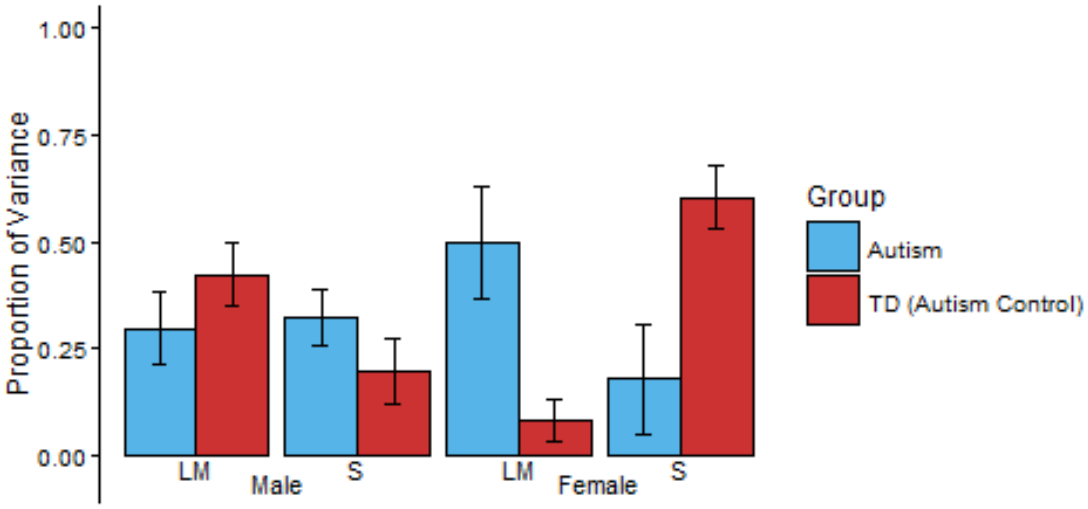



*Figure 4-13– The total amount of hue colour preference variance explained by cone contrasts for autism and TD groups.* These are further split by sex.

#### 4.4.6.2.2 Williams Syndrome

For the Williams Syndrome analysis, there was no significant difference between the Williams Syndrome and TD groups in the total amount of variance that was explained by the cone contrast values of the stimuli, $t$ (50) =1.18, $p$ = 0.2, see figure 4-14. The repeated measures ANOVA showed no significant main effect of colour axis, $F$ (1, 48) = 0.31, $p$ = 0.886. There was a significant interaction between colour axis and sex, $F$ (1, 48) = 7.966, $p$ < 0.01. This driven by an increased amount of variance explained by the blue-yellow axis in the female participants, $t$ (25) = 2.853, $p$ <0.01, no such difference was found in the males ($p$ = 0.147). There was no significant interaction between colour axis and group or three-way interaction (lowest $p$ = 0.536).

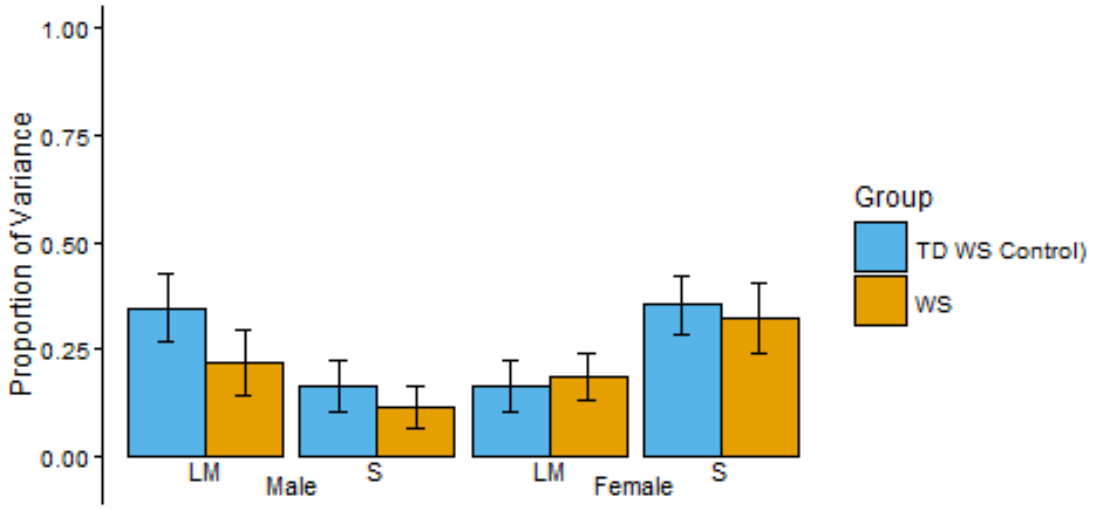



*Figure 4-14– The total amount of hue colour preference variance explained by cone contrasts for autism and TD groups.* These are further split by sex.

### 4.4.7 Relationship between colour preference, age, and chromatic discrimination

The colour preference descriptors obtained above (specifically hue and colour preference strength, cone contrast coefficients) were correlated with demographic descriptors and other test results (specifically the chromatic discrimination thresholds in the three dimensions, see Chapter 3) of the participants. Bonferroni corrected p-values were used to correct for multiple comparisons. Correlations were conducted on participants divided either by group (autism/Williams Syndrome vs. TD) or by group and sex.

#### *4.4.7.1 Autism*

At the group level, there were no significant relationships between chronological age and any colour preference measure for either the TD (lowest $p = 0.206$) or the autism group (lowest $p = 0.096$). However, there were significant correlations with performance on the CCDT. At the group level, there were significant correlations between Blue-Yellow axis thresholds and strength of hue preference for both the TD, $r = -0.592$, $p < 0.05$, and the autism group, $r = -0.634$, $p < 0.05$ (see figure 4-15). This was a pattern that was seen in the male participants of both TD and autism groups. There were no significant correlations with individual cone-contrasts and any demographic or experimental measures in either autism or TD groups (lowest $p = 0.125$).

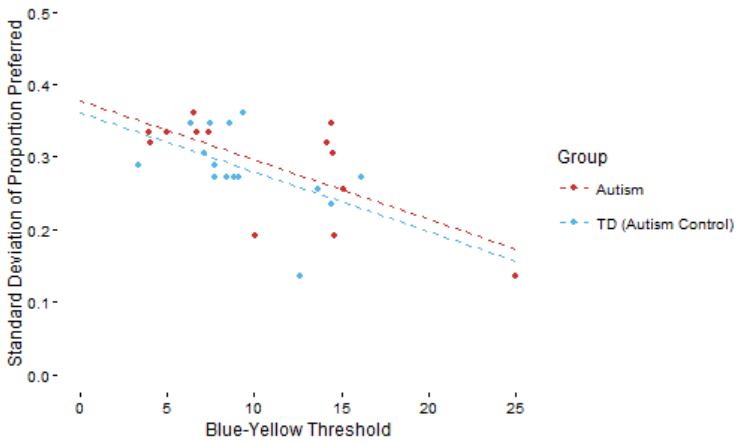



*Figure 4-15– The total amount of hue colour preference variance explained by cone contrasts for autism and TD groups.* These are further split by sex.

#### *4.4.7.2 Williams Syndrome*

In both TD and Williams Syndrome there was a significant correlation with Red-Green Thresholds and L-M cone contrast weights. For the TD group, there was a significant negative correlation between Red-Green threshold and the L-M cone-contrast weighting, $r = -0.486$, $p < 0.05$. In the Williams syndrome group, there was a positive correlation between Red-Green thresholds and standard deviation of hue preference indicating that variability in hue preference increases with better Red-Green chromatic discrimination, $r = 0.476$, $p < 0.05$. When split by sex, in the TD males there was a significant negative correlation between chronological age and S-cone cone-contrast weights, $r = -0.657$, $p < 0.05$. There were also significant positive correlations between verbal IQ, $r = 0.708$, $p < 0.01$, and non-verbal IQ, $r = 0.723$, $p < 0.01$, and S-cone cone-contrast weights. No other correlations were significant for either TD males (lowest $p = 0.121$) or Williams Syndrome Female groups (lowest $p = 0.136$). In addition, there was no significant correlations between measures of colour preference and demographic or performance on the CCDT in the TD female (lowest $p = 0.136$) or Williams Syndrome male groups (lowest $p = 0.136$).

## 4.5 Discussion

### 4.5.1 Summary of results

The main aim of this study was to determine whether colour preferences of atypically developing children, diagnosed with autism or Williams Syndrome, differed from mental age controls and whether the captured colour preference patterns were (a) age-appropriate and (b) otherwise similar to preference patterns of typically developing individuals. All three groups, the autism, Williams Syndrome and both TD groups, display colour preferences that depend on hue, lightness and

saturation, and are readily captured by a simple paired comparison test (see table 4-7 for summary). Hue preference patterns, at constant lightness and saturation, were similar, on average, in all groups. All groups showed sex-specific differences, with males being more likely to prefer bluish and greenish hues, and females more likely to prefer purple and pinkish hues. These sex-specific differences are also seen in the weightings on the cone-contrast components of the hue preference curves: males are more likely to prefer "greenish" hue contrasts (negative weights on the L-M component), while females are more likely to prefer "reddish" (positive weights on the L-M component) and "bluish" (positive weights on the S component) contrasts, although both the autism and Williams Syndrome groups cone-contrast weightings were weaker than both TD control groups. All groups preferred colours with higher saturation or higher lightness, on average, to colours with lower saturation or lightness, independent of hue. Yet the strength of colour preference varied between groups, with the Williams Syndrome group showing significantly weaker hue preference patterns (flatter preference curves as a function of hue), at the mid lightness/saturation combination only, independent of sex. They also show reduced variance explained cone-contrasts and greater inter-individual variations. The autism group showed weaker sex-specific differences in hue preference (as revealed in less sex dependence of the cone-contrast weights) and weaker colour preference dependence on lightness. The results suggest the colour preference is atypical in both autism and Williams Syndrome but that this atypicality varies between these groups.

*Table 4-7 – Summary of Colour Preference Results.*

| Measure | Group | Result |
|---|---|---|
| Hue Variants | Autism | Sex-specific but no difference between groups. |
| Hue Variants | Williams Syndrome | Sex-specific but no difference between groups. |
| Saturation Variants | Autism | High Saturation variants preferred over mid saturation variants. No group differences. |
| Saturation Variants | Williams Syndrome | High Saturation variants preferred over mid saturation variants. No group differences. |
| Lightness Variants | Autism | Females prefer lighter colours, males prefer darker colours. No group differences. |

| | | |
|---|---|---|
| Lightness Variants | Williams Syndrome | Females prefer lighter colours, males prefer darker colours. No group differences. |
| Strength of Colour Preference | Autism | No group or sex differences |
| Strength of Colour Preference | Williams Syndrome | Trend towards less variation in the Williams syndrome group. |
| Individual Colours | Autism | No group or sex differences |
| Individual Colours | Williams Syndrome | No group or sex differences |
| Cone-Opponent (Weights) | Autism | Sex differences between cone opponent axes. Sex response is greater in TD group for both sexes. |
| Cone-Opponent (Weights) | Williams Syndrome | No group, sex or axes effects. |
| Cone-Opponent (Variance) | Autism | No group or axes effects. |
| Cone-Opponent (Variance) | Williams Syndrome | No groups, sex or axes effects. |

### 4.5.2 Relationship to previous Studies of Hue, Saturation and Lightness Preference

The colour preference patterns for variations across hue, in general, were like those found in previous studies of children and young adolescents (considering varying degrees of control of the other attributes of colour) (Bonnardel et al., 2006; Child et al., 1968a; Ling & Hurlbert, 2011). Sex differences in hue preference are also like those found in previous studies, with male preferences peaking for bluish/greenish hues, and female preferences peaking for purplish or pinkish hues, on average (Bonnardel et al., 2006; Child et al., 1968a; Franklin, Bevis, et al., 2010; Hurlbert & Ling, 2007; Ling & Hurlbert, 2011; Palmer & Schloss, 2010). Whilst the majority of previous research has been conducted in adults, the hue preference curves are similar to similar aged participants across all three groups (Child et al., 1968a; Ling & Hurlbert, 2011) suggesting that the disparity in chronological age between the autism, Williams Syndrome and TD groups does not affected their colour preference for hue variants.

Although the Williams Syndrome group's hue preference patterns show similar sex differences, the hue preference strengths were weaker in comparison with the TD group. Conversely, the dependence of the Williams Syndrome group's colour preference on lightness and saturation is like that of the TD group, with similar strengths, and similar sex differences. It must be emphasised, though, that these statements are true only on average for the group, and that in the Williams

Syndrome group there was considerable inter-individual variation in colour preference, with participants being more strongly affected by one or more of hue, lightness and saturation.

Participants in all groups were more likely to prefer high saturation and high lightness colours. This pattern echoes previous studies' findings that young children and adolescents prefer highly saturated and high lightness colours (Child et al., 1968a; Ling & Hurlbert, 2011). Yet, while the females in both groups of the Williams Syndrome analysis (Williams Syndrome and TD (Williams Syndrome Control) were more likely than males to prefer highly saturated colours, as in previous studies, this sex difference did not reach significance in the autism analysis. Given that there was a trend in that direction, the lack of significance in sex difference for the preference dependence on saturation likely reflects the reduced statistical power due to the reduced number of females in the autism group compared to the Williams Syndrome analysis, rather than other explanations such as the "extreme male brain hypothesis" (Baron-Cohen, 2002). There is little other experimental evidence for sex differences between individuals with autism (Mandy et al., 2012), particularly on differences in sensory processing.

For lightness preference, there were significant interactions with sex in both the autism and Williams Syndrome analysis, driven by increased preference for lighter colours by females. This finding echoes previous studies' findings for increased preferences for high lightness colours in young female children compared to male children (Child et al., 1968a). Although these preferences for lightness and saturation occur generally across all colours, there are clear variations in The dependence of the lightness and saturation preference patterns on which demonstrate that different hues may have different lightness-saturation combinations for "peak preference" Further work is needed to clarify the nature of these relationships between hue, lightness and saturation.

In this study, there were no significant correlations in any group between chronological age and measures on the colour preference task. The studies which find colour preference changes with chronological age examine a large age range or difference between ages, e.g. from infancy to early adolescents to throughout adulthood to elderly ages (Bonnardel et al., 2006; Franklin, Bevis, et al., 2010; Ling & Hurlbert, 2011). These results, taken together, suggest that colour preference changes with chronological age are coarse and that no effect is found in this study due to the much narrower age range.

The results therefore demonstrate that the colour preferences of children with Williams Syndrome and autism are broadly like their mental-age-matched typically developing peers. Since these mental-age-matched peers are younger in chronological age than the experimental groups, the question remains, though, whether the colour preference patterns of the atypically developing children are, in fact, chronologically age-appropriate (see Limitations section below for wider discussion). The fact

that colour preference does depend, albeit coarsely, on chronological age in typically developing populations, suggests that there is delayed development in colour preference in the autism and Williams Syndrome groups studied here.

The finding that preference patterns differ little between the autism and Williams Syndrome and their matched TD groups, respectively, also suggests that the social and cultural influences on preference are intact and typical in the atypically developing group. This result may seem surprising, in the light of the known atypicalities in social communication characteristic of both groups, and their reported difficulties in the extraction of social norms and behaviours (Bauminger, 2002; Gosch & Pankau, 1997; Kunce & Mesibov, 1998; Mervis & John, 2010; Rosner, Hodapp, Fidler, Sagun, & Dykens, 2004). In contrast to these reported difficulties, the results in this thesis demonstrate that, at a group level, both autism and Williams Syndrome groups could extract societal/cultural norms for colour preferences. Yet the previous work has almost exclusively focussed on behaviours with a heavy social aspect, and it is possible that there is difficulty in the extraction of norms when a highly social component is involved but not when it is absent. For example, it is debateable whether the same social norms are involved in understanding that there are culturally bound sex differences in colour preferences compared to identifying the appropriate behaviour for conversations or personal distance. The true test of the extent to which individuals with autism or Williams Syndrome can extract these societal norms for colour preference would be a cross cultural study of colour preference involving both typically developing and atypically developing groups. Nonetheless the results of this study suggest that individuals with autism and Williams Syndrome can extract non-demanding social norms, despite the difficulties in extraction of more socially demanding norms identified as part of the core characteristics of each condition.

### 4.5.3 Relationship of findings to models of colour preference.

Previous studies have demonstrated some degree of consistency, at the population level, to the pattern of hue preference variation in different populations (for example, young adults of either British or Chinese/Russian nationalities) (Hurlbert & Ling, 2007; Ling & Hurlbert, 2011; McManus et al., 1981; Sorokowski et al., 2014)). This regularity is captured by the fact that a small number of factors is needed to explain most of the variance in hue preference curves across the population. Hurlbert and Ling (2007) proposed that the neural mechanisms underlying low-level colour-encoding, i.e. the cone-opponent-contrast components, explained much of the variance in hue preference curves for young adult populations (up to 70%), and weights on these factors also captured sex differences in preference well. For the red-green component, males were more likely to prefer colours with a greenish contrast, whilst females preferred colours with a reddish contrast; for the blue-yellow component, both males and females preferred colours with a bluish contrast.

The results reported in this thesis, for young children and adolescents in the TD, autism and Williams Syndrome groups, also demonstrate an underlying regularity in hue preference, well captured by characterising preference patterns in terms of cone-contrast components, but with a variability that argues against complete universality and requires instead additional factors to explain individual variations. For example, although for the autism analysis, between 62% (autism-matched TD males) and 76% (autism females) of variance in hue preference curves is explained by weightings on cone-contrast components, the variances explained for the Williams Syndrome analysis are lower, ranging from 33% (Williams Syndrome males) to 54% (TD females, Williams Syndrome Control). This weaker regularity in the Williams Syndrome analysis might be partially explained by the younger chronological ages of the TD group, and the overall lower mental ages of both TD and Williams Syndrome, in comparison with the autism analysis and with the earlier studies of young adults.

The reduced dependence of hue preference on cone-contrast components might therefore represent a developmental change from young children to adults, in which the encoding of preferences develops with age alongside the basic discriminatory encoding of colours in terms of cone-contrast components. It is known that chromatic discrimination ability along the distinct cone-contrast axes does improve with age from childhood into early adulthood (Knoblauch et al., 2001; Paramei, 2012; Paramei & Oakley, 2014) (see also Chapter 3 for wider discussion). The finding that preference strength (variance of the hue preference curve) increases with increasing chromatic discrimination ability suggests that the development of preference may parallel the development of chromatic discrimination ability. On the other hand, the lack of correlation between the total preference variance explained by the cone-contrast components and chromatic discrimination ability, in all groups tested, argues against a directly causal link between the two. Importantly the finding of reduced chromatic discrimination ability associated with reduced preference strength in the autism group also argues against the hypothesis that hyposensitivity to stimuli induces obsessive interest in the stimuli (as in a fascination with brightly coloured objects (Bogdashina, 2003). The finding instead argues that increased chromatic discrimination is related to greater peaks/troughs in hue colour preference (see section 4.4.7). Previous research predominately utilising questionnaires has provided modest evidence for this relationship ((Boyd et al., 2010; Boyd et al., 2009; Wigham et al., 2015), see also Chapter 1 section 1 and Chapter 6 for wider discussion of these studies). However, it is important to note that strength of colour preference alone does not indicate repetitive behaviour. This link will be assessed in Chapters 6 and 7 where the between experimental measures, i.e. strength of colour preference and chromatic discrimination, and the presence/absence of colour affected behaviours.

It is important to note there was also wide inter-individual variability within each participant group in the amount of variance explained by the cone-contrast component coefficients, with some

participants' hue preference patterns almost exclusively relying on the variation along the two-colour axes, and others showing little relationship to these, suggesting that individual factors at other levels must influence colour preference. Despite this variability, the sex differences in cone-contrast component weights found in previous studies are also evident in all three groups, although the sex differences were lower in the autism and Williams Syndrome groups.

Colour preferences have been suggested to be related to socialised normative colours in young children (Jadva et al., 2010; LoBue & DeLoache, 2011) and it is possible that these colour object associations drive colour preference more strongly in children, but that these are not significant factors for adults. The ecological valence theory proposes that colour preference intrinsically tied to objects. Colour preference arises from the associated valence of these objects which transfers over to the colour (Palmer & Schloss, 2010). Palmer & Schloss (2010) found that colour-object associations accounted for more variation in colour preference, over a broad range of lightness and saturation combinations, than cone contrast component weightings for the same stimuli. To assess object valence requires additional testing of the same individuals or separate testing of a perfectly matched group. Given the relative rarity of Williams Syndrome and autism and reduced time commitments it was not possible to estimate these colour-object associations for the TD, autism and Williams Syndrome groups. However, the atypical cone contrast weights in both the autism and Williams Syndrome groups suggests that in both clinical groups that colour preferences do not necessarily arise through the visual system. Instead colour preferences could arise through colour-object associations as proposed by the ecological valence theory. A bespoke colour questionnaire was developed to explore the nature of colour affected behaviours in the same participants. From this questionnaire, it will be possible to assess the relationship between colour-affected behaviours and colour preference, providing similar information to that from a specific test of the ecological valence theory (see Chapters 6 and 7 for more details on the questionnaire and case studies).

### 4.5.4 Limitation

#### *4.5.4.1 Stability of preferences over time.*

There is consistent evidence that colour preferences change with chronological age (see above section on Developmental Changes in Colour Preference). Colour preference effects due to hue, saturation and lightness follow an inverted "*U*" with a peak in childhood and early adolescence, with the effect of hue, saturation and lightness decreasing with age (Child et al., 1968a; Dittmar, 2001; Ling & Hurlbert, 2011). Previous research studies have used a cross sectional design to compare colour preference across different ages. It is unclear the extent to which colour preferences change for an individual over the course of their life. Longitudinal studies would address this gap in the research, but given that changes in colour preference have been demonstrated to be slow and change from early childhood into late adulthood (65+ years) such studies might not be practical.

A related issue is the differences in chronological age between the autism and Williams Syndrome groups compared to their TD mental age control groups. The disparity between mental and chronological age of the participants with autism and Williams Syndrome means that both clinical groups are older than the TD control matches. As outlined above, the relative strength of colour preference decreases with chronological age. There may therefore be differences in colour preference for both clinical groups when they are compared to control groups matched on chronological age. In the absence of chronological age matches, the hue preference curves of the autism and Williams Syndrome groups were compared to those of a similar aged young adolescent group reported by Ling and Hurlbert (2011). The hue preference curves for the autism and Williams Syndrome groups were qualitatively like TD adolescents of approximately the same chronological age, where at the group level females prefer reddish and purplish hues over brownish hues whilst males on average prefer bluish and greenish hues and dislike hues that are reddish and purplish. One slight difference can be found in the Williams Syndrome group hue preference curves, where the curves appear to have less pronounced peaks and troughs indicating that at the group level the Williams Syndrome participants show less clear colour preferences compared to both mental and chronological TD groups. As discussed above, the mean preference curve does mask considerable inter-individual variability in the colour preference curves (also see Chapter 6 for wider discussion in relation to parental questionnaires). It is also unknown the extent to which, if at all, colour preference in autism and Williams Syndrome change with chronological age, like the unknown for TD controls. Given the lack of experimental research on colour preference in both autism and Williams Syndrome, it is not possible to compare the results of this study with analogous research in older participants with either condition. Future research should aim to investigate a wider and older age range to see whether the results found in this study continue into adulthood.

## 4.6 Conclusion

This chapter investigated colour preference in autism and Williams Syndrome relative to mental age-matched TD controls. Both autism and Williams Syndrome groups showed similar patterns to their respective TD control group at the group level and when divided by sex for colour preference along hue, saturation and lightness variations. There was also similarity between groups in the amount of colour preference that could be explained by the cone contrast components of the stimuli. Yet there were some group differences. The Williams Syndrome group were found to have less pronounced dependence on hue in their colour preference, whilst the autism group's colour preference was less related to the cone contrast values. This suggests that colour preference is atypical in both autism and Williams Syndrome groups, but that this atypicality differs between the groups. The next chapter will investigate another behavioural response of colour, colour naming, to see whether there are differences between different types of behavioural responses of colour.

# Chapter 5 - Colour Naming

## 5.1 Overview

The previous chapter assessed colour preference in autism and Williams Syndrome. This chapter will investigate another high-level use of colour; colour naming. Looking at multiple higher-level uses of colour (e.g. language, aesthetic judgements) will further reveal the nature of how colour is used by individuals with autism or Williams Syndrome, beyond initial low-level sensory processing. Colour naming involves the development of conceptual categories of an abstract relational space. This ability to name appropriately abstract spaces has previously been found to be poorer in Williams Syndrome, but not autism, suggesting a possible dissociation for colour naming. Colour naming also has a high language component, and given the documented difficulties in language for individuals with either autism or Williams Syndrome, it may be that higher-level responses to colour where there is a strong language component may be at more risk of being atypical.

## 5.2 Introduction

### 5.2.1. Overview of colour naming

Colour naming is the act of giving a linguistic label to denote certain regions of colour space. Its purpose is to divide the continuous light wave into separate discrete colour categories. Because of this continuum there are no limits on the number of colour categories that can be divided. The origin of these specific colour categories is unknown. There has been much debate about whether colours categories are universal or are shaped relative to language (e.g (Berlin & Kay, 1969; Davidoff, Davies, & Roberson, 1999; Franklin, Clifford, Williamson, & Davies, 2005; Kay & Regier, 2003; Roberson, Davies, & Davidoff, 2000)). There has been a recent reconciliation of these two “extreme” viewpoints, which suggests that there are underlying perceptual colour categories which change upon acquisitions of colour terms (Franklin, Clifford, et al., 2005; Franklin & Davies, 2004; Franklin, Drivonikou, Clifford, et al., 2008). An in-depth review of these theories is beyond the scope of this chapter but the central aspects of these theories and how they propose the origins of colour categories occur will be considered in the next section.

### 5.2.2 Universal or Relative colour categories?

There is the same underlying physiological basis of how colour is perceived in humans using a cone opponent mechanism (see Chapter 1 section 1.2). Despite this colour spaces are frequently divided into discrete categories using colour names. Berlin and Kay (1969) examined colour terms collected from twenty different languages. From their results, they noticed distinct overlap between the locations of colour term boundaries were like the areas of Munsell colour space. They identified eleven basic colour terms (BCTs or focal colours) that were present in all Western languages. These colour terms were black, white, red, green, blue, yellow, brown, pink, purple, orange and grey (see

figure 5.1). Other colour terms existed outside of these but were not consistent between languages. These less common colour terms were described as non-basic colour terms (non-BCTs or non-focal colours). The researchers also proposed a seven-stage hierarchy in the cultural development of these colour categories (see figure 5.1). The first stage distinguishes between dark and light (in English black or white). The second stage was the addition of red. Stage 3 was the use of either green or yellow, with stage 4 the addition of the missing term in stage 3. The next colour term added was blue in stage 5, with brown being included in stage 6. The final stage is where purple, pink, orange and grey are added. In this stage, there is no specific order in which these colours were added to the BCT list. This evolutionary hierarchal framework gives a potential structure in which the relative importance of each BCT is considered (Berlin & Kay, 1991). There are 6 primary colours that are perceptually unique (white, black, red, blue, green and yellow), and 5 secondary colours that are more complex (brown, orange, pink and grey). There have been recent suggestions of revisions the structure proposed by Berlin and Kay (1991) to consider evolutionary importance of certain colours between culture and languages. One suggested change has been to divide the primary colours into "warm" (e.g. red and yellow) and "cool" (blue and green) colours (Kay & Maffi, 1999). This notion of primary/secondary and warm/cool colours give a possible developmental order in which colour terms are learned by children (See sections 5.2.3 on Development of Colour Naming and Constraints of Learning Colour Terms).

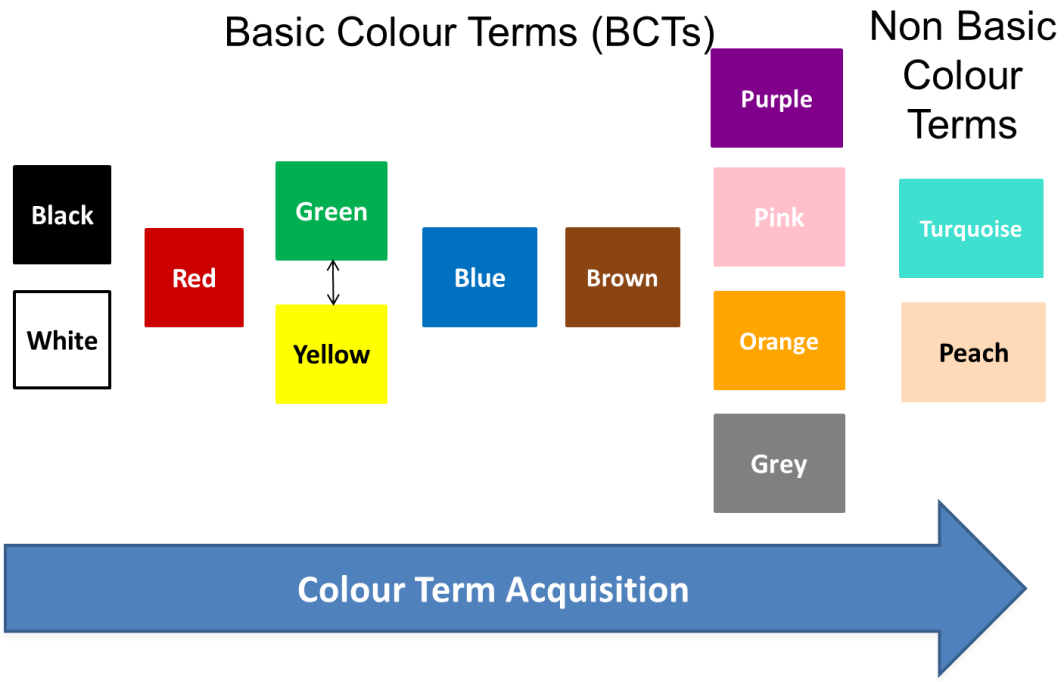


*Figure 5-1 – The eleven BCTs proposed by Berlin and Kay (1969).* The colours are ordered from left to right in terms of the order of colour term acquisition. Here primary colours (black, white, red, green, yellow and blue) are argued to develop before non-primary colours (brown, purple, pink, orange and grey). Once these eleven BCTs have been learnt then non-BCTs develop (illustrated here by Turquoise and Peach).

The linguistic relativity or Sapir-Whorf hypothesis states that thoughts are shaped by the language available to the person. With reference to colour naming this states that how you perceive colour is restricted by the number of colour terms that are available to you, i.e. that blue is perceived because there is a colour term for it, if there is no blue term then you are unable to perceive the colour as blue (e.g. (Davidoff et al., 1999; Roberson et al., 2000)). Thus, the mere presence of colour terms impact upon colour perception and cognition. This idea stems from how different languages divide up colour space using different colour terms. If colours are linguistically relative, then this means that speakers of different languages will qualitatively experience colour differently. For example, in Russian and Greek there are two commonly used terms for blue, one for light blue and another for dark blue, whilst in English there is only blue. This additional colour term has been found to benefit individuals on cognitive tasks such as visual search where there is enhanced performance in Russian/Greek speakers over English speakers, where there is a colour term between the colour space that the two original blue colour terms occupy (e.g. (Athanasopoulos, 2009; Winawer et al., 2007). Furthermore, this linguistic effect may be mediated by the dominant language. In bilingual Japanese and English speakers, the nature of between category effects changed depending on whether Japanese or English was the dominant language for that individual (Athanasopoulos, Damjanovic, Krajciova, & Sasaki, 2011). This effect of colour terms is not limited to between languages. There is faster identification of a coloured target when it is amongst distractors from a different colour category, but there is no accuracy benefit when the target and distractor are from the same colour category. Crucially in these experiments there is the same perceptual difference between targets and distractors. This means that faster identification of targets only occurs when a linguistic boundary is crossed (Franklin, Drivonikou, Bevis, et al., 2008; Holmes, Franklin, Clifford, & Davies, 2009; Roberson, Pak, & Hanley, 2008). Further evidence for the linguistic relativity hypothesis comes from psychophysical studies, where there has been no evidence found for distinct colour categories in the visual system (Boynton, 1997; Brown, Lindsey, & Guckes, 2011; Valberg, 2001; Webster, Miyahara, Malkoc, & Raker, 2000) although see (Witzel, Cinotti, & O'Regan, 2015) for unique hues discussion with reference to light spectra). If there are universal colour categories, then these findings suggest that they may not arise from cone-opponent mechanisms.

Despite these seemingly conflicting views, there has been a coming together of these two extreme positions. There are perceptual colour categories in infants (Franklin & Davies, 2004), however these perceptual categories are then warped once a reliable colour term has been acquired (Franklin, Clifford, et al., 2005). The same research group also found that acquiring a colour term causes a

categorical effect to occur at the colorimetric boundary of the new colour term (Franklin, Drivonikou, Bevis, et al., 2008; Franklin, Drivonikou, Clifford, et al., 2008). These findings suggest that the nature of these colour terms does vary somewhat with language, however even despite these inter language differences in colour terms there is still some consistency in the way that colour terms are used, i.e. that there is a hierarchy of colour terms, where certain colour terms are more likely to be included across difference languages (Lindsey & Brown, 2006; Regier, Kay, & Cook, 2005) but also that there is wide inter-individual variation of colour term use within a language (Lindsey & Brown, 2009).

### 5.2.2. Adult colour naming studies

A focus of adult colour naming studies has been on the extent to which BCTs are common and whether the colour terms used are the same as the BCT identified by Berlin and Kay (1969). Indeed the Berlin and Kay terms are the most frequent colour terms used by adults when using a free naming tasks (e.g. (Guest & Van Laar, 2000; Mylonas & MacDonald, 2015)). In addition to the initial eleven Berlin and Kay BCTs there have been suggestions for possible new BCT's to be added to this list. This suggestion of new terms arises from common use of non-BCTs that is of equal or near to frequency of BCT use. Peach, cream, lilac, mauve, lavender, maroon and teal/turquoise have all also been suggested as new possible BCTs in English to varying degrees of replicability (Boynton & Olson, 1987; Davies et al., 1991; Lindsey & Brown, 2014; Mylonas & MacDonald, 2015; Sturges & Whitfield, 1995; Witzel et al., 2015; Zimmer, 1982; Zollinger, 1984). Although suggested new possible BCTs may also be language dependent, for example in Greek and Russian where new BCTs have been proposed to divide between light and dark blues (Androulaki et al., 2006; Paramei, 2005; Winawer et al., 2007). Of those colours proposed as possible new BCTs, turquoise/teal has been the most replicated possible new BCT in English and other languages (Mylonas & MacDonald, 2015; Sturges & Whitfield, 1995; Witzel & Gegenfurtner, 2011; Zimmer, 1982; Zollinger, 1984). Mylonas and McDonald (2015) has conducted a series of large scale studies of colour naming in English speakers. Participants were shown 600 colours from the Munsell Renotation Dataset. The colours were chosen to sample a wide range of colours varying in Munsell value, chroma and lightness. From the participant responses, they constructed centroids within Munsell colour space to represent each colour term generated in the experiment. They found that the Berlin and Kay BCTs were used more frequently than non-BCTs by participants. The BCTs were found to occupy larger regions of colour space compared to the non-BCTs, except for yellow. From their results, they also argue for the inclusion of lilac and turquoise as additional BCTs, as these were some of the most consistently and frequently named colours. However, although they were used more frequently than other non-BCTs, the response time to name these colours was markedly slower than for BCTs. From these adult naming studies, it is clear that there is consistency in the presence of the eleven BCTs in adults. It is also apparent that there are

several non-BCTs (e.g. cream, peach, lilac and turquoise) are also used more frequently to describe certain colour space regions and that these more common non-BCTs are used more frequently used than other less common non-BCTs (e.g. gold, jade etc).

### 5.2.3. Development of colour naming

Young children have difficulty in learning colour names compared to the names of familiar objects or non-colour words (Bornstein, 1985; Carey, 1978). Prior to the development of colour naming, 6-month old infants can discriminate between different colour categories, even in the absence of colour terms (Bornstein, 1985; Bornstein, Kessen, & Weiskopf, 1976; Catherwood, Crassini, & Freiberg, 1989; Franklin & Davies, 2004). These pre-linguistic colour categories are perceptual instead of linguistic and they continue in pre-school children, where colour names have still not been learnt (Bonnardel & Pitchford, 2006). The development of colour naming is further complicated because young children are able to learn colour names and have colour as a separate semantic category, i.e. colours are different from animals, prior to them being able to reliably map the use of a colour name onto the physical stimulus of a colour (Backsheider & Shatz, 1993; Bartlett, 1978; Cruse, 1977; Istomina, 1963; Kowalski & Zimiles, 2006; Sandhofer & Smith, 1999; Soja, 1994). Furthermore it has also been suggested that colour may not be the most salient aspect of an object in comparison to other perceptual factors, e.g. shape, for young children (O'Hanlon & Roberson, 2006). Both these issues however primarily relate to learning the first colour word. Once an initial colour term has been learnt and not subsequent learning of later colour terms (Bartlett, 1978; Franklin, 2006). Bartlett (1978) suggested that once four BCTs have been learned then future judgements are made using perceptual adjacency (e.g. extending red to pink; see also (Soja, 1994)). This suggests that once colour words are initially acquired and are then accurately mapped onto the actual visual percept of that colour that new colour names are extensions of those that are like the colour names that the child possesses. The types of errors that are made by young children learning colour names supports this notion. Over-extension errors are made when learning colour names, for colours at colour boundaries, for example, naming orange as red or yellow (Bartlett, 1978; Davies, Laws, Corbett, & Jerrett, 1998; Pitchford & Mullen, 2003; Wagner, Dobkins, & Barner, 2013). Wagner and colleagues (2013) recently explored these patterns of errors during colour naming. They grouped 22-36 month old toddlers based on their accuracy of naming the eleven BCTs. Participants were asked to separately identify and name the Berlin and Kay BCTs. They found that there were fewer consistency errors once colour names were more reliably understood. There were also a greater number of overextension errors regardless of comprehension of colour terms. The number of consistency and overextension errors reduced over time as colour names become more reliably learnt.

Once colour terms have been reliably learnt there are strong linguistic effects. The perceptual colour categories that are present prior to learning colour names are shifted towards the colour name

boundaries (Franklin, Pitchford, et al., 2008). Children have also shown enhanced performance for matching and remembering focal colours compared to non-focal colours (Andrick & Tager-Flusberg, 1986; Farran et al., 2012; Heider, 1971; Mervis, Catlin, & Rosch, 1975). There are similar results when children find colours from different colour categories compared to when they are the same (e.g. (Farran et al., 2012; Franklin, Pilling, & Davies, 2005)).

*5.2.3.1 Constraints on Colour Naming*

As alluded to in the above sections, there are many different constraining factors on successful development of colour naming. This section will detail different constraints found on colour naming. It has been suggested that one of the reasons why colour naming is difficult because it requires the conceptualisation of an abstract relational concepts. However, colour is not alone in having this characteristic. When comparing the development of colour naming in comparisons to other abstract relational concepts, e.g. space or size, it has been found that the learning of abstract relational concepts was more difficult in general for children than concrete objects, e.g. car, person. This suggests that there may be a general difficulty in the development of naming abstract concepts (Bornstein, 1985; Pitchford & Mullen, 2001; Rice, 1980; Sandhofer & Smith, 1999). However, there are methodological problems with many of these studies. Pitchford and Mullen (2001) only examined red-green colour naming, so it is unclear whether this result is true for other areas of colour space. In the Bornstein (1985), Sandhofer and Smith (1999) studies also have methodological problems, where there were fewer choices for the size manipulation than the colour, meaning that the relative accuracy is unequal between the size and colour stimuli.

There is also evidence for inter-individual constraints on colour term acquisition, such as verbal ability (Pitchford & Mullen, 2003), where greater verbal ability was associated with greater comprehension of more complex BCTs (i.e. brown, pink, grey). Increased preference for a colour has also been associated with reliable naming of that colour in toddlers (Pitchford & Mullen, 2005). It is unknown if this relates to the use and development of non-BCTs. Preference has also been shown to also influence the accuracy of colour naming in older children (7 years old) adults but not in 4 year old children (Pitchford, Davis, & Scerif, 2009), suggesting a possible developmental effect of colour preference on colour term acquisition between pre-school age (2 years old) and in young children (7 years old) and use of colour information, although again the stimuli were limited in this study focusing solely on Blue-Purple colours.

Chromatic discrimination has not been found to be linked to naming in children (Pitchford & Mullen, 2002, 2005). However, in both studies the chromatic discrimination measure only assessed whether the participant could discriminate between two categorically different colours, (e.g. is this brown the same as blue?) and not within colour categories (e.g. do these two blues differ?). So, it is unknown if whether a more sensitive measure of chromatic discrimination measure would be related to BCT

accuracy or for non-BCTs use. However there has been no evidence for there to be a relationship between cone contrast values of BCTs and accuracy of colour naming (Brown et al., 2011; Saunders & Van Brakel, 1999), although this is only been shown in adults. Infants have shown clear perceptual colour categories (Franklin & Davies, 2004), yet it is not clear whether this related to the cone contrast values or whether increases in chromatic discrimination with development leads to faster development of wider colour names. It is possible that chromatic discrimination may affect colour naming, i.e. it is difficult to give a colour a unique name if you are unable to distinguish it from another, but such a developmental study is yet to be conducted.

There is wide range of inter-individual variability both in the chronological age at which reliable colour naming develops but also the order in which colour terms develop (Bornstein, 1985; Franklin, 2006; Heider, 1971; Mervis, Bertrand, & Pani, 1995; Mervis et al., 1975; Pitchford & Mullen, 2002; Sandhofer & Smith, 1999; Shatz, Behrend, Gelman, & Ebeling, 1996). There is mixed evidence for the order of colour term acquisition follow those proposed by Berlin and Kay (1969). In some cases the order in which colour terms are acquired was similar to the order proposed by Berlin and Kay (1969) in English speaking pre-school children (Johnson, 1977). Furthermore there is evidence for learning primary colours before secondary colours in English speaking 3 year olds (Pitchford & Mullen, 2002). However, the order of colour term acquisition is not the same in cross-cultural studies. Children in Botswana and Russia also learnt colour terms in a different order to English speaking toddlers (Davies, Corbett, McGurk, & Jerrett, 1994; Davies, Corbett, McGurk, & MacDermid, 1998). However an advantage for primary over secondary colours is not always found in English children either (Bartlett, 1978; Shatz et al., 1996). Other research has suggested a more nuanced developmental of colour term acquisition. Brown and Grey (two secondary colours) accuracy and comprehension have been found to develop consistently later (around 46-49 months) than other BCT colours (around 36-39 months). This suggests that there is not necessarily an advantage between the other nine BCTs and that the order in which colour terms are acquired for these nine BCTs are not necessarily constrained to a given order (Pitchford & Mullen, 2002, 2005).

In summary, these studies seem to suggest that there is an initial ability to discriminate between different perceptual categories in infants. These perceptual colour categories are then subsequently modified and shaped by the linguistic colour categorical boundaries as colour terms are reliably learnt. There are further inter-individual constraints, such as verbal ability, chromatic discrimination and colour preference that are related to the development of colour naming in typically developing children.

### 5.2.4. Colour Naming in Williams syndrome and Autism

Both autism and Williams syndrome have been shown to have either atypical verbal ability which is often accompanied by delayed onset of language (Baron-Cohen et al., 1996; Loveland & Landry, 1986; Mervis & Klein-Tasman, 2003) and delayed vocabulary development, e.g. (Le Couteur et al., 1996; Mervis & John, 2012), or language regression ((Lord, Shulman, & DiLavore, 2004); see (Tager-Flusberg, Paul, & Lord, 2005) for review of language and communication abilities in autism). However, there is a dissociation in language ability between autism and Williams syndrome. In Williams syndrome language is considered a relative strength relative to non-verbal ability, whilst in autism non-verbal ability is a relative strength compared to language ability (M. Martens et al., 2008; Tager-Flusberg et al., 2005).

#### *5.2.4.1 Colour Naming in Williams syndrome*

In Williams syndrome development of other abstract relational language concepts (e.g. space and number) are poorer than concrete language ability (e.g. nouns), which are a relative strength during language development (Mervis & John, 2008). This poorer use of spatial language has been repeatedly found in Williams syndrome (e.g. (Heinze, Osório, Lens, & Sampaio, 2014; Mervis & John, 2008, 2010; Phillips, Jarrold, Baddeley, Grant, & Karmiloff-Smith, 2004). This is not surprising given the visuospatial ability difficulties that are also found in Williams syndrome. Therefore it is possible that these two deficits in visuospatial ability and language are related (e.g. (Laing & Jarrold, 2007; Landau & Hoffman, 2005)). As discussed above colour naming develops at a similar time as reliable use of spatial terms. In addition to colour being defined as an abstract relational concept, suggests that learning colour terms could potentially be difficult for individuals with Williams syndrome. Scant research has been conducted on colour naming in Williams syndrome. One study looked at colour naming in the blue-green colour region in Williams syndrome. It found that categorisation of blues and greens was typical in Williams syndrome relative to mental and chronological age matches (Farran et al., 2013). Here there were two versions of the naming task conducted using the equal distance stimuli from the blue-green colour boundary. One task was perceptual categorisation where participants had to choose whether a colour was more like either a blue or green exemplar. The second task was a verbal naming of coloured patches as being either blue or green. However, both these tasks used a forced choice method between two classes (blues/green) rather than allow free naming. Whilst this study shows Williams syndrome that colour categorisation between blue and green is typical in Williams syndrome, it is not whether colour naming itself is typical when naming colours is not constrained. Furthermore, a limited range of colours were used, so it is unknown whether this would be consistent across for other colours.

There has been no study of free colour naming in individuals with Williams syndrome. There is however anecdotal evidence for atypical colour naming on a task that used free naming (Farran et

al., 2012). Free naming of colours was used to assess memory of both focal and non-focal colours during a route learning task. On the non-focal colours the Williams syndrome abnormal colour names were given in some instances. For example, “toothpaste colour” (Farran et al., 2012, p904). However, the frequency of these terms was not recorded, so it is not known the frequency of which these terms were used or the richness of their colour vocabulary. Furthermore, since the study did not use calibrated stimuli or colours that varied systematically making it difficult to replicate the colours that were used in this study. However additional evidence for atypical colour naming in Williams syndrome comes from a case study of a young child with Williams syndrome (Capirci, Sabbadini, & Volterra, 1996). Here consistency of primary colour names developed later (5 years) than what would be expected in typical children. Instead the child with Williams syndrome had to use prototypical object names instead of using a colour name, for example using ketchup instead of red or chocolate instead of brown. The results of these studies suggest that there may be difficulty of colour naming in Williams syndrome, whether this is due to inconsistent use of terms, delayed onset or use of atypical names.

#### *5.2.4.2 Colour Naming in Autism*

Little research on vocabulary development in autism has focused on whether there is a difference in relational vocabulary relative to concrete vocabulary development. Ungerer and Sigman (1987) found that 5-year olds with autism could sort items based on colour and on other visual aspects such as form (e.g. shape). Although this study only used four primary colours (red, blue, green and yellow) and did not control the coloured stimuli. Nonetheless, this combined with enhanced or typical performance on spatial cognitive tasks, e.g. (Bertone et al., 2005; Edgin & Pennington, 2005), suggest that unlike individuals with Williams syndrome, representation of spatial information may not be a reduced for individuals with autism, either perceptually or in relational language.

As is the case with Williams syndrome research, there have been no direct studies of colour naming in ASC. Colour naming has been assessed as part of the various studies on colour and autism (P. Heaton et al., 2008; A. K. Ludlow et al., 2014; Ludlow et al., 2012; Ludlow & Wilkins, 2009; Ludlow et al., 2006) in these studies the colour naming is used as a control task for stimuli or as a screening test for participation. There were no reported incidences or different colour naming in these studies between the autism and control groups. Further suggestion that colour naming may be typical in autism comes from word generation studies. There was also no difference between colour name generation between adolescent individuals with autism and chronological age and verbal ability controls (Boucher, 1988). However only the number of terms was compared, not the content of the generated list of colour names, i.e. BCTs were not compared against non-BCTs. In addition, this study used a word generation task and does not test whether a colour name is the same in response to an appropriate colour stimuli. It is important to map the colour names used onto physical stimuli

because otherwise it is not known whether there is a match between of a colour name to its appropriate physical stimuli. For example, someone with achromatopsia may still have knowledge of colour names but their ability to reliably name these colours in response to coloured stimuli will be very poor compared to a normal trichromat.

It should be noted that because in all the studies listed above that the focus of the study is not on colour naming. To this end, the BCT and non-BCT term usage in Williams syndrome and autism have not been assessed, nor have closer examinations of the colour lexicons been explored or only assess colour name generation, not the mapping of colour names onto colour stimuli. Therefore, these studies do not record enough information as to ascertain whether colour naming is typical in either autism or Williams syndrome.

Colour naming confers the ability to map individual colour terms onto an abstract perceptual space. Understanding this process more in Williams syndrome and autism may shed light onto both language development but also a unique opportunity to understand the relationship between low level perception and how this may be related to higher order processing. Further understanding of colour naming in both Williams syndrome and autism is also crucial because colour has a role in interventions (e.g. Picture Exchange Communication System) or is used in an educational role in schools (e.g. class/wayfinding). Knowledge of whether colour naming is typical will aid the efficacy in the use of colour in these behavioural interventions. This experiment will assess colour naming in Williams syndrome and autism. Unlike previous studies in these populations it will use more representative and controlled colour stimuli in a free naming task that includes more than the 11 BCTs. It will also analyse potential constraining factors, such as chromatic discrimination, verbal ability, chronological age, on colour naming.

### 5.2.5 Rationale and Hypothesis

The rationale for this chapter was to explore qualitative reported differences in colour naming for individuals with Williams syndrome but not in autism. Unlike previous studies which either used uncontrolled stimuli or used word generation paradigms. If the development of colour naming is linked to the development of abstract relational knowledge, then this may be atypical within Williams syndrome but not autism.

## 5.3 Methods

### 5.3.1 Participants

A total of 74 participants took part in the study. There were, 23 children with Williams syndrome, 7 with autism and 44 TD control children. The number of children is fewer than the total number of children tested because this chapter was included after testing had initially begun. The demographics of the participants who took part in this study can be found in table 5-1.

*Table 5-1 – Colour Naming participant demographics. Chronological age is reported in months. Scores on the RCPM are raw scores. VIQ is the standard scores on either the WISC-III, WISC-IV or WPPSI. Standard deviations are reported in brackets.*

| Group | Chronological Age | RCPM | VIQ |
|---|---|---|---|
| TD (n=44) | 67.16 (9.59) | 17.64 (4.35) | 107.93 (12.91) |
| Williams syndrome (n=23) | 153.91 (37.54) | 15.48 (4.52) | 70.36 (13.58) |
| Autism (n=7) | 170.1 (13.99) | 30.29 (4.42) | 82.67 17.97) |

### 5.3.2 Stimuli

The MacBeth ColorChecker chart was used to present the colour stimuli (McCamy et al., 1976). The chart contains a total of twenty-four matte painted colours arranged in four rows of six. Eighteen of the colours are chromatic, with the remaining six consisting of a grey scale. Six of the chromatic colours are primary colours used in printing (red, blue, green, yellow, magenta and cyan), with the remaining twelve colours varying predominately in saturation and lightness. These include two skin tones, two representing sky and chicory flower blues, with the remaining eight colours varying to provide a good estimate across a general colour gamut. The naming experiment used the same setup as the light box and grey sheet of paper as described in Chapters 2 and 3.

### 5.3.3 Coding of Colour Terms

Participant responses were coded to separate out different patterns of naming. Firstly, responses were categorised as either a BCT or a non-BCT. Whether a colour term was a BCT or a non-BCT was decided based on the Berlin and Kay (1969) universal colour terms. A second aspect of the coding was whether a modifying term (e.g. light/dark) was used in conjunction with either a BCT or non-BCT. Separate from this was whether a colour was named using two colour terms, this could be either using two BCTs (e.g. Blue Green), a BCT and non-BCT (e.g. Blue Turquoise) or two non-BCTs (e.g. Brick Burgundy). If a participant response was “don’t know”, then this was coded as a separate category. This meant that there was a total of seven possible ways in which a colour name could be coded. Similar coding strategies have been have been used in adult colour naming studies, e.g. (Mylonas & MacDonald, 2015).

### 5.3.4 Concordance Rating

Concordance rating refers to the general agreement between individuals for the name of a coloured patch. In this study, a concordance rating was calculated using a weighted proportional measure that considers the relative frequency of the colour terms and modifier term. The concordance value was 3:1 weighted in favour of the colour term over the modifier term. When more than one colour or modifier term each term was given an equal weighting. This higher weighting on the colour term means that greater emphasis on differential colour naming, not differences in use of modifiers will predominately drive any potential differences between the TD and Williams syndrome groups. Separate concordance ratings were calculated for each participant group. This returns a total value between 0 and 1, where the lower the value the less concordance there is for the individual compared to other participants.

Three different concordance ratings were calculated. One for each participant group (Williams syndrome and TD) for a measure of group level colour naming concordance. The final concordance measure was calculated using all participants. This will give a measure of colour naming concordance for individuals of this mental age ability. Separate concordance ratings were calculated for overall concordance (all colours), chromatic concordance (colours 1-18) and achromatic concordance (colours 19-24). Since there are fewer participants with autism who took part in this study, concordance ratings were not calculated for this group.

It is important to note that concordance is different from consistency, which refers to how stable colour naming is for an individual over time. As data was collected at a single time point it was not possible to calculate colour naming consistency. Therefore, it is only possible to determine whether there are differences in colour naming concordance for one time point.

### 5.3.5 Comparison with other variables

The effect of different constraining factors was also analysed to assess their possible impact on colour naming. The following constraining factors were included; Chromatic discrimination, favourite colour, verbal ability and chronological age. Again, due to the small number of participants in the autism group, the relationship between constraining factors and colour naming was not assessed for this group.

#### *5.3.5.1 Discrimination thresholds*

One factor that may influence the ability to name colours is chromatic discrimination (see Chapter 3 for more details). Performance of participants on the CCDT test used in Chapter 3 was used as a measure of chromatic discrimination. This threshold test is more sensitive measure of chromatic discrimination than what has been used in previous child colour naming studies (Pitchford & Mullen, 2002, 2003, 2005).

The psychophysical task from Chapter 3 measured both chromatic and luminance (achromatic) discrimination. Given that the Macbeth ColorChecker chart has both chromatic and achromatic colours, it is possible to use the performance on the colour discrimination task to assess whether there is a different relationship for chromatic and achromatic low level sensory processing and colour naming for chromatic and achromatic coloured patches. An average chromatic discrimination threshold was calculated by using the average of thresholds on the "red-green" and "blue-yellow" colour axes. These were then correlated with chromatic concordance ratings and frequency of BCTs and non-BCTs. Luminance thresholds were correlated with achromatic concordance ratings.

#### *5.3.5.2 "Favourite Colours"*

Another factor that may influence colour naming is whether a colour is liked or not. As part of Chapter 4 the same participants were required to choose their favourite colours from the Macbeth ColorChecker Chart (McCamy et al., 1976). This chosen colour was used for a separate analysis to assess participants' concordance ratings for their respective favourite colours.

#### *5.3.5.3 Verbal Ability*

Verbal ability was assessed using standard scores from either the WISC-III, WISC-IV or WPPSI-IV (which ever age test was appropriate). Standard scores were used rather than raw scores to ensure comparability since verbal ability scores were collected across different IQ tests. Standard scores were correlated with concordance ratings, and frequency of BCTs and non-BCTs.

### 5.3.6 Procedure

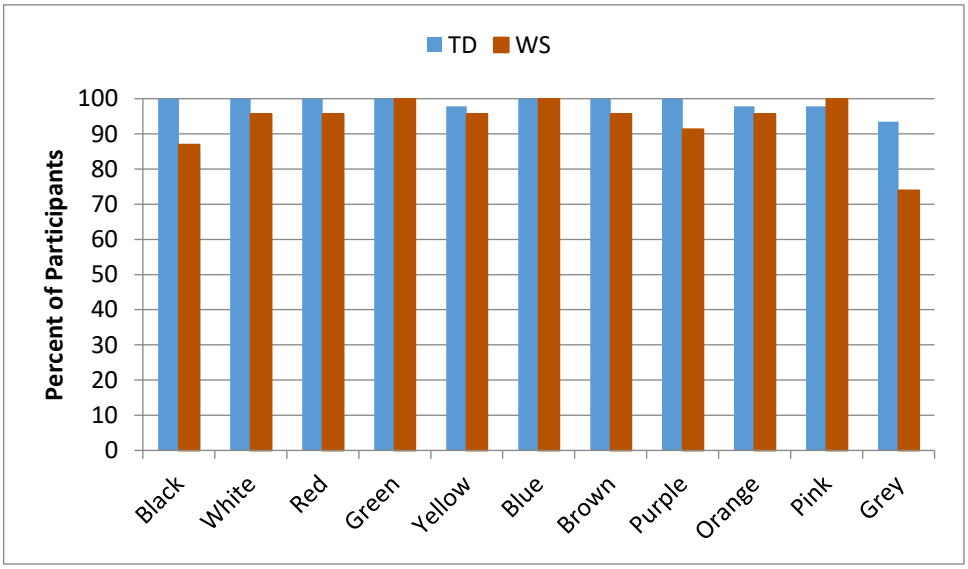
Participants were sat down in front of the light box and the Macbeth ColorChecker chart was placed centrally in front of them. Participants were asked to name all the colours on the colour checker chart sequentially from the top left to the bottom right colour patches. All colours were visible to the participant for the duration of the naming experiment. One by one each colour was pointed to by the experimenter to reduce any confusion over which colour was meant to be named by the participant. Participants were encouraged to name all colours if they could. All responses for each colour (including use of any modifiers) were recorded by the experimenter. When a participant gave two responses to a colour, for example, "blue no green", both colour names were recorded. If a participant did not name give a name to a colour they were prompted to give a name. If they did not, then this was recorded as "Don't know". Coding was completed after the testing session had finished using the scheme outlined in 5.3.4.

## 5.4 Results

### 5.4.1 Frequency of Colour Term Use

A wide range of unique BCT and non-BCT terms were used by participants in all groups (see Figure 5.1 for full list of terms used by all groups). In both the TD and Williams syndrome groups the most

common terms used were the eleven BCTs. Although the Williams syndrome group were less likely than the TD group to use all eleven BCTs, where thirty percent of Williams syndrome participants (7/23) did not use all 11 BCTs compared to six percent of TD participants (3/44). This difference in percentage of the eleven BCTs used approached significance, where the Williams syndrome group (mean rank = 29.78) were lower than the TD group (mean rank = 36.2), $U = 603$, $p=0.063$. The most commonly omitted colour in both the Williams syndrome and TD groups was grey, although some Williams syndrome participants missed other terms BCTs. Turquoise and peach were the most common non-BCTs for both Williams syndrome and TD groups. There was also a wide range of infrequent non-BCTs used by both groups (e.g. lime, lemon). In the autism group, all 11 BCTs were used by every participant. Frequency of non-BCT use was reduced compared to BCT. The most common used non-BCT was turquoise (3/7 participants). Other non-BCTs were used less frequently. All groups followed the same qualitative pattern where BCTs were more frequent than non-BCTs. Certain non-BCTs (e.g. Turquoise) were used more frequently than others, but there is a gradual reduction in the frequency of rarer non-BCTs



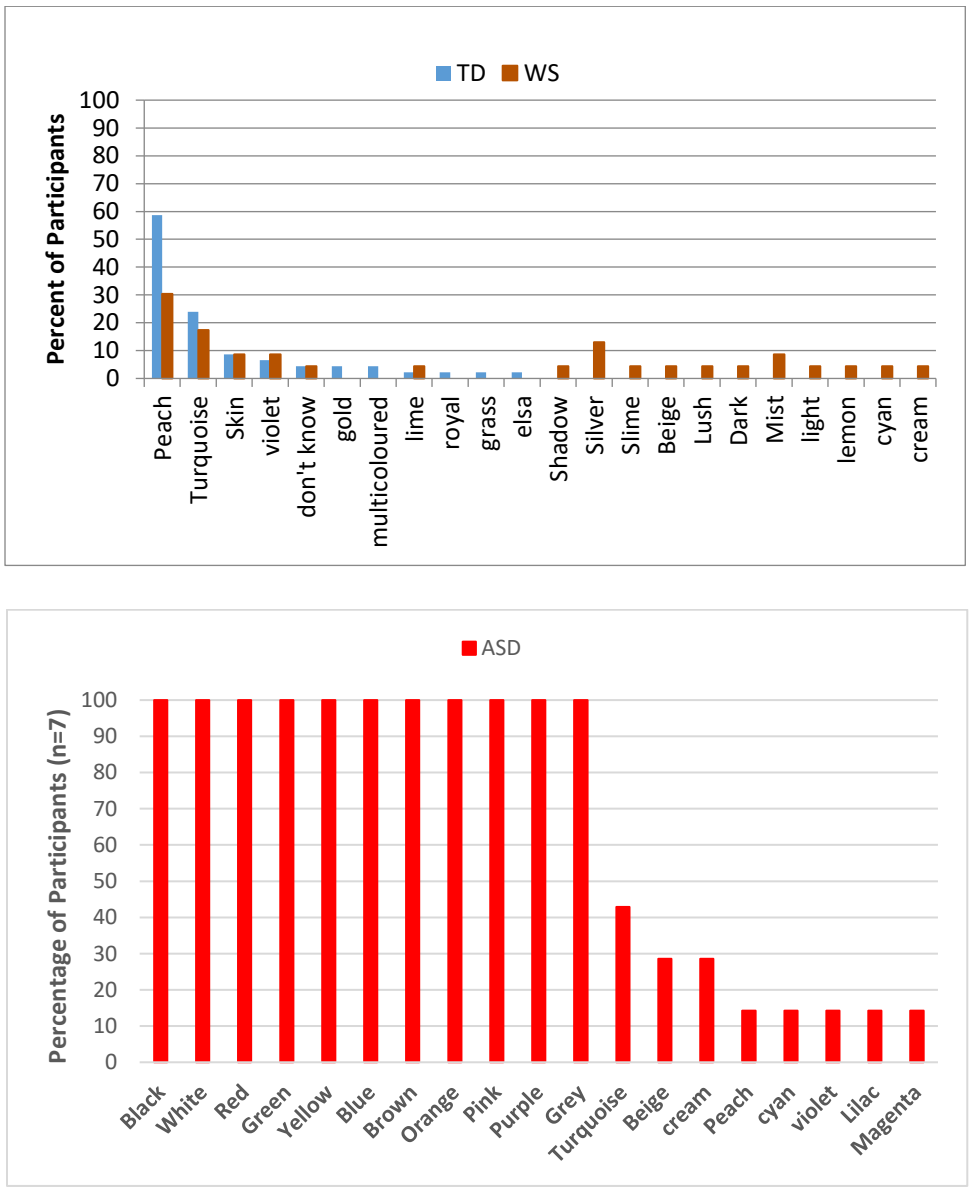


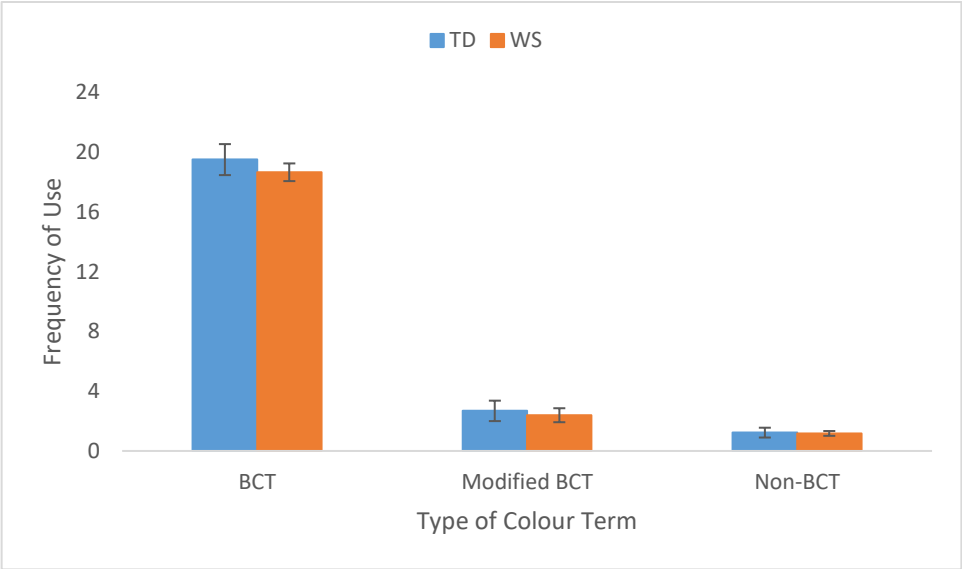

*Figure 5-2 - Frequency of colour terms used by both TD and Williams syndrome groups.* A) Shows the frequency of BCTs and B) frequency of non-BCTs for both groups. C) The frequency of BCT and non-BCT use for the autism group.

### 5.4.2 BCT/NBCT, Use of Modifiers

The frequency of type of colour term was assessed between Williams syndrome and TD groups. Due to low numbers in the autism group, no statistical tests were undertaken with this group. The total for each type of colour term (BCT, modified BCT, non-BCT). On visual inspection, it was noticed that some types of colour terms were not used frequently by participants (modified non-BCT, two BCTs, BCT with NBCT and two non-BCTs), as such these were omitted from the analysis. A repeated measures ANOVA with type of colour term (n=3) as a within subject factor and group (TD/Williams syndrome) as a between subject factor found a main effect of type of colour term, $F$ (2, 130) = 395.27, $p$<0.001. Post hoc tests revealed that singular BCTs were used more frequently than both modified BCTs, $t$(66)=18.61 $p$<0.001, and non-BCTs, $t$(66)=28.42 $p$<0.001. Modified BCTs were also significantly more common than non-BCTs, $t$(66)=3.66 $p$<0.001. There was no main effect of group or

group by type of colour term interaction (lowest $p$=0.56). In the autism group again there was a similar pattern to the TD and Williams syndrome groups where BCTs were most frequently used types of colour terms, followed by modified BCTs and then non-BCTs. However there was qualitatively different pattern in the relative difference between relative frequency of BCTs and modified BCTs where this is much less pronounced in the autism group than either the Williams syndrome or TD group. There was no discernible difference in the non-BCT use.

A clear pattern emerged in all three groups where there were differences in the frequency in type of colour name given. All groups used BCTs more frequently than modified BCTs and non-BCTs. On average two BCTs, two non-BCTs Terms that compromised of either two BCTs, two non-BCTs, modified non-BCTs or a BCT combined with a non-BCT were not used frequently in any participant group.



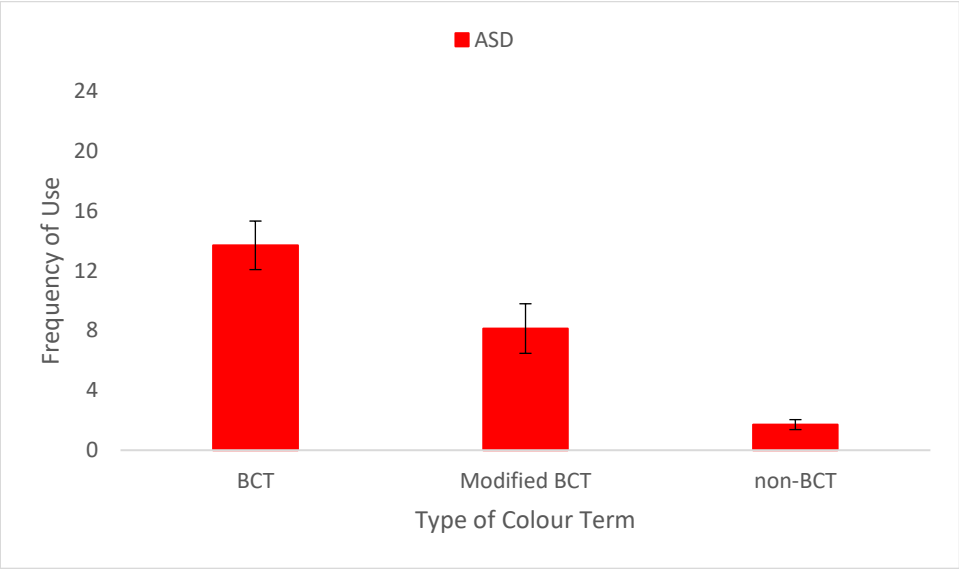



*Figure 5-3 - BCTs, modified BCTs or non-BCTs use. This is shown for each participant group across all twenty-four colour patches shown.*

## 5.4.4 Concordance

Concordance ratings were calculated for each individual colour and averaged for overall concordance (across all colours), chromatic concordance (colours 1-18) and an achromatic concordance (colours 19-24). Concordance ratings were calculated for each individual group and for the whole sample. Group concordance ratings reflect colour naming concordance within each participant group (i.e. TD or Williams syndrome), whilst overall concordance ratings represent colour naming concordance for all participants across both groups.

### *5.4.3.1 Group Concordance Ratings*

Independent samples t-tests were conducted to assess the concordance rating for each individual colour in the ColorChecker Chart. Bonferroni corrected p-values were used to control for multiple comparisons. The colours where there was a significant difference between Williams syndrome and TD concordance ratings are listed in table 5-2. There was no significant difference for any other colours (lowest $p$=0.29).

*Table 5-2– Colours that significantly differed between TD and Williams syndrome groups.* . The colour name is denoted by the name given in the ColorChecker Chart. The mean concordance ratings for each colour are displayed for both TD and Williams syndrome groups. Bonferroni corrected p-values are reported.

| Colour Name | Mean Rating TD | Mean Concordance Rating Williams syndrome | DF | t-score | p-value |
|---|---|---|---|---|---|
| Dark Skin | 1 | 0.76 | 65 | 4.01 | 0.001 |
| Light Skin | 0.39 | 0.13 | 65 | 6.45 | 0.001 |
| Blue Flower | 0.75 | 0.45 | 65 | 4.12 | 0.001 |
| Neutral 8 | 0.55 | 0.25 | 65 | 5.1 | 0.001 |
| Neutral 6.5 | 0.59 | 0.27 | 65 | 4.19 | 0.001 |
| Neutral 5 | 0.53 | 0.28 | 65 | 3.94 | 0.001 |
| Neutral 3.5 | 0.56 | 0.34 | 65 | 3.55 | 0.01 |
| Black | 0.91 | 0.42 | 65 | 7.56 | 0.001 |

There was also significant differences in group concordance ratings for all colours, $t(65)=6.72$, $p<0.001$, where the Williams syndrome group showed significantly lower concordance ratings (i.e. less agreement within the Williams syndrome compared to agreement within the TD group). To explore these differences in concordance further a repeated measures ANOVA was conducted with colour type (2 levels: chromatic and achromatic) as a within subject factor and group (2 levels: Williams syndrome and TD). There was a significant main effect of colour type, $F(1, 64) = 81.34$, $p < 0.001$. This was driven by much significantly lower concordance rating for achromatic colours than chromatic colours for all participants, $t(66)=5.82$, $p<0.001$, see figure 5.4. There was also a significant group by colour type interaction, $F(1, 65) = 41.18$, $p<0.001$. Post hoc t-tests revealed that there was significantly lower concordance for both chromatic, $t(65) = 3.22$, $p<0.005$, and achromatic colours, $t(65) = 7.91$, $p<0.001$. Here is should be noted that this effect is much stronger for the achromatic colours. There was also a difference in the relative concordance ratings between chromatic and achromatic colours, where the Williams syndrome showed significantly lower concordance for the achromatic colours than chromatic colours, $t(22) = 8.45$, $p<0.001$, whilst this was not the case in TD group, $t(43) = 2.39$, $p=0.084$.

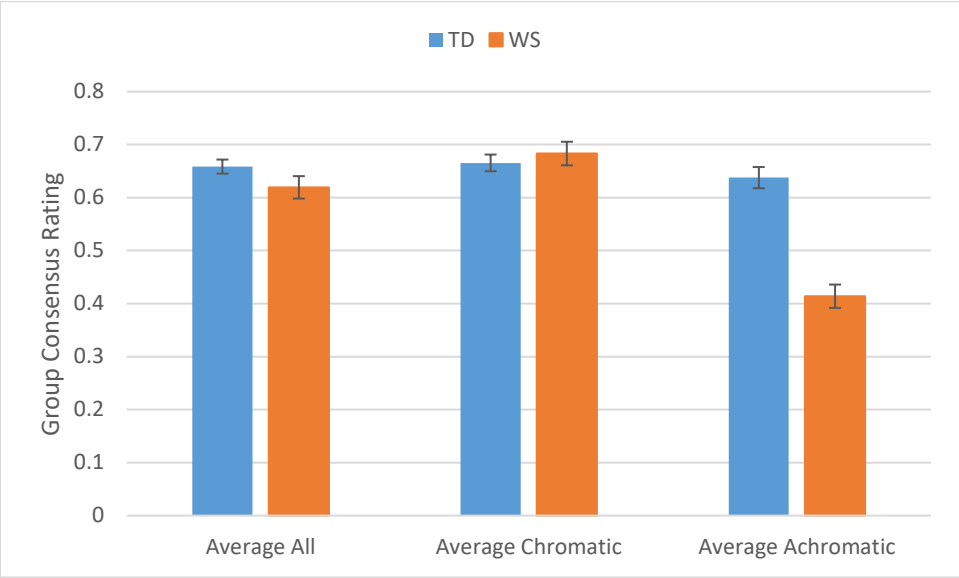



*Figure 5-4 – Group Concordance ratings across all, chromatic and achromatic colours.*

### *5.4.3.2 Overall Concordance Ratings*

In the overall concordance analysis the "Dark Skin" colour patch approached significance, $t(65)=3.17$, $p=0.072$, where the Williams syndrome group showed less concordance than the TD group. There were no other individual colours that approached significance (lowest $p=0.46$). Williams syndrome showed less concordance than the TD group for average concordance rating for all colours, $t(65)=2.7$, $p<0.05$. Similar to the group concordance ratings, the Williams syndrome showed less concordance than the TD group for the achromatic colours, $t(65)=3.5$, $p<0.001$ (see figure 5.5).

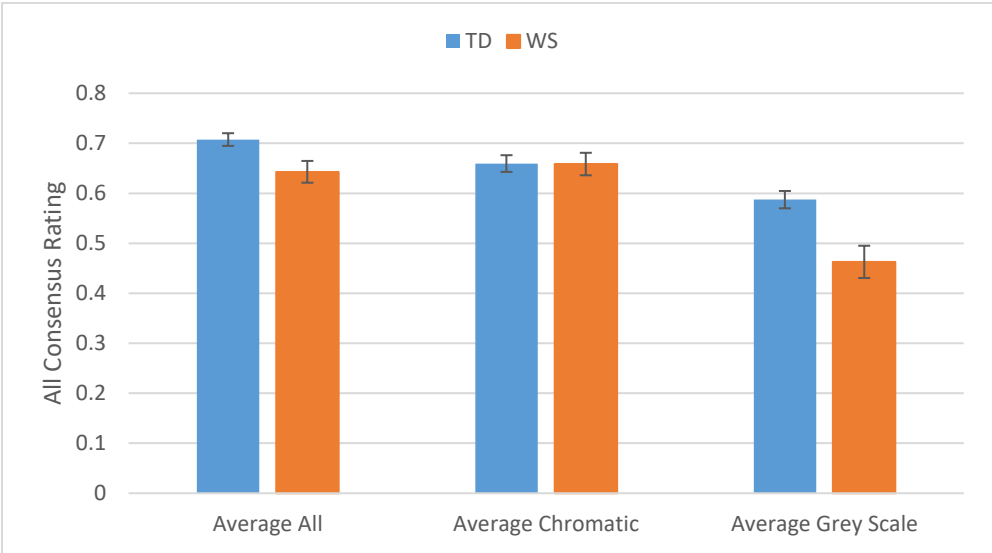



*Figure 5-5 - Concordance ratings for all participants across all colours, chromatic and achromatic colours only.*

### 5.4.4 Relationship between Colour Naming and Verbal ability

To see whether colour naming was related to verbal ability, correlations were between standard scores on the verbal subtest of either the WISC or WPPSI and frequency singular and modified BCT and non-BCT. For the TD group, there was a significant negative correlation between verbal ability and BCT frequency, $r$=-0.42, $p$<0.005. There was a significant positive correlation of VIQ with modified BCTs, $r$=0.31, $p$<0.05. Finally, there was a significant positive correlation between VIQ and non-BCT frequency, $r$=0.43, $p$<0.005. There were no significant correlations in the Williams syndrome group (lowest $p$=0.6).

Correlations were also conducted between verbal ability and concordance ratings (see figure 5.6). For the TD group, there were significant negative correlations between verbal ability and overall concordance rating, $r$=-0.41, $p$<0.01, and chromatic concordance rating, $r$=-0.45, $p$<0.005. There was no significant correlation between verbal ability and achromatic concordance ($p$=0.93). Again, there were no significant correlations in the Williams syndrome group (lowest $p$=0.26).

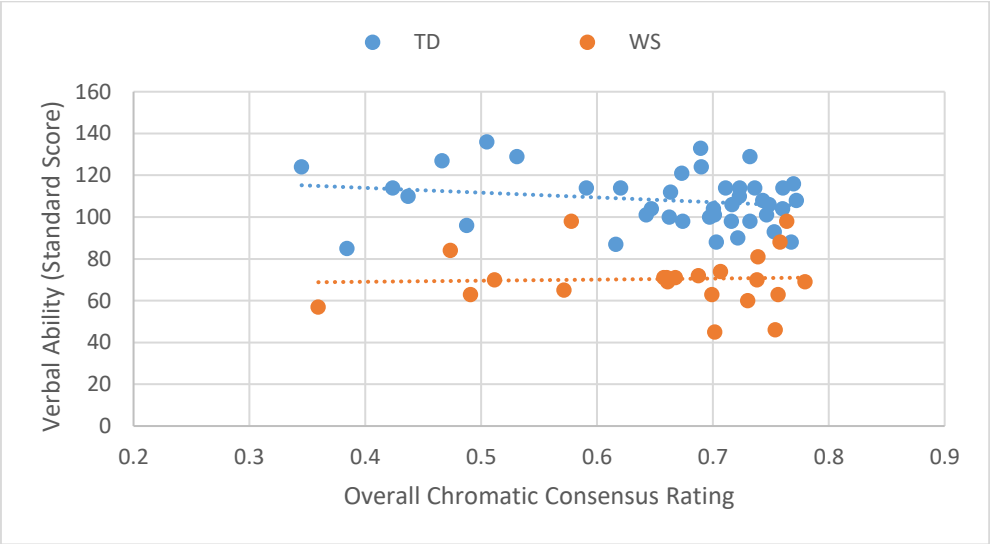



*Figure 5-6 - Overall chromatic concordance rating plotted against verbal ability. The dotted lines show the least squares fit for both TD and Williams syndrome groups.*

### 5.4.5 Relationship between Colour Naming and Chromatic discrimination

Correlations were carried out between performance on the CCDT (see Chapter 3) and type of colour term use and concordance ratings. Significant correlations were found between chromatic discrimination and frequency of non-BCT use in the TD group, $r$=-0.43, $p$<0.005. No other correlations were significant in the TD group (lowest $p$=0.516). For the Williams syndrome, there were no significant correlations between CCDT thresholds with overall and group achromatic concordance rating (lowest $p$=0.133).

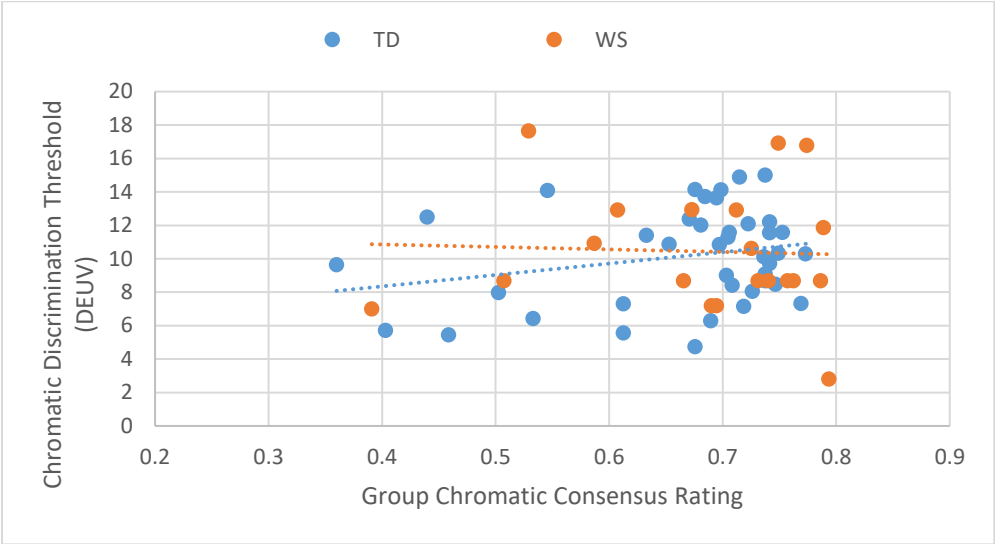



*Figure 5-7: Group chromatic concordance rating plotted against Chromatic Discrimination thresholds. The dotted lines show the least squares fit for both TD and Williams syndrome groups.*

### 5.4.6 Relationship between Colour Naming and Chronological age

For the TD group, there were no significant correlations between chronological age and frequency of singular or modified BCTs. There was a significant correlation between chronological age and frequency of non-BCTs, $r$=0.52, $p$<0.001. In the Williams syndrome group, there was a significant negative correlation between chronological age and frequency of BCT, $r$=-0.51, $p$<0.05, and a significant positive correlation between total modified BCTs and chronological age, $r$=0.48, $p$<0.05. No other correlations were significant (lowest $p$=0.182). There were no significant correlations between any concordance rating and chronological age in either TD (lowest $p$=0.35 or Williams syndrome (lowest $p$=0.14) groups.

### 5.4.7 "Favourite" Colour Concordance between groups.

The type of colour term used to describe the participant's favourite colour was examined between groups. This was the same colour that was chosen in chapter 4. Group and overall concordance ratings were compared between TD and Williams syndrome groups. A one sample t-test was used to assess whether there were differences in group concordance ratings for the participant's favourite colour. The Williams syndrome group (mean = 0.84, S.D. = 0.23) had a higher average concordance rating for their favourite colour than the TD group (mean = 0.66, S.D. = 0.39), $t(61)$=1.84, $p$<0.05. For the overall concordance rating the Williams syndrome group (mean = 0.86, S.D. = 0.2) again showed higher concordance rating than the TD group (mean =0.68, S.D. = 0.36), $t(61)$=1.84, $p$<0.05.

### 5.4.9 Parental Questionnaire: Colour Naming

The results of the Williams syndrome naming data was supplemented by parental questionnaire results about their child's colour naming. This section of the included questions relating to: the age at which colour terms were reliably learnt, examples of elaborate or impoverished colour learning (See

Chapters 6 and 7 for more details). The results in this section are for the Williams syndrome group only, one participant from the Williams syndrome group did not complete the questionnaire. Twenty participants of the Williams syndrome group were reported by their parents to name colours reliably, of these 16 parents could give a specific age at which this occurred. The mean age that these sixteen participants could reliably name was 5.68 years (Standard Deviation = 1.96 years). Parents also reported that only two participants would give unusual colour names. These two participants were also reported to not name colours reliably by their parents. Three parents said that their child would use elaborate colour terms, whilst eight parents said that their children's colour terms had basic colour vocabulary. To further explore correlations were conducted between the differences the age at which colour terms were reliably named and age at testing with the concordance ratings or the type of colour term used in the TD and Williams syndrome participants who reported their child to have reliable colour naming. There was no significant correlation with either overall concordance, chromatic concordance or achromatic concordance (lowest $p$=0.303). There were also no significant correlations with BCT use ($r$ = -0.486, $p$ = 0.048), use of modified BCTS ($r$ = 0.538, $p$ = 0=0.026), or non-BCT use ($r$ = 0.091, $p$ = 0.728).

## 5.5 Discussion

The results show that colour naming is atypical in Williams syndrome relative to TD controls. Both groups used BCTs more than non-BCTs. However, there was less concordance in colour naming, the achromatic colours, in the Williams syndrome group. Different constraining factors between groups were associated with colour naming. In the TD group; verbal ability, chronological age and chromatic discrimination thresholds were associated with more complex colour naming. Whilst in the Williams syndrome group there were no significant correlations with any constraining factors.

Like previous research on both child and adult colour naming studies, singular BCTs were the most frequent type of colour name given by participants in all groups (e.g. (Mylonas & MacDonald, 2015; Pitchford & Mullen, 2001; Wagner et al., 2013)). All eleven BCTs were used more frequently than non-BCTs and multiple term BCTs, a pattern that was observed in all groups. However, this masks more nuanced differences in frequency of BCTs use in the Williams syndrome group. In this group, only three BCTs (Green, Blue and Pink) were used by all participants, other BCTs were used by most Williams syndrome participants but to varying degrees. This was most pronounced for grey, where this term was used significantly less than in the TD group. The use of non-BCTs by participants in this study also mirrored that found in adult naming studies, e.g. (Mylonas & MacDonald, 2015), where the two most common non-BCTs reported were turquoise and peach used by both Williams syndrome and TD groups. This suggests that these two colour terms may be the first non-BCTs to develop in young native English speaking children. Although only turquoise was seen in the autism group as the most common non-BCT, but this is likely to be due to the reduced number of

participants in that group. The greater increase of frequency of both peach and turquoise colour term use in younger TD and Williams syndrome populations over other reported “common” non-BCTs (e.g. mauve/lilac, cream, maroon etc.) suggest that these non-BCTs may develop earlier before other non-BCTs. The use of the turquoise term was also the most common non-BCT in the autism group, even with the greatly reduced numbers, although the low numbers of participants in this group may mean that this needs to be replicated with a larger number of participants with autism. It is also possible to note that the MacBeth ColorChecker Chart does not sample all regions of colour space equally. It is possible that the twenty-four patches on the ColorChecker do not give prototypical versions of other non-BCTs that are also more commonly suggested (e.g. cream, maroon, lilac etc). To fully test this, future studies should use prototypical colours from each of these non-BCTs to see whether there is the same frequency of colour terms.

Similar concordance ratings were seen between the TD and Williams syndrome groups for across all patches and all the chromatic patches. However, this masked subtler differences in colour naming between the two groups. There were group differences in the concordance ratings of different coloured patches. The Williams syndrome group showed less within group concordance (more variability) in naming achromatic colours. Lower concordance was also seen for colours that were less saturated. This reduced concordance was caused by the Williams syndrome group using a wider range of terms to name these colours. Taken together with the reduced naming concordance of achromatic colours in general, these results indicate that the individuals with Williams syndrome may struggle to name colours that are not prototypical BCTs (except for grey). This reduced naming could be an example of over-extension errors seen when typically developing children learn vocabulary. However, it is not possible to separate out whether the chromatic colours are at the edge of a colour boundary. Unfortunately, it is not possible to map out where individual colour category boundaries are due to the limited number of stimuli in the Macbeth ColorChecker Chart. The exception to this is for the colour “Magenta”. For this colour the Williams syndrome group showed increased concordance (less within group variability). In this case however, the increase is due to a reduced colour lexicon compared to the TD group. For greyscale where the Williams syndrome group showed reduced frequency in using “grey”, and less concordance in how to name the achromatic patches. It has been suggested that grey is one of the last BCTs to develop in typically developing children (Pitchford & Mullen, 2005). This may be why Williams syndrome group struggle with the naming of this colour. This is supported by the Williams syndrome group showing significantly less concordance for other non-BCTs but crucially not for BCTs where there are prototypical exemplars of these colours in the MacBeth ColorChecker Chart.

Differences in colour naming are not due to low level perceptual factors. As seen in chapter 3, there was no difference on CCDT performance between Williams syndrome and TD controls. Despite this,

naming complexity and concordance in colour naming was differentially correlated with CCDT thresholds between the TD and Williams syndrome groups. Existing research has highlighted the importance of chromatic discrimination in colour naming TD children (Pitchford & Mullen, 2001, 2002, 2005). However, those studies used a coarse measure of chromatic discrimination where judgements were made on the basis of whether a colour was within the same or different colour category. Here a psychophysical study was used to measure chromatic discrimination, which gives a more accurate measure chromatic discrimination. Despite these differences however there was a similar result whereby enhanced chromatic discrimination led to more complex naming in the TD group. Although this relationship is not found in adult colour naming studies (Brown et al., 2011; Saunders & Van Brakel, 1999), suggesting that chromatic discrimination may only be a constraining factor for colour naming in children rather than adults. No such relationship between chromatic discrimination and complexity/concordance of colour naming was found in the Williams syndrome group. This is different for spatial language use, where deficits in spatial language are in line with visuospatial deficits found in Williams syndrome (Mervis & John, 2008, 2010, 2012). It is not possible to determine whether the difficulties in colour naming are due to this deficit in visuospatial ability in general, since no measure of visuospatial ability was taken. Studies in typically developing children have found that the ability to abstract relational concepts is delayed in children (Bornstein, 1985; Pitchford & Mullen, 2001; Rice, 1980; Sandhofer & Smith, 1999). Although these studies use no measure of visuospatial ability or sensory discrimination, so it is unclear whether the late development of naming all abstract relational concepts has the same underlying sensory process or is due to cognitive maturation. This is even the case where abstract relational language has been looked at in Williams syndrome (Mervis & John, 2010, 2012), visuospatial performance is not assessed in the Williams syndrome participants. It may be that these studies are not sensitive enough to capture the variability in visuospatial ability in Williams syndrome and whether this variability is related to abstract relational language use in Williams syndrome. Colour naming in the Williams syndrome group was found to be delayed in comparison to TD children, but the age at which reliable colour naming develops was not associated with performance. In combination with individual's difficulty in using spatial terms, the results here give further evidence for later general development of abstract relational concepts in Williams syndrome that is not in line with their mental age, but is in line with the cognitive profile of Williams syndrome.

The changes in BCT and non-BCT frequency with respect to chronological age also support previous research. In the TD group increased non-BCT was significantly correlated with an increase in chronological age. The Williams syndrome group showed a reduction in frequency of BCT use was correlated with an increase in chronological age. There was also an increase in use of modified BCTs was associated with chronological age. This increase in the use of more complex colour names, either

through non-BCTs or through modified terms, seen in both the TD and Williams syndrome groups is in line with previous research finding that the complexity of colour terms increases with age (Pitchford & Mullen, 2001).

Individuals with Williams syndrome have been shown to have delayed learning and deficits in using abstract relational concepts (e.g. space and number). This has been linked to the visuospatial deficits associated with Williams syndrome (Mervis & John, 2010, 2012; Phillips et al., 2004). Visuospatial perception is primarily associated with dorsal stream function, whilst the results here suggest that there is also difficulty in representation of colour as an abstract relational knowledge space that is predominately associated with ventral function. In this study, there was no correlation between chromatic discrimination and concordance rating for either group. There was a correlation between chromatic discrimination and relative complexity of colour term, giving additional evidence for the relationship between perceptual abilities constraining the development of abstract relational language in both TD and Williams syndrome groups. Previous research by Pitchford and Mullen (2005) has found an association between chromatic discrimination and colour naming have found the relationship in younger children (2-5 years). Taken together the results from this study and Pitchford and Mullen (2005) suggest colour naming is more constrained by chromatic discrimination and verbal ability in younger children, however the strength of association between chromatic discrimination and colour naming changes reduces as individuals get older. This notion also fits in with cross cultural studies (Franklin et al., 2005) where initial perceptual categories are “warped” with the reliable acquisition of colour terms.

In the autism group, every participant used all 11 BCTs suggesting that colour naming may be typical for individuals with autism. This is in line with previous research where individuals with autism have been shown to be able to categorise objects on the basis of colour (e.g. (Ludlow et al., 2006; Ungerer & Sigman, 1987)) but also display an appropriate retrieval of colour names as a semantic category (Boucher, 1988). This study however used more controlled and a wider range of colour stimuli than those used in previous studies, and explicitly show that individuals with autism can appropriately name colours on a free-naming task. In this group, there was an increase in the relative frequency of modified BCT use to name colours in this group compared to both the TD and Williams syndrome group. Unfortunately, due to the small sample size it is not known whether this difference would still be found with a much larger sample. For the same reason, it is not possible to assess the extent to which constraining factors (e.g. verbal ability, chronological age) may affect colour naming. For example, increased frequency of modified BCTs is associated with an increase in chronological age. The autism group is on average older than the Williams syndrome and TD groups and so the difference in colour naming complexity may be simply due to the fact the autism group are older.

Wide inter-individual variation was observed in all participant groups but this variation was not necessarily present for all colours. There were some colours that had high levels of concordance in their naming across the TD and Williams syndrome groups (e.g. white, red, yellow, blue, green and orange), but some colours had a low level of concordance rating across both TD and Williams syndrome groups and overall and group concordance ratings. These were predominately desaturated colours (Colour patches: “Light Skin” and “Blue Flower”) and the “Bluish green”. The “Bluish green” patch variation in naming was due to participants naming this patch either blue, green or turquoise. This variation between these two BCTs (blue and green) and the most common non-BCT (Turquoise) may suggest a developmental chronological age and verbal ability where the division of the blue-green region of colour space into a new colour category of turquoise may occur. Likewise, for the “Light Skin” patch where the most common terms were brown, orange and peach. This wide variation could again reflect the development of peach as a separate colour category from orange and brown. The “Blue Flower” and “Light Skin” patches are both relatively desaturated colours compared to the other chromatic colours in the ColorChecker chart. The wide variation found here may therefore also represent a more general difficulty in giving names to colours that are not in a prototypical BCT category. This can be seen in the wide variation of colour terms but also relative increase in non-BCTs to describe these colours.

### 5.5.1 Limitations:

A limitation of the free naming method is that the size of an individual’s colour lexicon was not measured. Previous research has suggested that prior to reliable colour naming children have a conceptual category of colour (Kowalski & Zimiles, 2006; Pitchford & Mullen, 2003). It is currently unknown whether the size or variability of colour terms in this pre-reliable colour naming phase affects the speed at which colour naming develops, and the extent to which the disruption of this pre-reliable colour naming causes delay or deviancy for the subsequent development of reliable colour naming, and how this may affect individuals with Williams syndrome. Atypical language acquisition could potentially cause disruption to the development of pre-reliable colour names or colour as a separate semantic category. Adolescents and young adults with Williams syndrome generate words from semantic categories in a different order compared to a moderate learning disability group, suggesting that their semantic category organisation is less structured compared to a verbal matched moderate learning disability group (Jarrold, Hartley, Phillips, & Baddeley, 2000). It is possible that the Williams syndrome group may give inappropriate colour names due to difficulty in recalling information from their semantic categories. It may also be that the participants with Williams syndrome have a different semantic structure of their colour lexicon or that they may not be able to reliably use the correct colour term when retrieving a colour term from their long term memory. Furthermore, the size of colour lexicon may be different in Williams syndrome compared to

mental age controls. Whether the Williams syndrome group show difficulty in recalling objects from semantic categories or naming abstract spaces in general is unknown. Previous work on visuospatial naming has found that there was an association in ability to use visuospatial terms and visuospatial abilities in Williams syndrome participants. However, in this study difference in colour naming is found but this is not related to colour discrimination ability, suggesting that naming different abstract spaces is generally atypical in Williams syndrome but that the nature underlying that atypicality differs with the abstract space, where visuospatial ability underlies ability of naming of abstract spatial concepts but other cognitive factors (not verbal ability) or chronological age underpin colour naming.

A related issue is colour naming consistency. This is the extent to which there is homogeneity of colour naming over time. On a word generation task, young adults with Williams syndrome were shown to generate words in an unusual order but that overall the content of the generated words was the same as verbal ability control group (Jarrold et al., 2000). It is possible that the Williams syndrome group were less likely to correctly retrieve an appropriate colour term from their long term memory. However, the colours terms given by the Williams syndrome group were plausible names with only one incidence where a participant didn't give a colour name. When there was less concordance in naming it is for colours that were far from good exemplars or close to a colour boundary. Other incidences of differences in colour naming concordance were through reduced colour lexicon in the Williams syndrome group. For example, for the "Bluish-Green" patch the Williams syndrome group were less likely to use turquoise, instead they were more likely to use either green or blue. Whereas the TD group were more likely to use all three terms. These all suggest that the atypical colour naming seen in the Williams syndrome group is not due atypical retrieval of colour names from long term memory or a failure to comprehend the task.

## 5.6 Conclusion

Colour naming is atypical in Williams syndrome but not autism. There was lower concordance for naming achromatic and less saturated colours in the Williams syndrome group. This difference is not due to a difference in chromatic discrimination ability between Williams syndrome and mental age equivalent TD groups (see chapter 3). Instead other constraining factors were identified, such as chromatic discrimination and chronological age, which are more important for colour naming in Williams syndrome, suggesting that different factors influence colour naming in Williams syndrome. In the autism group, all BCTs were used suggesting that colour naming using these BCTs is intact in adolescents with autism. Taken together this shows a dissociation between the autism and Williams syndrome, relative to typically developing children, in colour naming in the frequency of BCT and the consensus between participants for desaturated colours.

# Chapter 6 – Parent Reported Colour Affected Behaviours

## 6.1 Overview

Chapters 3, 4 and 5 have examined colour discrimination, colour preference and colour naming using rigorous experimental procedures. However, these findings may not necessarily reflect how individuals respond to colour outside of experimental conditions. Colour affected behaviours refer to behaviours of individuals which is prompted or associated with a response to colour. This chapter will address this issue using parental questionnaires to complement the results of the experimental chapters. The questionnaires consist of both structured and unstructured questions with free text opportunities for parents to describe their child's behaviour. This allows for comparisons of colour affected behaviours between typically developing children with and without either autism or WS. A selection of case studies will also illustrate potential links between parent reported colour affected behaviours and performance on experimental tasks already outlined experiments as reported in the thesis Chapters 3, 4 and 5. Therefore an individual's response to colour is compared across a mixture of psychophysical, cognitive experimental tasks and questionnaires. The chapter highlights the benefits of using a mixed-methods approach to study sensory processing to gain further insight about the relationship between an individual's colour perception measured on different tasks.

## 6.2 Introduction

### 6.2.1 Background to questionnaires

#### *6.2.1.1 Sensory Profile and Short Sensory Profile*

A small number of authors have published questionnaires for the assessment of different aspects of sensory processing. One of the most commonly used (especially by occupational therapists) is the Sensory Profile (SP) (Dunn, 1999). The SP is a questionnaire that is completed by parent/carer about their child's sensory processing, with separate sub sections that relate to each sensory modality (vision, auditory, tactile, olfaction and taste). The scores for each participant (the reported child) can then be classified along two different dimensions, Neurological Threshold and Behavioural Response. Neurological Threshold is posited by the author as representing the underlying biological response to the stimuli. With a high threshold said to represent a habituation to the sensory stimuli, and a low threshold said to denote sensitization to sensory information. The Behavioural Response is defined as the way that the participant acts either in accordance or discordance to their Neurological Threshold. Using a combination of these scores, the author reports that an individual's response to sensory information can be allocated into one of the following four different categories: Poor Registration, Sensation Seeking, Sensitivity to Stimuli and Sensation Avoiding (see Figure 6-1).

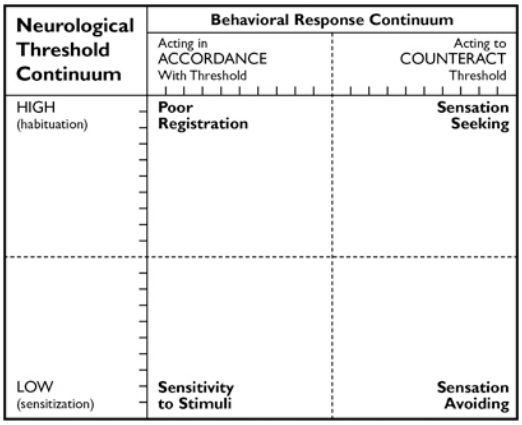



*Figure 6-1- Sensory processing as assessed by the Sensory Profile. Different questions load onto different quadrants, enabling an individual's sensory processing to be classified. Picture adapted from Dunn (1999).*

In the original publication, Dunn (1999) reported several aspects of the psychometric properties of the SP. These included Cronbach's alphas of 0.47 to 0.91 across the different subsections, (indicating poor to very good internal consistency). The Cronbach's alpha for visual items was 0.748. The factor structure was supported by internal correlations ranging between 0.25 – 0.76 suggesting that the Sensory Profile has good internal consistency across different factors, and that the different factors are independent from each other (Dunn, 1999). Content validity was assessed during multiple iterations during the development phase of the SP. This included a literature review by eight experienced sensory integration therapists. Once the set of questions were finalised they were categorised into separate set categories (i.e. each sensory modality) by 155 sensory integration occupational therapists who were not involved in the development of the SP. There was good agreement between these therapists on the selection of the individual questions into individual categories (Dunn, 1999).

However there has been little independent evaluation of the SP. One study published in the same year as the Sensory Profile, reported an independent evaluation of the construct validity using the School Function Assessment, a questionnaire of student's school performance including social and sensory issues (Coster, Mancini, & Ludlow, 1999). There was good correlation between different subscales on both questionnaires. The two subsections that correlated highly were related to sub scores around fine sensorimotor behaviour and behaviour modulation. Yet it is unclear whether this correlation between the School Function Assessment and the SP mean that the SP captures sensory processing or whether it reflects more general atypical behaviour. A more comprehensive assessment of content validity would be to compare SP scores with other sensory questionnaires.

In the field of autism research and practice, the Sensory Profile is the most widely used sensory questionnaire (Dunn, 1997; Green & Ben-Sasson, 2010; Green et al., 2012; Green et al., 2015; Green et al., 2013; Kern et al., 2006; Lidstone et al., 2014; Rogers et al., 2003; Tomchek & Dunn, 2007;

Wigham et al., 2015). Interestingly these studies highlight high inter individual variation of sensory processing between different individuals with autism, where there are commonly individuals located within each of the four quadrants seen in Figure one (for wider discussion see Chapter 1 sections 1.1.1 and 1.2.1). There is also variability between individuals with autism for the sensitivities of different sensory modalities. However, it is not possible to place each sensory modality into each quadrant, meaning that the location of an individual into one of the quadrants is taken from the sum of each sensory modality.

The Sensory Profile has also been used with individuals with WS (Janes et al., 2014; Riby et al., 2013). The results found that individuals with WS aged between 5 and 15 years showed variation in their sensory profile but that visual sensitivity was less affected compared to other sensory modalities such as proprioception, gustatory and vestibular processing.

It is unclear the extent to which the Sensory Profile reflects physiological, neurological or sensory processing thresholds of the individual. Surprisingly very few studies have investigated the relationship between sensory processing as measured by the Sensory Profile and performance on psychophysical tasks. Instead most studies have compared Sensory Profile scores with scores using behavioural questionnaires, such as anxiety or repetitive behaviours (Green & Ben-Sasson, 2010; Green et al., 2012; Janes et al., 2014; Lidstone et al., 2014; Riby et al., 2013; Wigham et al., 2015).

There is one study that has compared auditory discrimination (as measured by a frequency discrimination task) with auditory behaviours as defined using the SP in adults with autism. Jones and colleagues (2009) split performance for their autism group on their auditory discrimination task around the mean giving two groups of participants, those defined as having relatively good/poor discrimination. They then assessed scores on the Sensory Profile for their autism group. They found that participants who had lower auditory discrimination thresholds had higher scores on auditory items on the sensory profile (Jones et al., 2009). This suggests that better auditory discrimination leads to more auditory affected behaviours. However, it is not clear whether these individuals have higher overall sensory affected behaviours or whether this increased sensory discrimination leading to an increase in sensory (auditory) affected behaviours is associated with other sensory modalities. Nor is it possible to identify the direction of causality between sensory discrimination and different types of sensory affected behaviours. Further research is needed assessing psychophysical performance for all sensory modalities and the relationships with specific types of sensory behaviours.

It is also unclear the extent to which the Sensory Profile reflects underlying cortical activation. Currently there are only two studies that have attempted to link scores on the Sensory Profile with neuroimaging data. Ludlow and colleagues (2014) investigated Sensory Profile scores using an ERP

mismatch negativity paradigm of meaningful and nonsense speech. Using 11 high-functioning adults with autism they found a reduced neural response to both meaningful and nonsense speech in the autism group compared to IQ and chronological age matched controls. The authors also conducted a preliminary analysis to investigate whether sensory processing predicted the mismatch negativity response for frontal or central-parietal scalp regions. Bootstrapping with replacement was used to boost the number of participants (5000 replacements with 95% confidence levels). They found that only scores for the sensory sensitivity quadrant predicted mismatch negativity across frontal electrodes only (Ludlow et al., 2014). However, caution should be used when interpreting the results of this study. It is important to note that this study used bootstrapping for many iterations on a small sample on a new task in a population group that show high inter individual variability in sensory processing and neural responses. It is not clear the extent to which the participant group are representative of a wider sample of autism. The size of the effect also needs to be considered. Even with the multiple bootstrapping the effect size is still relatively small.

In a separate study, Green and colleagues (2015) found a relationship between primary sensory cortices activation and sensory sensitivity as measured by the sensory profile. The authors hypothesised that sensory over-responsivity, as measured by the Sensory Profile, would be reflected by reduced habituation to sensory stimuli in primary sensory cortices. To test this, they used a composite sensory score across all five sensory modalities and correlated this with functional brain anatomy in adolescents with autism. They found that there was a significant positive correlation between sensory over-responsivity and cortical excitability in primary auditory and somatosensory cortices in response to low level auditory, tactile and joined sensory stimuli, whilst using anxiety measures as a covariate (Green et al., 2015), as previous research has hypothesised a link between anxiety and sensory processing (Lidstone et al., 2014; Wigham et al., 2015). However, this study has a few limitations. First the auditory and tactile stimuli were selected based on a prior study and were aimed to maximally differentiate between individuals with and without autism. Therefore, the results need to be replicated with neutral stimuli and for other sensory modalities. Secondly it is not reported whether the individual sub scores on auditory and tactile items on the sensory profiles also correlate with primary auditory and somatosensory cortices. By using a composite score, sensory over responsivity scores may be conflated by other senses that were not assessed in the study. Further inspection of the raw data seems to suggest that the correlation is primarily increased by a small sub group (four out the nineteen participants) who show increased sensory responsivity and cortical activation. It is not clear whether those participants who are not at the extreme scores also show a similar relationship between sensory over responsivity and cortical activity.

These studies support a possible relationship between scores on the Sensory Profile and neural activity, however because of both the limited number of studies and methodological uncertainties

about how best to investigate this relationship, remains difficult to know how best to conceptualise and ascertain the nature of this relationship.

There has also been no research as far as this researcher is aware, investigating a potential relationship between the Sensory Profile responsivity scores and other physiological responses (electrodermal activity). This is perhaps surprising in the light of the replicated finding of differences in physiological responses between young adolescents with and without autism (McCormick et al., 2014; Schoen, Miller, Brett-Green, & Nielsen, 2009). This latter finding suggests that it may be important to distinguish between physiological responses and other measures of sensory affected behaviours, though it is not clear whether there is relation in younger or older participants. Further although inspection of individual items on the Sensory Profile suggest that they measure affected behaviours with a sensory aspect (for example, "*Touches people or objects"* (Q18 Short Sensory Profile), the interpretation of the underpinning c*ause* of the type of response is unclear. Using Q18 as an exemplar- the sensory affected behaviour might be a consequence of either hyper or hypo responsivity to the tactile sensation. Several authors have also identified a range of other behaviours which might be a consequence of either hyper or hypo sensitivity. For example, "*Reacts emotionally or aggressively to touch*" (Q4 Short Sensory Profile). These questions are taken from the Short Sensory Profile but are representative of questions in the SP. In this case an inappropriate response could be caused by a hyper response of increased sensitivity to touch, or alternatively it could be a hypo response, where the touch does not elicit an expected response because of decreased sensitivity. These concerns highlight that the lack of specificity to enable a distinction to be made between the hypothesised nervous system hyper and hypo sensory responses as originally proposed by Dunn (1999). Instead the Sensory Profile measures different behaviours, some of which can be inferred to be related to a sensory response in a particular sensory modality.

Further it is still not established whether these sensory behaviours as measured by the Sensory Profile are related to psychophysical thresholds (see Chapter 2 section 2.5). Psychophysical thresholds may act as a marker of physiological reactivity, for example colour discrimination experiments reflect specifically defined neurological pathways from the retinal ganglion cells to LGN and the cortex (see also Chapter 1 section 3 for more details). Therefore, they could represent a missing intermediate step between the neurological activity and more sensory affected behaviours. Further understanding of possible relationships between the neurological system, psychophysics and sensory affected behaviours is likely to provide a mechanism for gaining a greater understanding of sensory processing in individuals including those with autism and WS.

Finally, and of most relevance to this thesis, a major limitation of the sensory profile is that the SP does not include any questions that focus specifically on responses to colour. There are numerous published anecdotal accounts (from high functioning individuals with autism and parents of children

with autism), that describe particular sensory experiences with clear accounts of how certain sensory stimuli can lead to definite changes in behaviour and mood; and how some sensations are experienced as causing physical discomfort /pain (Bogdashina, 2003, 2011; Grandin, 2009; Lawson, 1998; A. K. Ludlow et al., 2014; Williams, 1994). However, to date these accounts have not been investigated alongside either the use of questionnaires or performance on experimental tasks. Some of these anecdotal accounts include colour affected behaviours as well as differences in colour perception See Chapter 1 Section 3.1.1 and 3.2.1). Thus, the omission of colour from the Sensory Profile had important implications for the design of this research and is likely to mean that particular sensory sensitivities relating to colour are unlikely to be identified.

#### *6.2.1.2 Alternative Sensory Measures*

In response to the limitations of the sensory profile other sensory questionnaires have been created. This section will give a brief overview of two other recently developed sensory questionnaires. The Glasgow Sensory Questionnaire (Robertson, 2012; Robertson & Simmons, 2013) and the Sensory Processing Quotient (Tavassoli et al., 2014).

#### *6.2.1.3 Glasgow Sensory Questionnaire (GSQ)*

Like the Sensory Profile, the Glasgow Sensory Questionnaire is a parent questionnaire. Questions map onto the seven sensory modalities, with an equal number of questions that focus on hyper- and hypo-reactivity for each sensory modality. Hyper-sensitivity was defined as an “overload” for a type of sensory stimuli. Hypo-sensitivity was noted as being an under-reaction to some sensory stimuli, and that this may lead to sensory seeking behaviours. This procedure provides a more comprehensive coverage of this aspect of sensory processing than the Sensory Profile. To date there are few studies that have used the GSQ. One study investigated a general population sample of adults using the GSQ and the Autism Quotient (Robertson & Simmons, 2013). This study reported an increase in Autism Quotient Score was positively correlated with an increase in scores on the Glasgow Sensory Questionnaire, indicating that an increase in autistic traits was associated with an increase in both hyper and hypo sensory processing. Freyberg and colleagues (2015) investigated binocular rivalry in adults with autism. They found that there was a weak-medium positive correlation for all participants between scores on the visual subscale of the Glasgow Sensory Questionnaire and number of switches between images, but there was no correlation when comparing each individual group (Freyberg, Robertson, & Baron-Cohen, 2015). Although these findings will need replication including testing across other visual functions such as motion or colour perception, they suggest that the GSQ visual questions may relate to existing knowledge of aspects of visual functions.

The authors report some aspects of the psychometric properties of the GSQ, including a good level of reliability for all items (Cronbach’s alpha = 0.935). Unfortunately, reliability was not reported for

individual sensory modalities. The questions were also independently verified by five other academic staff who agreed that the questions measured sensory processing. To date there is no data available comparing the use of the Glasgow Sensory Questionnaire with other measures such as the SP.

However, the Glasgow Sensory Questionnaire has several limitations in common with the Sensory Profile. First, there are no specific questions that assess responses to colour. Although reference is made to colour as an example of an "obsession over a visual stimulus", colour is combined with sparkling lights and mirrors. Furthermore, it only includes hyper responses to colour, and makes no reference to aversive responses to colour which have been stated by anecdotal accounts of parents or high functioning individuals with autism. (Bogdashina, 2003, 2011; Lawson, 1998; Williams, 1994).

*6.2.1.4 Sensory Perception Quotient (SPQ)*

The Sensory Perception Quotient has been designed as a self-report measure of sensory processing in adults (Tavassoli et al., 2014). The questionnaire assesses all sensory modalities and like the Glasgow Sensory Questionnaire it makes explicit distinction between hyper and hypo sensory responses for each sensory modality. Unlike the Sensory Profile and the Glasgow Sensory Questionnaire, further distinctions are made for each modality. For example, the visual questions are divided into separate questions on acuity, brightness, motion and colour. This expansion of topics provides an opportunity for the Sensory Perception Quotient scores to be mapped onto (and compared with) the underlying sensory functions for each sensory modality, meaning that there is potential for greater specificity in identifying where atypicalities of sensory processing may lie. The Sensory Perception Quotients reliability and validity was assessed using a large sample of adults diagnosed with autism (n = 196) and adults without autism (n = 163). The questionnaire has good internal reliability (Cronbach's Alpha = 0.92), although again this was not reported for each sensory modality or sub modality. Nonetheless scores on the Sensory Perception Quotient have been shown to correlate with the scores on the adult Sensory Profile scores (Tavassoli et al., 2014), suggesting that there is good concurrent validity between the two questionnaires. Tavassoli and colleagues (2014) compared performance on the Sensory Processing Quotient and Autism Quotient between adults with and without autism. The authors reported a positive correlation between SPQ and AQ in both groups, replicating a similar finding using the Glasgow Sensory Questionnaire (Robertson & Simmons, 2013). They also found that there was an increase in scores on the Sensory Processing Quotient in adults who have autism compared to those who did not.

As a new measure of sensory processing, there are still some unknowns regarding the Sensory Processing Quotient. First, although the questionnaire is designed to investigate the different functions within each sensory domain, it is unclear at present whether scores on these questions do map onto the results of psychophysical or neural processing testing of these sub domains, i.e. does chromatic discrimination map onto the colour questions. Secondly closer inspection of colour

questions indicates that the primary focus of the questions is on the ability to match colours together and a single question on preferring to wear muted colours. However other aspects relating to colour affected behaviour, such as the colours of walls, toys etc, are not included. This again suggests that potential aspects of colour affected behaviours are likely to be missed. There is also no reported selection criteria for these questions over other aspects of colour perception.

### 6.2.2 Sensory Questionnaires Summary

This section has outlined three sensory processing questionnaires. The Sensory Profile is the most commonly used measure, but it remains unclear whether it is a valid measurement of sensory processing rather than just sensory affected behaviours. Two newer questionnaires have different limitations. The Glasgow Sensory Questionnaire and Sensory Processing Quotient both make more rigorous attempts to capture hyper and hypo sensory processing across a range of sensory modalities. However, for both questionnaires it is unclear how well they map onto psychophysical thresholds. Furthermore, in relation to this thesis, all three questionnaires do not capture colour affected behaviours. This omission meant that for the purposes of this research thesis, a bespoke parent report questionnaire would need to be developed to capture colour affected behaviours (see section 6.3 and Appendices 4 and 5). However, to identify how best to undertake this task, it was also necessary to review the existing literature to compare the benefits and limitations of using questionnaire and direct experimental testing methods to obtain reliable and comprehensive information about sensory processing. The next section will summarise this comparison.

### 6.2.3 Qualitative Studies on Sensory Experiences

Structured questionnaires can be useful to assess sensory processing in large numbers of participants/subjects; however, they are not able to assess how sensory atypicalities develop or the impact on the individual beyond the focus of the individual questions. The use of qualitative methods using interviews and open questions, provides an opportunity to explore in greater detail aspects of sensory processing. For example, through enabling the parent or child to describe their own experiences of sensory sensitivities. This work as far as this researcher is aware, has only been conducted in autism (Kirby et al., 2015; Robertson & Simmons, 2015). No analogous work having been identified in either WS or TD children.

In comparison to studies using questionnaires, the number of studies using qualitative methods to study sensory processing in autism is sparse. Kirby and colleagues (2015) conducted interviews with children and young adolescents with autism about their sensory experiences. These children had Sensory Profile scores that indicated that they had atypical sensory sensitivities. They found that sensory experiences were generally either seen to be positive or negative, but that the child's valence towards these sensory experiences also changed over time. For example, one child did not

like to have their hair brushed when they were younger as it was associated with pain. However, hair brushing changed to a positive experience when they were older because it was now associated with making the child look pretty. The authors also identified that children with autism will take a variety of precautionary steps to cope with their sensory sensitivities, such as deep breathing or choosing not to participate in an event. Some of the children also stated that their negative sensory experiences lead to uncontrollable physical responses such as itchiness, choking, vomiting or shaking (Kirby et al., 2015).

Similar results were found in adults with autism. Robertson and Simmons (2015) conducted a semi-structured interview with six adults who had autism. The adults described how particular aspects of sensory stimuli were either positive or negative and could induce definite changes in mental states. For example, one of the adults reported that "strong colours" were a problem and that they were distressing to deal with and "hurt". For some unpleasant sensory stimuli were described as strong enough to cause physical sensations, similar to those identified by children with autism (Kirby et al., 2015). This suggests that although the sensory experience may change in its valence over the lifespan for some individuals with autism, there is still a coupling between a negative sensory stimulus and a physical response. However, Robertson and Simmons (2015) also found that these negative impacts could be controlled if the adult with autism could take some control in advance over their environment (e.g. through the wearing of ear plugs), suggesting that there may be strategies that an individual can use to be better able to cope with such negative sensory stimuli. The researchers also identified that sensory stimuli and sensory sensitivities led to disruptions of behaviour (such as an adult having to leave the room or discontinue an activity). These themes are common across other qualitative studies of sensory processing in adults with autism (Jones, Quigney, & Huws, 2003; Robledo, Donnellan, & Strandt-Conroy, 2012; Smith & Sharp, 2013).

These studies are useful to gain insights into sensory behaviours and the detailed experience of individuals. However, these behavioural experiences have usually not been linked to either sensory questionnaires or behavioural results on experimental sensory tasks (e.g. psychophysical or cognitive tasks), making the results of these studies difficult to directly compare with the studies using quantitative questionnaires. Furthermore, due to the nature of qualitative methodology studies, they are usually conducted on a small number of participants. Given the highly heterogeneous nature of autism it is unclear whether the participants in these qualitative studies can be considered representative of their respective groups, and further whether they are representative of the larger samples of individuals recruited to studies that have reported questionnaire and/or other quantitative measures.

### 6.2.4 Summary Questionnaire and Interview studies on Sensory Processing

Both qualitative and quantitative studies of sensory processing in autism have shown highly variable sensory sensitivities, where some individuals show either increased or decreased sensitivity across one or more sensory domains. In WS there is also some reported evidence of differential sensory processing using quantitative measures, although no qualitative methods were identified. However, only using qualitative or quantitative methodological approaches to studying sensory processing leads to an incomplete account of the types of experiences of affected individuals. Furthermore, there have been limited attempts to link sensory processing measures to cognitive or psychophysical tasks. The combination of a mixed methods approach (including quantitative, qualitative and experimental methods) is likely to best progress our understanding of an individual's sensory processing, the processes that contribute to this and how best to characterise the sensory experiences.

### 6.2.5 Chapter Rationale and Aims

This chapter will utilise a combination of methods. A bespoke questionnaire was designed and used in this research to assess the frequency of colour affected behaviours. The questionnaire included a set of structured questions focussing on visual sensitivities (in line with the Sensory Profile) in combination with more open ended questions about colour affected behaviours. The results from the questionnaire will be presented and then be compared with results from the other experimental chapters (Chapters 3, 4 and 5). These comparisons will allow the exploration of possible relationships between low-level perceptual processing of colour (chromatic discrimination), cognitive uses of colour (colour reference and naming) and observable colour affected behaviour. In addition to this, individual case studies will also be presented to further illustrate the richness of the data available when the information from the different methods are combined. This chapter has three main aims:

1) The development of a new bespoke sensory processing questionnaire designed to capture colour affected behaviours and to assess the psychometric properties of the bespoke questionnaire.
2) To assess the frequency of colour affected behaviours and whether there are any characteristics that are specific to individuals who display such behaviours.
3) To investigate potential links between sensory processing and cognitive uses of colour across TD, autism and WS groups.

## 6.3 Method

### 6.3.1 Participants

Ninety-eight participants who took part in the study: twenty-one with autism; twenty-six children with WS; and fifty-one TD participants. The WS and autism participants were not individually

matched to TD participants due to not all questionnaires being returned. The response rate and participant demographics for those who completed questionnaires can be seen in table 6-1.

*Table 6-1 - Participant demographics for Questionnaire Data.* Chronological age is reported in years. Standard deviations are reported in brackets. The response rate is reported as a percentage of participants who returned both versions of the questionnaire of the total of participants who took part in the study.

| Group | Chronological Age | Males | Females | Verbal IQ | Non-Verbal IQ | RCPM | Response Rate (%) |
|---|---|---|---|---|---|---|---|
| Autism (n=21) | 12.9 (2.31) | 16 | 5 | 81.82 (21.10) | 87.45 (17.87) | 28.29 (6.13) | 80.77 |
| WS (n=26) | 12.65 (3.16) | 13 | 13 | 69.68 (14.01) | 54.04 (8.75) | 15.65 (4.39) | 96.3 |
| TD (n=51) | 6.61 (1.57) | 21 | 27 | 112.74 (13.14) | 107.56 (12.52) | 22.25 (6.39) | 66.23 |

### 6.3.2 Questionnaire Development

There were two versions of the questionnaire that were used in this study. This section will outline the initial development and then subsequent revisions of the first questionnaire. Both versions one and two of the questionnaire can be found in the appendices (appendix 4 and 5 respectively).

#### *6.3.2.1 Version 1: Basic Version*

The parental questionnaire was designed to identify colour affected behaviours (as reported by the parent). The initial basic version questionnaire included questions based on different colour affected behaviours. The colour affected actions were eating food, wearing clothes, toys played with and the colours of rooms. These different actions were chosen as they are the commonly reported behaviours that are affected by colours (see introduction). Each question consisted of two parts. The first part assessed whether the parent indicated that their child displayed that behaviour (Yes/No). The second part of the question provided a free text opportunity for the parent to expand on how that behaviour is displayed by their child. Further, each colour affected behaviour had two questions, relating separately to positive or negative experiences. This gave a maximum of eight possible colour affected actions (four behaviours that are both positive and negative). There were additional structured (Yes/No) items focussing on specific reactions to bright lights and “obsessions” with visual objects or spinning objects. There was also a section to identify a possible familial history of colour blindness. Parents of four participants with autism and one participant with WS completed this version of the questionnaire.

*6.3.2.2 Version 2: Expanded question set including Structured Visual questions.*

A decision was made early in the project to expand the parental questionnaire. The parents of all TD participants, 17 autism participants and 25 WS participants completed this version of the questionnaire. There were an additional in six topics: that were added to Version 2 of the questionnaire. These six topics were:

**(i) Basic visual functions**

Twenty-two structured questions to assess basic visual functions and the visual questions from the Sensory Profile were added. However, since the sensory profile does not cover all aspects of vision, questions on depth, stereopsis and visuo-motor function were designed by the researcher and added to the questionnaire. All questions were reported on a 5 point Likert scale ranging from always to infrequent displays of the behaviour. Parents were asked to report whether these behaviours were shown in the previous 6 months. However, if the behaviour had been shown prior to the previous 6 months then they were asked to indicate at what age this occurred. The scores on the visual questions were totalled separately from the sub-set of items derived from the Sensory Profile and across all basic visual question scores (both Sensory Profile questions and other basic visual function questions). Higher scores on the Sensory Profile Items or on all Basic Visual functions indicate greater atypicality in vision related behaviours. The highest possible score on Sensory Profile items was thirty-five, whilst the highest score for all basic visual functions was ninety.

**(ii). Colour affected actions questions**.

In addition to questions on food, clothing, rooms and toys, questions focussing on the skills of drawing and colouring/painting behaviours were added. These questions were more age appropriate for the TD participants, but also from anecdotal reports about intense interest in or aversions to colours in adults with autism.

**(iii)**. **Emotional responses to colour**

These questions asked whether parents thought their child had an emotional response to one or more colours. It includes both positive (happy, relaxing, and excited) and negative (sad, avoiding) emotional responses. This meant that there is a maximum score of five for this measure, with three emotions being positive and two emotions being negative.

**(iv) General aspects of colour perception**:

A second set of questions were designed to relate to two different higher order functions of colour perception. These were colour naming and colour preference.

**(v). Colour naming**

Seven new questions probed colour naming. This included reliability of colour naming and elaborate or impoverished colour vocabulary.

**(vi) Colour preference combinations**

Two further questions assessed whether there were different colour combinations that were either liked or disliked.

All the TD participants and 17 autism participants and 25 WS participants completed this version of the questionnaire.

### 6.3.3 Procedure

The questionnaires were completed by parents and in all cases parents were blind to the performance of their child on the experimental tasks. For participants who were tested at home/University, questionnaires were given to the parents whilst the child participated in the experimental tasks outlined in previous chapters. When participants were tested at school, the questionnaires were sent to parents after the completion of the experimental tasks and returned in the post. All TD participants were assessed at school, whilst all but one of the WS participants were tested at home with the other tested at school. The autism group were assessed either at home/University (n = 7) or at school (n = 14).

### 6.3.4 Data Analysis

The results section is split into several sections due to the different versions of the questionnaire and different approaches were used for the different subsections. Firstly, the psychometric properties of the questionnaire were assessed. After this all three groups were compared against each other to assess responses across the different parts of the questionnaire.

#### *6.3.4.1 Colour affected behaviours:*

Frequencies of colour affected behaviours were calculated from the sum of emotional responses to colour and colour affected actions. Separate calculations were then conducted for emotional responses and colour affected actions. All participants whose parents had completed version 2 of the questionnaire were included in the colour affected behaviours and emotional responses to colour analysis. Due to the overlap in questions between versions 1 and 2 for colour affected actions meant that all participants who had returned questionnaires could be included in the colour affected actions analysis. Where free text comments were provided by the parents, the colour (if mentioned) and how the behaviour is affected behaviour was also recorded. This information was included in the case studies. As in previous chapters the comparison between the autism and WS groups with the TD group will be of approximate mental age equivalents, despite the large differences in chronological age. Unlike previous chapters a direct comparison will be made between the autism and WS groups.

This is to assess syndrome specificity between the autism and WS groups for similar chronological age.

## 6.4 Results

### 6.4.1 Questionnaire Psychometric Properties

Face validity: During the piloting of the questionnaire, once parents had completed the questionnaire, they were asked if they had could understand both instructions that were given in the questionnaire and the structured and open-ended questions. Further examples of face validity came from parental comments, such as being able to distinguish problematic behaviours that do and do not originate because of colour (see case study number 4 for an example).

The internal reliability of the items was assessed separately for the structured questions and those items that included facility for parents to make free text contributions. Factor analysis was used to assess the underlying structure of the structured basic visual function questions. The results of Kaiser-Meyer-Olkin measure of Sampling Adequacy (0.685) and the results of the Bartlett's Test of Sphericity was also significant indicating the questionnaire data were suitable for exploratory factor analysis. Principal Components Analysis was used to split the questions onto different factors. Varimax rotation was used, with factor loadings constrained to 0.4. The Cronbach's alpha for all the questions was 0.76, indicating "good" internal reliability. This was like the Cronbach's alpha for the visual questions from the Sensory Profile (0.748). The basic visual function questions loaded onto five different factors. Factors were considered if they had an eigenvalue greater than one. From these five factors were identified; Visual Recognition, Lightness, Visual Attention, Visual Seeking and Visual Avoiding. Cronbach's alpha was conducted for each of these factors and is reported alongside item loadings in table 6-2.

*Table 6-7 – Initial factor loadings for each individual question item onto the five different factors. The Cronbach's alpha for each factor is also reported.*

| Question | Component | | | | |
|---|---|---|---|---|---|
| | Visual Recognition | Lightness | Visual attention | Visual Seeking | Visual Avoiding |
| 16. Does your child have difficulty reading words from a book or computer screen? | .754 | | | | |
| 12. Does your child have a hard time finding objects in competing backgrounds? | .729 | | | | |
| 13. Does your child have difficulty in identifying moving objects against a background? (e.g. a bird flying or moving car) | .706 | | | | |
| 10. Does your child look very carefully or intensely at objects/people? | .627 | | | | |
| 11. Does your child really like to look at one special object? | .611 | | | | |
| 15. Is your child bothered by visual changes in a room? | .538 | | | | .467 |
| 4. Does your child try to get away from bright lights? | | .879 | | | |
| 2. Do bright lights bother your child (even after having enough time to adjust to them)? | | .831 | | | |
| 7. Does your child prefer to be in the dark? | .412 | .575 | | | -.453 |
| 21. Does your child have difficulty seeing things near to them? | | | .768 | | |
| 22. Does your child have difficulty seeing things that are far away from them? | | | .715 | | |
| 20. When watching television or on the computer does your child sit near to the screen? | | | .623 | | |
| 14. Does your child notice visual changes in a room? (e.g. turning on/off a light) | | | -.552 | | .424 |
| 1. Does your child like bright lights? | | | | .834 | |

| | | | | | |
|---|---|---|---|---|---|
| 3. Does your child try to get near bright lights? | | | | .824 | |
| 5. Does your child enjoy watching spinning objects? | | | | .729 | |
| 7. Is your child happy to be in the dark? | | | | | -.833 |
| 9. Does your child cover their eyes or squint to protect their eyes from light? | | | | | .498 |
| Cronbach's Alpha | 0.788 | 0.745 | 0.513 | 0.735 | 0.171 |

The Cronbach's alpha for each factor suggested the removal of three individual items. These questions were; Question 6: "Does your child prefer to be in the dark?", Question 7: "Is your child happy to be in the dark?", and Question 14: "Does your child notice visual changes in a room? E.g. turning on/off a light)". These corresponded to items that loaded onto multiple items. The factor analysis was then conducted with the removal of these items using the same parameters as listed above on the same dataset. The Kaiser-Meyer-Olkin measure of Sampling Adequacy (0.746) and the results of the Bartlett's Test of Sphericity was also significant indicating the questionnaire data was again suitable for exploratory factor analysis. The Cronbach's alpha for the revised questions was 0.795. This is an improvement from the previous factor analysis with all basic visual function questions. This time there were four factors separated out which were; Visual Recognition, Visual Acuity, Visual Seeking and Visual Avoidance. The Cronbach's alpha for each of these factors ranged from 0.74 to 0.837, indicating acceptable to good internal consistency for the items. The psychometric properties of this version of the questionnaire can be found in table 6-3.

The psychometric properties of the colour affected behaviour questions were not assessed. This is due to the differences in the classes of the data between the two questionnaire sections. Instead the colour affected behaviours will be discussed with respect to the relevant frequencies across each participant group.

*Table 6-8- Revised factor loadings for each individual question item onto the four different factors. The Cronbach's alpha for each factor is also reported for the second iteration of the general visual function questions.*

| | Component | | | |
|---|---|---|---|---|
| | Visual Recognition | Visual Acuity | Visual Seeking | Visual Avoiding |
| 12. Does your child have a hard time finding objects in competing backgrounds? | .768 | | | |
| 13. Does your child have difficulty in identifying moving objects against a background? (e.g. a bird flying or moving car) | .743 | | | |
| 16. Does your child have difficulty reading words from a book or computer screen? | .719 | | | |
| 11. Does your child really like to look at one special object? | .709 | | | |
| 10. Does your child look very carefully or intensely at objects/people? | .608 | | | |
| 22. Does your child have difficulty seeing things that are far away from them? | | .832 | | |
| 21. Does your child have difficulty seeing things near to them? | | .787 | | |
| 20. When watching television or on the computer does your child sit near to the screen? | | .762 | | |
| 3. Does your child try to get near bright lights? | | | .849 | |
| 1. Does your child like bright lights? | | | .841 | |
| 5. Does your child enjoy watching spinning objects? | | | .733 | |
| 4. Does your child try to get away from bright lights? | | | | .896 |
| 2. Do bright lights bother your child (even after having enough time to adjust to them)? | | | | .859 |
| Cronbach's Alpha | 0.783 | 0.74 | 0.737 | 0.837 |

### 6.4.2 Questionnaire Results

#### *6.4.2.1 Basic Visual Function Questions*

The visual questions from the Sensory Profile were compared between groups. An independent samples t-test revealed the autism group had significantly higher scores than the TD group, $t$ (63) = 7.43, $p < 0.001$ (Figure 6- 2). There was also a significant difference for the Expanded Visual Question set, $t$ (63) = 8.083, $p < 0.001$. The WS group were also reported to have significantly higher scores than the TD group on the visual Sensory Profile items, $t$ (72) = 6.979, $p < 0.001$ (Figure 6-2). This was also the case for the Expanded Visual Question set, $t$ (72) = 7.965, $p < 0.001$. There were no significant differences between the autism and WS groups for either the sensory profile questions ($p$ = 0.827) or for all basic visual functions ($p$ = 0.334).

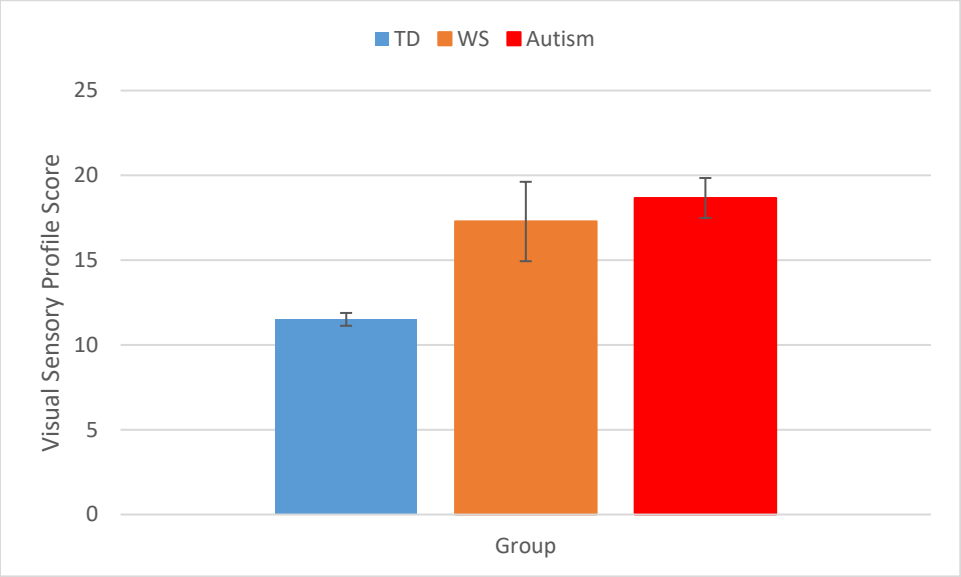



*Figure 6-2 – Mean scores on the visual questions of the Sensory Profile Questions.* The maximum score is 65. Colour Affected Behaviours

#### 6.4.2.2.3 Frequency of Colour Affected Behaviours

The total amount of different types of colour affected behaviours was compared between the autism and TD groups. There was significantly more colour affected behaviours in the autism group than the much younger TD group, $U$ = 342, $p$ <0.01. There was a significant increase in both positive responses to colour, $U$ = 287, $p < 0.001$, and negative responses to colours, $U$ = 348, $p < 0.05$. To further explore this, difference, colour affected behaviours were classified into either colour affected actions (e.g. wearing clothes of a colour) or emotional responses to colour. For both classifications, individuals with autism showed significantly increased colour affected actions compared to the TD group, $U$ = 302, $p < 0.005$, and increased emotional responses to colour, $U$ = 278, $p < 0.05$ (see figure 6-3). For colour affected actions the autism group displayed significantly higher positive actions, $U$ = 309, $p$ <

0.005, but not negative emotions, $U = 408$, $p = 0.146$. Further analysis was conducted to see whether there were differences in the groups for colour affected actions. Bonferroni corrections were made for multiple comparisons. There were significantly more affected behaviours for individuals with autism relating to coloured rooms than the TD group, $U = 275$, $p < 0.05$. There were no significant differences between the autism and TD groups for food, $p = 0.656$, clothes, $p = 0.248$, or toys, $p = 0.088$, colour affected behaviours (see Figure 6-4). For the WS group there were no differences in the total number of colour affected behaviours between the TD and WS groups, $U = 584$, $p = 0.537$. This was also the case when behaviours were divided into colour affected actions, $U = 523$, $p = 0.102$, and emotional responses, $U = 561$, $p = 0.329$. Furthermore, there was no difference in either positive or negative actions or emotions, lowest $p = 0.135$ (see Figure 6-3).

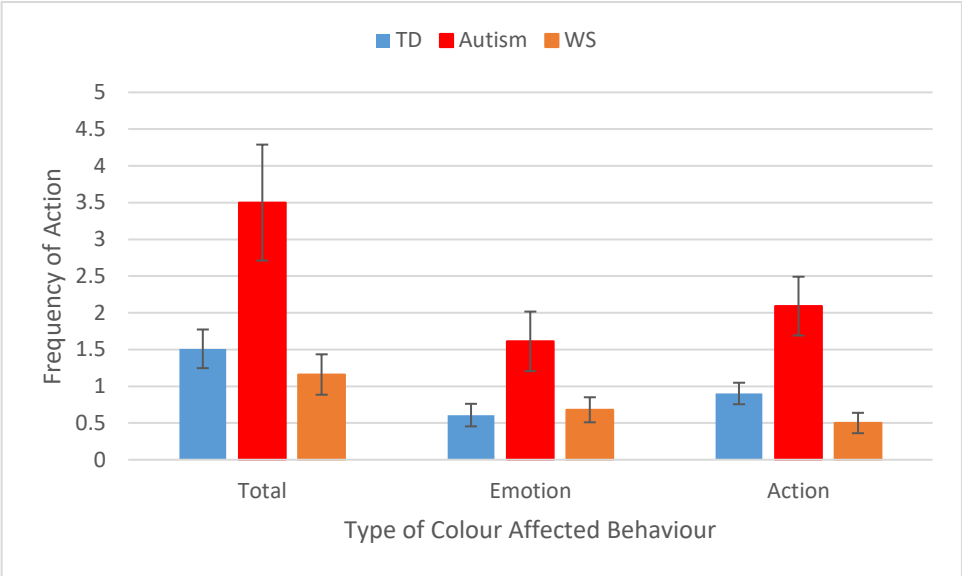
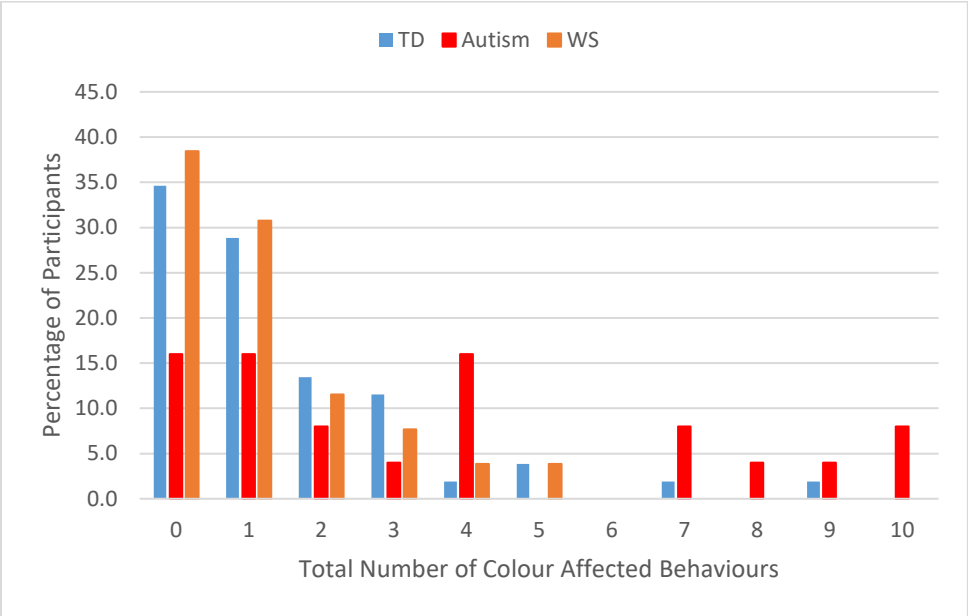
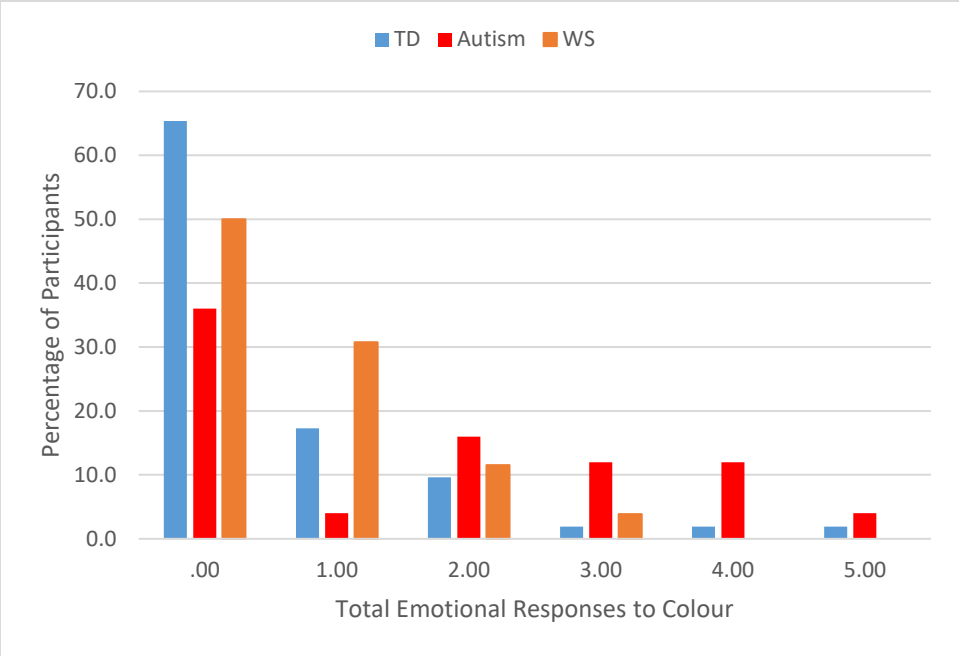



*Figure 6-3 – Average frequency of colour affected behaviours.. There was significantly more colour affected behaviours in the autism group compared to the WS and TD groups. This pattern was found for Total colour affected behaviours, and for emotion and action sub divisions. The Frequency of Actions is from a maximum of 16 as reported in the questionnaire.*

The frequency of colour affected behaviours can be seen in figure 6-4b. This shows several differences in the distribution of the number of colour affected behaviours. For both the TD and the WS groups there is a positively skewed data towards lower occurrences of colour affected behaviour. This was also the case for the emotional responses to colour and colour affected actions. Although the tail was longer for the TD group. A different pattern was seen in the autism group. For the total number of colour affected behaviours there is a clear bimodal distribution where there is a subgroup whose behaviour is severely affected by colour (24% of autism participants). There is also a relatively higher number of participants in the autism group who also displayed a moderate number of colour affected behaviours compared to both TD and WS groups, as indicated by a skewness towards lower values, whilst the autism curves are flatter and represent a more platykurtic distribution.

For emotional responses to colour (figure 6-4c) there is a similar distribution but the tail is much shorter. Colour affected actions were also found to have a higher and longer distribution tail, suggesting greater variability in colour affected behaviours compared to emotional responses to colour.



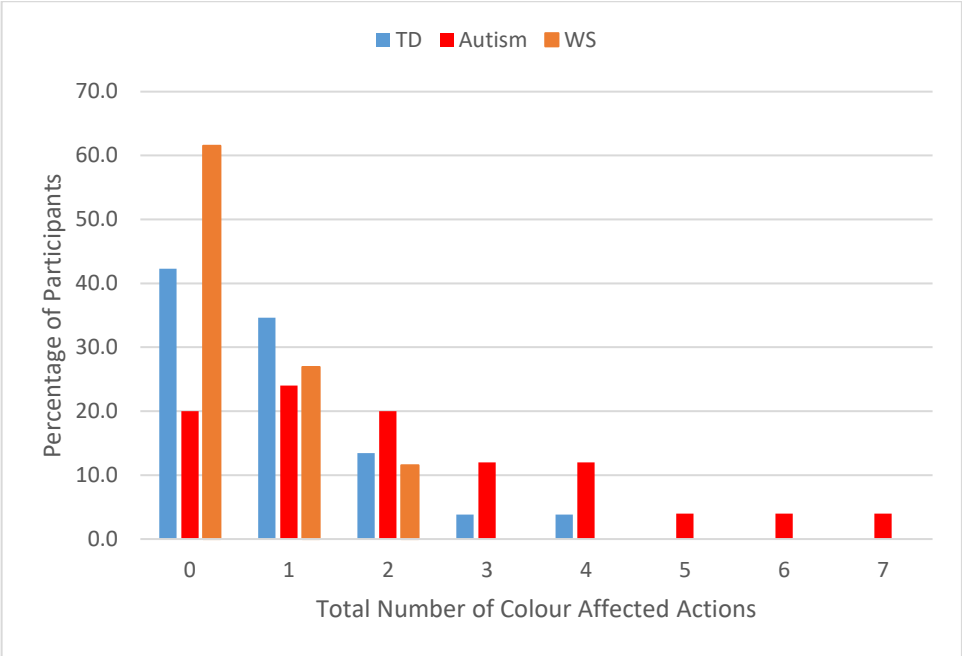



*Figure 6-4 – The distributions of the TD, Autism and WS groups.* Distributions are shown for: a) Total number of Colour Affected Behaviours, b) Total emotional responses to colour and c) Total number of colour affected actions.

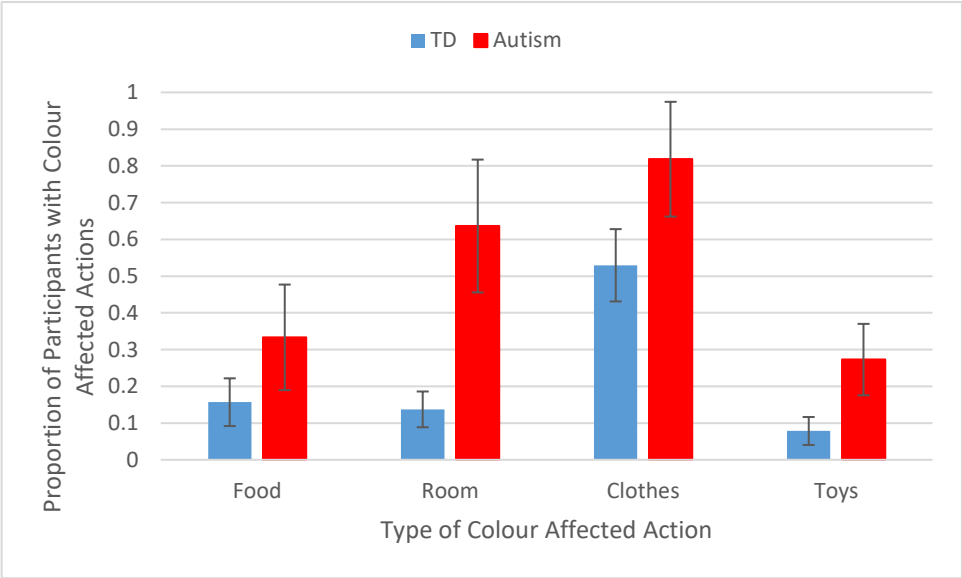



*Figure 6-5 - Proportion of colour affected actions for the autism and TD groups. There is significantly more colour affected actions relating to room colour in the autism group compared to the TD group. Whilst the most commonly affected action in both groups is the colour of clothes.*

For emotional responses to colour the autism group were reported to have significantly more negative emotional response to colour than the TD group, $U = 275$, $p < 0.001$ (see also Figure 6-6) Further analysis was conducted to see whether there were differences in the groups for emotional responses to colour. After using Bonferroni corrections for multiple comparisons, the autism group found colours more relaxing, $U = 399$, $p < 0.05$, and sad, $U = 275$, $p < 0.001$. There were no significant

differences for excited, $p = 0.84$, happy, $p = 0.18$, and avoiding, $p = 0.8$ emotional responses to colour.

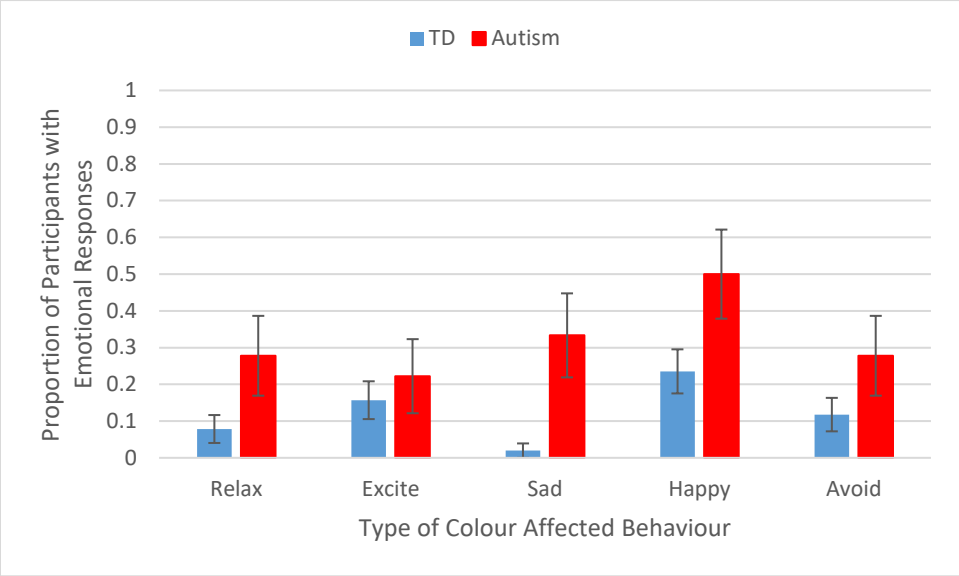



*Figure 6-6 – Proportion of participants with emotional responses to colour.* There are sad and relaxing emotional responses to colour in the autism group compared to the TD group.

The comparison between the autism and WS groups revealed syndrome specific responses. There were significantly more colour affected behaviours in the autism group compared to the WS group, $U = 131.5$, $p < 0.05$ (see Figure 6-3). Significant increases in colour affected actions in the autism group were also found, $U = 128.5$, $p < 0.001$, but not emotional responses to colour, $U = 165$, $p = 0.114$. To further explore the difference in the colour affected actions, additional analyses were conducted for the type of colour affected actions (food, room, toys and clothes). There were significantly more incidences of behaviour being affected by the colour in the autism group for room colour, $U = 169$, $p < 0.001$, clothes, $U = 149$, $p < 0.005$. There was no difference between groups for being affected by either the colour of toys ($p = 0.292$) or food ($p = 0.816$).

#### *6.4.2.3 Relationship between Basic Visual Function Score and Colour Affected Behaviours*

To investigate whether there was a relationship between scores on the quantitative and qualitative sections of the questionnaire. Participants were split by diagnostic group. There was no correlation between scores on the visual Sensory Profile questions and the total number of colour affected behaviours (lowest $p = 0.071$). There was also no significant correlation in any group for either emotional responses to colour (lowest $p = 0.428$), or colour affected actions (lowest $p = 0.096$). When collapsed across all participants, there was a significant positive correlation between Sensory Profile scores and the total number of colour affected behaviours, $r = 0.279$, $p < 0.01$. There was also a

significant positive correlation between visual Sensory Profile scores and emotional responses to colour, $r = 0.233$, $p < 0.05$, and colour affected actions, $r = 0.229$, $p < 0.05$.

In the TD group, there were significant positive correlations between Extended Visual Questions scores and the total number of colour affected behaviours, $r = 0.458$, $p < 0.001$, emotional responses to colour, $r = 0.3$, $p < 0.036$, and colour affected actions, $r = 0.431$, $p < 0.002$. There were no significant correlations for both the autism (lowest $p = 0.411$) and WS groups (lowest $p = 0.532$). When collapsed across all participants, there were significant correlations between Extended Visual Questions scores and the total number of colour affected behaviours, $r = 0.305$, $p < 0.003$, emotional responses to colour, $r = 0.265$, $p < 0.011$, and colour affected actions, $r = 0.229$, $p < 0.03$.

#### *6.4.2.4 Relationship between Questionnaire scores and Experimental Tasks*

##### 6.4.2.4.1 Basic Visual Function

The three primary experimental tasks from Chapters 3, 4 and 5 were correlated with scores on the visual Sensory Profile questions. The Chromatic Contrast Discrimination Test thresholds (Chapter 3 Experiment 2) were correlated with scores on the visual questions of the Sensory Profile. For all groups, there were no significant correlations between either chromatic or luminance discrimination thresholds with scores on the visual Sensory Profile questions (lowest $p = 0.298$). The standard deviation of colours of mid lightness and mid saturation level were also correlated with scores on visual Sensory Profile questions. There was a trend towards a significant negative correlation for the autism group, $r = -0.542$, $p = 0.056$. There was no significant correlation for either the TD, $r = -0.06$, $p = 0.691$, or WS groups, $r = -0.021$, $p = 0.921$. For the naming correlations, the overall, chromatic and grey scale concordance values were correlated with Sensory Profile scores. There were no significant correlations in either the TD or WS groups (lowest $p = 0.307$). No correlation was conducted for the autism group due to the small number (n=7) numbers of participants who completing the naming test (see Chapter 5 Section 5.3.1 for more participant details). There was no significant correlation of Sensory Profile scores with chronological age in any group (lowest $p = 0.118$).

Like previous studies, Additional correlations were conducted on all participants rather than split by group. For the Chromatic Contrast Discrimination Test (CCDT) there was no significant correlation of Sensory Profile scores with either luminance threshold, $r = 0.141$, $p = 0.416$, or chromatic threshold, $r = 0.192$, $p = 0.17$. There was a significant negative correlation between Sensory Profile scores and standard deviation of hue preference variants, $r = -0.241$, $p < 0.05$. There was no significant correlation between Sensory Profile scores and naming concordance ratings (lowest $p = 0.375$). There was a significant correlation between Sensory Profile scores and chronological age, $r = 0.616$, $p < 0.001$. However, this result is due to the increase in chronological age of participants in the autism

and WS groups who also have higher scores on the Sensory Profile and therefore not likely to be a true effect.

A similar set of analysis was undertaken for the basic visual function questions and performance on experimental tasks in the previous chapter. This separate analysis was done because the basic visual function questions were found to have better internal reliability than the Sensory Profile questions. For the CCDT there were no significant correlations in any group between basic visual function questions score and either luminance thresholds (lowest $p = 0.079$) or Chromatic Thresholds (lowest $p = 0.244$). There was a significant negative correlation in the autism group between variation in hue preference basic visual function question scores, $r = -0.774$, $p < 0.005$. There were no significant correlations for the TD ($p = 0.694$) or WS groups ($p = 0.459$). There were also no significant correlations between Extended Visual Questions scores and naming concordance in either the TD (lowest $p = 0.235$) or WS groups ($p = 0.525$). When collapsed across all participants there was a significant positive correlation between the basic visual function question score and luminance thresholds, $r = 0.24$, $p < 0.032$. There was also a trend toward significance between chromatic discrimination thresholds and basic visual function question, $r = 0.211$, $p = 0.059$. There was also a significant negative correlation for variation in hue preference and basic visual function question, $r = -0.315$, $p < 0.003$. There was no significant correlation between naming concordances basic visual function question scores (lowest $p = 0.189$).

#### 6.4.2.4.2 Colour Affected Behaviours

Colour affected behaviours were also correlated with performance on the experimental task from Chapters 3, 4 and 5 (Chromatic Discrimination, Colour Preference and Colour Naming). In the TD group, there were no significant correlations between experimental performance for any of three experimental tasks and total colour affected behaviours (lowest $p = 0.218$), emotional responses to colour (lowest $p = 0.339$), and colour affected actions (lowest $p = 0.431$). For the WS group there were also no significant correlations between experimental performance and total colour affected behaviours (lowest $p = 0.435$), emotional responses to colour (lowest $p = 0.269$), colour affected actions (lowest $p = 0.205$). In the autism group, there was a significant negative correlation between luminance discrimination threshold and emotional responses to colour, $r = -0.657$, $p <0.011$. No other correlations were significant for total number of colour affected behaviours (lowest $p = 0.103$) and colour affected actions (lowest $p = 0.199$). When data from all participants were pooled, there were no significant correlations between experimental performance and total colour affected behaviours (lowest $p = 0.552$), emotional responses to colour (lowest $p = 0.415$), colour affected actions (lowest $p = 0.38$).

## 6.5 Discussion

The new bespoke questionnaire developed in this chapter was found to found to have good validity and reliability as a measure of basic visual function and of colour affected behaviours. Moreover, the basic visual function questions had better internal consistency than the visual questions on the Sensory Profile. The results from the visual function questions revealed significantly higher scores for both Sensory profile questions and Expanded Questions for autism and WS groups compared to TD groups. However, scores on these questions did not correlate with performance on experimental tasks, with the exception in the autism group where variation in hue preference was negatively correlated with visual function scores. For colour affected behaviours there were significantly more affected behaviours in autism, including both emotional responses to colour and actions, than reported in the TD and WS groups. This was most pronounced for room colours. There were no significant differences between the WS and TD groups. Overall, patterns of colour affected behaviours were not associated with experimental tasks (except autism and variation of preference). Significant correlations were also observed between the total numbers of colour affected behaviours and basic visual function questions.

The questionnaire developed for this study was found to have a satisfactory level of Cronbach's Alpha. Further iterations of the questionnaire are needed. When compared to other sensory questionnaires, the one developed in this study underwent less revisions. Further development of the questionnaire is needed to include questions based colour affected behaviours and additional consultation with parents and clinical professionals to include possible missing item and check the wording of individual questions further. Additional validation needs to also be carried out in other samples, for example typically and atypically developing adults. The Cronbach's Alpha for this questionnaire was lower when compared to the Cronbach's Alpha overall for the Sensory Profile, Glasgow Sensory Questionnaire and Sensory Perception Quotient. However, when compared for visual items only it out performed the Sensory Profile (individual modalities were not reported for the Glasgow Sensory Questionnaire or Sensory Perception Quotient). Indeed, many of the questions that were removed during the factor analysis were those adapted from the Sensory Profile. Further research is needed to assess whether the visual questions in the Sensory Profile are adequately assessing visual function or are measuring behaviour that may be related to vision.

The finding of increased scores on visual functions questions (using both Sensory Profile and expanded questionnaires) in the autism group are in line with other studies that have used either the Sensory Profile, Glasgow Sensory Questionnaire or Sensory Processing Quotient (Freyberg et al., 2015; Green et al., 2015; Green et al., 2013; Kern et al., 2006; Lidstone et al., 2014; A. K. Ludlow et al., 2014; Rogers et al., 2003; Tavassoli et al., 2014; Tomchek & Dunn, 2007; Wigham et al., 2015). The higher scores amongst the WS group for visual scores is slightly different profile compared with

previous findings of WS individuals of a similar age where gustatory, auditory and proprioceptive senses had atypical sensitivities but visual functioning did not have atypical sensitivity (Janes et al., 2014; Riby et al., 2013). However, because only the visual functioning was assessed in the questionnaire study it is not possible to say whether the overall sensory profile is similar or different to the finding by Janes and colleagues (2014).

The dissociation between significant correlations for colour affected behaviours and experimental performance and the basic visual function questions suggests that the visual function questions do not correspond well to performance on sensory tasks. The only exception in this study, was a significant negative correlation between hue preference variation and scores on the visual function questions in the individuals with autism. It should be noted however that this means that reduced hue preference variation is associated with higher scores on the visual function scores. This reduced hue preference is still an atypical response to colour preference as it indicates a colour preference that is not dependent on hue (see Chapter 4 introduction and discussion for more details on colour preferences). In the autism group, only there was a significant negative correlation between luminance thresholds and emotional responses to colour. This could either reflect a "hyper-sensitive" response where greater discrimination is associated with increased likelihood of behavioural problems. Nonetheless there is no association with CCDT on chromatic axes performance, given that this is the most direct measure of sensory processing (chromatic discrimination) used in this thesis the lack of significant correlations of with performance on chromatic axes may suggest that these questions may not be a good measure of actual underlying chromatic sensory sensitivities as measured by psychophysical tasks. Whilst there is a relatively small sample size in this study, there is growing evidence from other researchers who have failed to find a relationship between the Sensory Profile and sensory processing or underlying neurological behaviour. There has been a failure to find a relationship between scores on the Sensory Profile and a measure of physiological reactivity, in this case electrodermal activity in adults with autism (McCormick et al., 2014; Schoen et al., 2009). For this study the lack of an association with performance on the colour experimental tasks may be due to the lack of appropriate questions of visual function, i.e. no items about colour in the Sensory Profile. The only previous study that has attempted to link visual function to performance on psychophysical tasks was the study of binocular rivalry in adults reported above. In this study the researchers found a significant correlation between number of switches between images on a binocular rivalry task and visual items on the Glasgow Sensory Questionnaire (Freyberg et al., 2015). Like this study there was a significant relationship when participant groups were collapsed together, whilst no group individually was shown to have this correlation. It is unclear whether this is because of a lack of range of sensory profile scores within each group to give a significant correlation. However, in both this study of young children and the Freyberg et al (2015) adult study, the clinical

groups were found to have significantly higher sensory sensitivities. Unfortunately, it is difficult to identify whether this correlation is due to the differences in sensory sensitivities or due to impaired performance on binocular rivalry by the autism group. Alternatively, this finding may simply indicate that the relationship between scores on questionnaires and results from psychophysical tasks is weak. This weak relationship could be due to the lack of appropriate questions or that a more comprehensive assessment of visual performance through multiple psychophysical tasks is needed. Given that this study and the one by Freyberg and colleagues (2015) are the only studies that this researcher has identified that have investigated this relationship between scores on an SP questionnaires and psychophysical performance, further research is needed to progress our understanding of these potential inter-relationships and differences between different sensory processing assessments. One solution to this could be to adopt a mixed methods approach to the study of sensory processing. The merits of this approach are considered in Chapter 7.

Overall the number of colour affected behaviours reported by parents across all groups, suggests that the inclusion of questions relating to colour would be a useful and meaningful addition to both the Sensory Profile and Glasgow Sensory Questionnaire. At present, no specific questions about colour are included in either measure. The Sensory Perception Quotient does have specific questions that relate to colour. However, these questions focus on matching similar colours (3/5 questions on the Sensory Perception Quotient), response to bright colours (1/5 questions on Sensory Perception Quotient) and wearing muted colours (1/5 questions on the Sensory Perception Quotient). The findings from on the Sensory Perception Quotient do not fully account for the behaviours reported in this study. The findings from this study indicate that choice of clothing (and not necessarily muted colours) is a common behaviour affected by colour; that colour affected behaviour was most commonly reported in the autism group; and that emotional responses to colour were also more common in the autism group compared to TD. Unfortunately, the Sensory Perception Quotient does not include any questions about emotional responses to colour. However, the results from this study suggest that the colour items in the Sensory Perception Quotient may not capture some of the colour affected behaviours most frequently reported by parents in this study. Given that this measure uses closed questions it possible that these behaviours are not adequately captured by the Sensory Perception Quotient.

The results from this part of the questionnaire have shed light on the nature of colour affected behaviours and the use of quantitative questions for the study of sensory behaviour in individuals with autism or WS. However, these results simply report the extent to which colour affected behaviours are present in each group, this may not be the best way to study some of the underpinning processes (such as the different biological, developmental and environmental factors that may be relevant) that might contribute to the behaviours observed by parents at a time in the

child's development arise nor the "cause" of the behaviours. Further, simply studying the frequencies for the groups may not be the most appropriate way to identify relationships between experimental results and behaviour noticeable to parents. Especially for the diagnostic groups, given the high heterogeneity in autism and WS, it may be more appropriate in the first instance to study sub-groups or indeed individual cases to explore potential relationships at the individual level. This might then lead to a more refined approach to the design of specific hypothesis testing in future research studies.

## 6.6 Conclusion

This chapter used a new questionnaire to assess parent reported basic visual function and the presence of colour affected behaviours. This new questionnaire was found to have good psychometric properties. On the basic visual function section, there was greater atypicality in both the autism and WS groups relative to a younger TD sample. There was also more reported colour affected behaviours in the autism group compared to both the WS and TD groups. Yet it is not clear about the nature of these colour affected behaviours and to what extent they fit into the diagnostic criteria in DSM-5 for sensory reactivity. The next chapter will report a selected number of case studies of individual participants from both the autism and WS groups will be described in more detail to explore the extent to which colour affected behaviours can be described under the restricted and repetitive behaviours domain.

# Chapter 7 - Case Studies

## 7.1 Overview

Chapter 6 reported the results of a new parental questionnaire to assess basic visual function and the presence of colour affected behaviours, and parent-reported *how* many participants had displayed colour affected behaviours. Yet little is known about the behavioural manifestation of these colour affected behaviours, the extent to which they affect the individual or *why* they occur. This chapter will extend the results of the previous chapter through a series of case studies of individuals whose parents reported either a pronounced behavioural and/or emotional responses to colours, or no strong colour affected behaviours. The cases can be classified in different ways depending on the child's experimental and performance related to and the parent (informant) questionnaire results.

## 7.2 Introduction

### 7.2.1 The Mixed Methods Method

This chapter will adopt a mixed methods approach for each individual case study. In recent years, there has been the rise in mixed methods methodology. This methodology incorporates the use of multiple methods (usually both quantitative and qualitative) in the study of the same phenomena. The methodology was born out of the need to reconcile the view that there is incompatibility between extreme positions of qualitative and quantitative research, and a desire to adopt a more pragmatic approach to research (Johnson & Onwuegbuzie, 2004). There is currently no established methodology or data analysis techniques for mixed methods methodology. For example, the different individual methodologies can be deployed either simultaneously or sequentially (Johnson & Onwuegbuzie, 2004; Palinkas, Aarons, et al., 2011; Palinkas, Horwitz, Chamberlain, Hurlburt, & Landsverk, 2011; Schifferdecker & Reed, 2009). Sequential mixed methods are where the results of one methodology inform the design of the second study. For example, using the results of an interview to inform experimental design or for validation of quantitative results with qualitative methods. Typically, this involves a transformation of a concept between the two different methodologies. For example Luckstead and colleagues (2009) used a questionnaire with open ended questions to support the quantitative findings of a peer-to-peer mental illness intervention (Lucksted, McNulty, Brayboy, & Forbes, 2015). Simultaneous mixed methods are when all methods are collected at the same time. The concurrent collection of different methodologies allows for a "*triangulation*" between the data from different methods. This allows for the direct comparisons between methods of different epistemological standpoints and allows for validation of one methods results with those from another. For example, Swain and colleagues (2010) used both quantitative and qualitative closed and open questions during a phone interview investigating the standard procedures of mental health agencies to establish matches and mismatches between the

quantitative and qualitative questions. Whether a simultaneous or sequential mixed methods design is used depends on the research question. In this study the aim is to assess to establish whether there is convergence between the different experimental tasks and the open/closed questions on the questionnaire (see aims above), as such the simultaneous mixed method design will be most appropriate to test this aim.

A mixed methods approach can also be applied to case study research (Yin, 2013), where the range of methods used can give greater information of the wider profile of the subject. Yet whilst the mixed methods approach has been used widely in psychiatric and educational research but it has seldom been applied to the study of sensory processing in autism or WS. One study has used a simultaneous mixed methods design to assess sensory processing in adult with autism (Robertson, 2012; Robertson & Simmons, 2013). This was during the development of the Glasgow Sensory Questionnaire (see Chapter 6 section 6.2.1.3) where the quantitative questions were supplemented with qualitative questions to further validate the Glasgow Sensory Questionnaire. The case study outlined earlier by Ludlow and colleagues (2014) is also a sequential mixed methods study. In this study the case study was identified and then from this information, the subject completed three different colour based tasks and was compared relative a similar mental ability autism and TD groups. However, it is unclear the extent to which there is variability in the relationship performance between the tasks in general. To circumvent this issues, this chapter will use multiple cases to further understand the relationship between the measures used in thesis.

### 7.2.2 Colour affected behaviours as Restricted and Repetitive Behaviours

Hyper- or hypo-reactivity to sensory input have been classified under the Restricted and Repetitive Behaviours domain in DSM-5 (American Psychiatric Association, 2013). These atypical responses to sensory input can manifest as an extreme response to a specific sensory stimulus (e.g. fascination with lights/spinning objects or excessive touching or smelling). Restricted and repetitive behaviours can be divided into two subcategories: Repetitive sensory motor and Insistence on sameness. Repetitive sensory motor actions, which include basic motor actions (e.g. hand flapping), sensory seeking behaviours and repetitive use of objects. Insistence on sameness refers to behaviours which are characterised by excessive following of a routine or rituals and difficulties with changes to those routines (Cuccaro et al., 2003; S. Leekam et al., 2007). Different possible manifestations of colour affected behaviours (emotional and actions influenced by colour) were identified in the Chapter 6. Yet it is unclear the extent to which these can be categorised within Restricted and Repetitive Behaviours domain. Colour affective behaviour could feasibly manifest as an example of either insistence on sameness (e.g. wearing clothing of a colour) or repetitive sensory motor actions (e.g. an unusual sensory dis/interest of a colour). Although atypical reactivity to sensory input is included in DSM-5, little is known about what the phenotype is for these behaviours.

### 7.2.3 Rationale for Chapter:

1) To use a simultaneous mixed methods design to investigate performance across the experimental tasks and questionnaire used in this thesis.
2) Evaluate a series of case studies on the nature of colour affected behaviours and whether these are related to experimental performance.
3) To investigate whether colour affected behaviours can be categorised as restricted and repetitive behaviours.

## 7.3 Method

### 7.3.1 Selection of Cases

The in-depth cases were selected based on those individuals who were reported on the parental questionnaire to have a colour affected behaviour. Individuals who had the most available qualitative data were more likely to be selected. The best exemplars of each case study classification were chosen (see section below). Case studies were selected from the same participant sample as outlined in Chapter 6.

### 7.3.2 Data Analysis

The data for the case studies were compiled from the individual performance across all experimental results, as well as parental and participant qualitative results. Details around colour affected behaviour were compared to performance on the chromatic discrimination, colour preference and colour naming (see Chapters 3, 4 and 5 for more details on each experimental task). This also included semi-structured interviews with parents in the autism and WS groups where they were tested at home or at Newcastle University (see Chapter 2 Section 2.3 for details on where participants were tested). The semi-structured interview used the similar prompts based around the colour affected behaviours in the questionnaire. There were also semi-structured interviews following the same protocol done with each of the participants. The case studies were classified into different types from category and relationships based upon the data based on qualitative inspection of the data and the identification of different themes describing the relationship between the qualitative and quantitative data. The case studies will be split between the autism and WS groups due to the differences on experimental task performance (see Chapters 3, 4 and 5) between the autism and WS groups relative to their respective TD control groups.

### 7.3.3 Classification of Cases

The most common identified classification of the case studies is listed below:

a) Colour Object/Action Associations:

This classification denotes where there is reported to be an association between a colour and an object or action. This association can be either positive or negative. It should be noted that this is separate from colour preferences that are identified in the colour preference task.

b) Emotional Responses to Colour

This is where there are reported emotional responses to colour from the parental questionnaire...

c) Other incidences of Colour Affected Behaviours

Incidents where the individual shows a colour affected behaviour but that are infrequent and can't be classified as either Colour Object/Action Associations or Emotional Responses to Colour.

d) No colour affected behaviours

Participants who were reported to show no colour affected behaviour.

## 7.4 Results

### 7.4.1 TD group

As expected from the results earlier in Chapter Six there were fewer potential case studies to select from as there were significantly fewer incidences of colour affected behaviours in the TD group. Despite this there were 33.3% and 56.9% participants who reported as either having either an emotional response or action affected by colour.

#### *7.4.1.1 Colour Object Associations: Case Study TD1*

Case Study TD1 was a TD participant aged 4 years and 10 months. He was reported to have colour affected behaviours around the colours red and pink. He did not like wearing pink coloured clothes, as this was a "*girl's colour*". There were several colours affected behaviours for the colour red. He was reported to "*love"* red in a room. Furthermore, he liked having a red coloured bike. His previous bike was not coloured red and he was not confident in riding it. Yet of his new bike, "*as soon as he saw it was red he was confident of riding it*". There were historical reports of his behaviour being affected by red when he was three years old. When offered a blanket he initially refused it, however when he was told that the blanket was red he would accept the blanket. He now has a white blanket and is less concerned by the colour of the blanket suggesting that colour affected behaviours may reduce with chronological age in typical development. In comparison to his experimental results, on the colour preference task he selected his favourite colour to be red. This colour was selected the highest number of times, though there was no dislike of colours that were close to pink (high lightness and high saturation variants for the reddish hue) His naming concordance was also good for chromatic colours (average concordance = 0.74) and for reddish hues (average concordance = 0.88).

This shows that he is more likely to name colours with of reddish hues in line with his peers, more so than other colours.

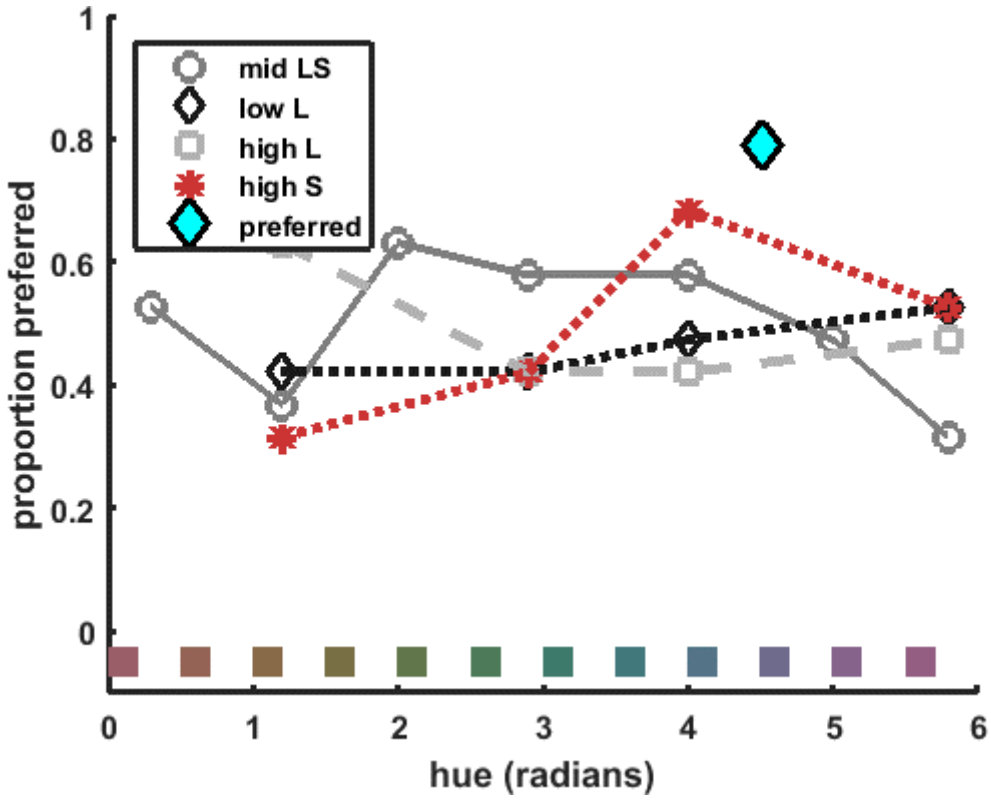



*Figure 7-1 - Individual colour preference curves for Case Study TD 1. The individual does not show a pronounced colour preference pattern based on hue, lightness or saturation. However, their favourite self-selected colour red was chosen the most times.*

### 7.4.2 Autism Case Studies

Cases were selected and included as a case study to illustrate different profiles of parent reported colour affected behaviours and the results from the experimental tests. There were 80% and 57.1% of participants who reported to have either an action or emotional response to colour respectively.

#### *7.4.2.1 Colour Object Associations: Case Study Autism 1*

This was a 16-year-old male who wears aqua tinted glasses. Both mother and step-father completed the questionnaire. They agreed that the participant's favourite colour was purple, and that there was a recent development in the last four months of a dislike of orange. The step father says this is because orange reminds him of drug dealers and that he doesn't like ginger haired people. His mother added that he also doesn't like to be in rooms with orange in them. However, when the participant was asked by the researchers, the participant wasn't sure why he didn't like orange. The participant also showed various colour affected behaviours. In the past he had a friend who had pink gloves and these pink gloves used to help the participant relax. He would prefer to be in purple coloured rooms and to wear clothes that were black, grey, brown or black/white. Whilst refusing to

wear clothes that were brightly coloured. The participant dislikes playing computer games where there is a high contrast of colours. Furthermore, his mother recognised that the participant does not like to use colour in their artwork. When asked why, the participant stated, “I can't use colour well, I don't like colour in my art”. However, was confident enough to say that purple and green do not go together. Both parents and the participant identified that the purple/lavender was relaxing, because this “*reminds me of incense*”. His mother also believed that pink could be relaxing for him as well as purple on some occasions. In addition, the participant stated that pink and purple make him happy and that he was excited by “bright white”. As alluded to earlier, orange would be avoided where possible. The participant says that they like the combination of purple with white and cream, however this was a mismatch between their parents who instead believes that he likes black with other colours (purple, pink or yellow). The preference curve of this participant is in keeping with the colour affected behaviours described by himself and his parents. There is a clear dependence on hue, where there is a relative peak for purple and pink hues for all variants except low lightness colours. There is also a clear dislike for brownish hues for all manipulations, including high lightness and saturation manipulations these colours that are closer categorically to orange than the brownish hue variant.

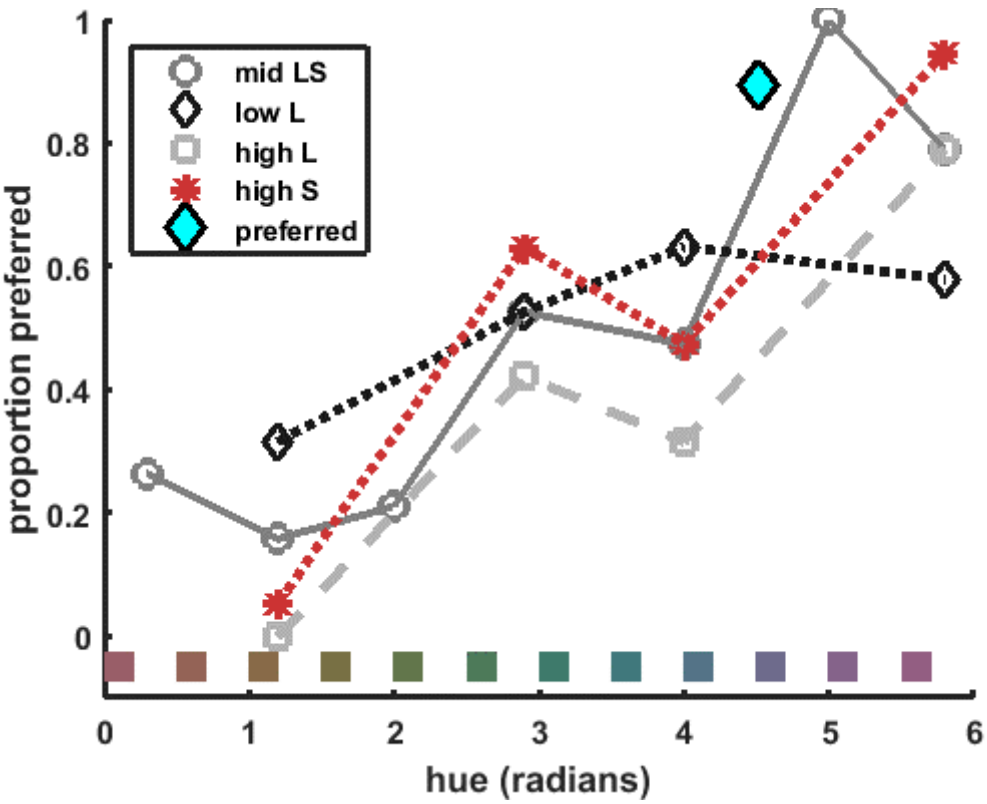



*Figure 7-2 - Preference curves for Autism Case Study 1.* There is a clear preference for purplish hue, as well as a dislike for brownish lightness and saturation variants which are close orange.

*7.4.2.2 The Colour-Taste Synesthete – Autism Case Study 2*

This participant is a 15-year-old male with autism. He was currently wearing purple tinted glasses, but was asked to remove them for the colour experiments. His parent recognised that there are colours that he likes and dislikes but was not able to specify them. He was reported to have emotional responses to different colours. There is a specific negative emotional response to different types of reds. Where red is associated with sadness and is also avoided, more generally "dull" colours are avoided. He was also reported to like blue and purple together, but dislike red and blue together. He has several behaviours that are affected by colour. There was a dislike of rooms that have red tones in them (but did not insist on a colour for a room) and a dislike of playing with pink toys. He likes to wear colours that are dark, and when drawing or painting would prefer to use colours such as purple, grey and black. During the test sessions, he would remark about how certain colours looked vibrant and bright because this was in line with his favourite art style, and would verbally comment on how he disliked dull colours (mimicking the parental questionnaire). He mentioned that he associated certain colours with positive and negative emotional states. Blue and sky blue were associated with calmness, green was peaceful, red is anger (which also "*hurt*" his brain) and brown/grey were depressing. Upon seeing colour of low lightness, he would call these colours "*dull*", which is somewhat at odds with his parent's comment that he likes to wear dark colours. He also commented on how yellow text or a combination of red/blue as either text/background combinations were also negative. Finally, he made comments about being synesthetic for the colour red which tasted sweet to him. No other colour gave a synesthetic response. His preference results (Figure 7-3) reflect both his own comments and those from the questionnaire. There is a clear dislike for low lightness variants and high lightness and highly saturated colours are generally more preferred over mid lightness and saturated hue variants. The variations per hue are peak for colours (green/blue) that he associated with positive emotions. This case shows that there is a synesthetic response to the colour red. It shows how his response to red had primarily negative associations. Previous research has suggested that "*sensory overload*" (see Chapter 6 introduction) may be a problem for individuals with autism. This case would qualitatively fit that criteria as there is dislike of red which is related to the presence both taste and visual sensory response, unlike the other case studies where whether the colour affected behaviour is caused by sensory overload is harder to determine.

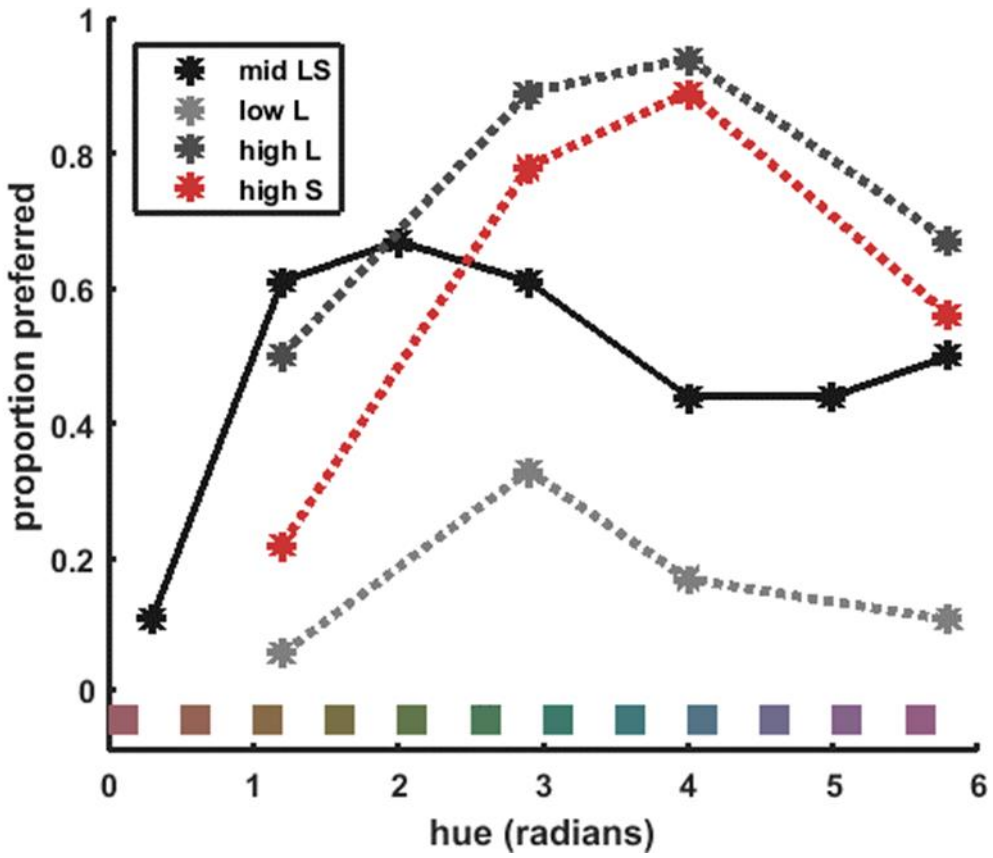



*Figure 7-3 - Preference curve for Autism Case Study 2. The x-axis is the hue radians, whilst the y-axis is the proportion that the colour was preferred.*

### *7.4.2.3 Emotional Responses to Colour: Autism Case Study 3*

This case study represents the best example of an emotional response to colour in the autism group. Autism Case study 3 was a 14-year-old male with autism. His parent reported his favourite colour to be blue, however the participant said that it's brown. Both parent and participant agreed that white was disliked because it is "*empty and feels plain*". There were several colour affected behaviours of the participant reported by the parent. His parent reported that despite disliking white, he would like to eat white food but instead disliked eating red and green coloured foods. The colour of the rooms would also affect behaviour. He would dislike being in a white room, but would instead prefer to be in a green and white room because the addition of green means that it is, "no longer [a] plain" colour. He would also sit in a pale yellow coloured room to relax after finishing school. Coloured clothing was also an issue, where he would prefer to wear either dark green or brown. His parent commented on how he likes to wear a brown coat and specifically chose this colour (whilst his brother chose a more colourful one). If he was asked to wear a brightly coloured or pink items of clothing, then he would refuse to wear this. He would also like toys because of their colours, for example his parent said that he liked a Spiderman toy when he was younger because of the bright colours (red and blue). The participant also commented how they liked bright colours in other toys,

e.g. Warhammer (blue and gold). His parent believed that this was because, “He likes other things to stand out, not himself”. When drawing/painting he would prefer to use darker colours and will only draw using a grey pencil. He would also display several emotion responses to colour. The colour gold would excite him and make him feel happy, especially when combined with blue, as these colours complemented each other. They also reflect the primary colours in his Warhammer interest. He would avoid pinks and purples as these are “feminine” colours. Orange and black would make him feel sad as these colours were difficult to mix with other colours. Yellows and browns would be relaxing because of the previously mentioned relaxation room. He had learnt colours by 5 years of age and per his parent can name colours reliably. The participant’s preference curve can be seen in figure 7-4. Unlike previous case studies in this chapter, this participant’s preference curve does not fit the responses from their questionnaire. For basic hue manipulations, there is a decreased preference for purple and pink hues which were considered to be feminine. Other responses from the questionnaire suggest that the preference curves may represent his preference for brown and darkish coloured clothes. This suggests that in the case where there are multiple behaviours affected by colours, that some colours may be more influential to the participant’s colour preference.

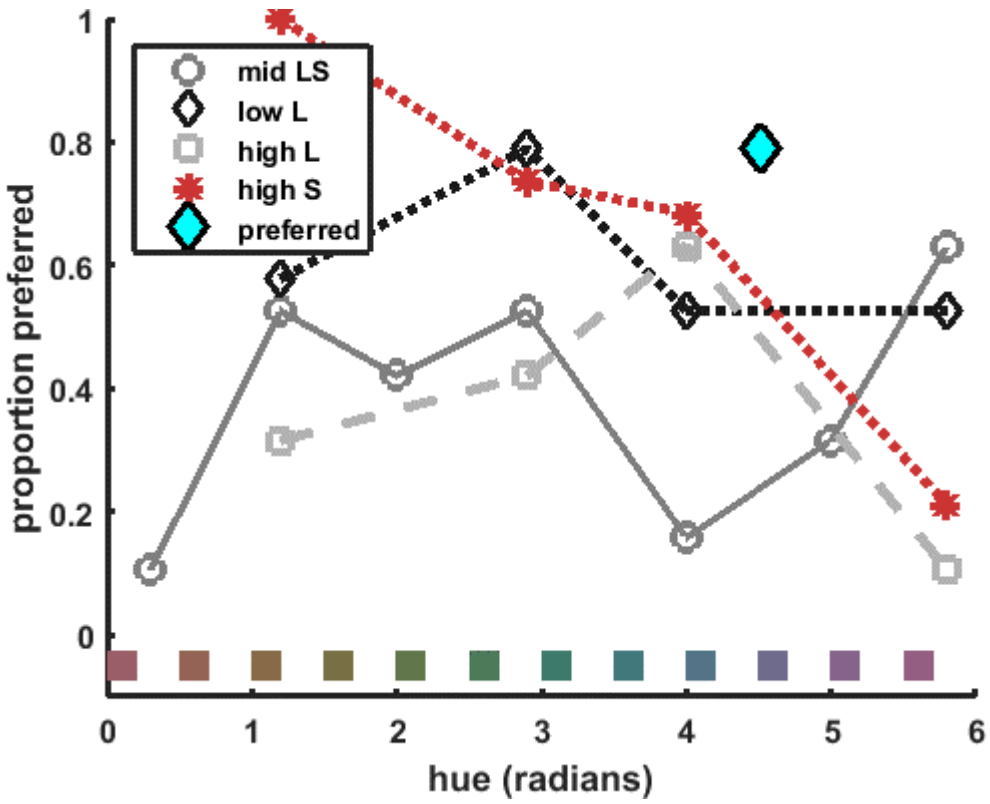



*Figure 7-4 - Preference curves for Autism Case Study 3.* There is a dislike of “feminine” colours and a relative preference for brown and low lightness variants

#### *7.4.2.4 Other Anecdotes from autism participants*

Unlike the WS there were many other participants in the autism group who displayed colour affected behaviours. For example, there was a female participant who refused to wear pink clothes, their preference curve shows a trough for pinkish hues. This participant would also take to coloured ink being used to mark their school work, instead preferring it to be marked it black. If red ink was used, then the participant would rewrite their whole notebook to "correct mistakes". There is another case of a male participant who is distrustful of red colours in their food, or the child who dislikes yellows, and will also not eat bananas. Another participant was reported to like green because it reminded them of nature. They also liked bright neon colours and "colourful computer games" which was at odds with their hypo-reactiveness to other senses. These participants show clear colour preferences that are dependent on their object associations with a colour. However as was the case in the WS group there are participants who showed either less pronounced colour preference patterns or showed a preference for some hues, colours preferences that were not associated with a colour affected behaviour, suggesting again that it is not only object associations are driving colour preference (see also chapter 4).

### 7.4.3 WS Case Studies

#### *7.4.3.1 Positive Colour Affected Behaviours – Williams Syndrome Case Study 1*

This section will describe the case report of one child with WS whose parent described some positive colour affected behaviours and consider these behaviours in relation to the pattern of results from the same child's colour preference experiment test results.

Williams Syndrome Case Study 1 was a 13-year-old male with WS. His favourite colour/and colour preference was reported by both the child himself and his parent, to be orange. He also had several colour affected behaviours. These included his willingness to eat green foods (specifically peas and broccoli). He will choose orange and green coloured clothing when given the option and chose a light green colour for their bedroom walls. His mother also reported several positive emotional and behavioural responses to orange: the colour excites him and makes him happy (as does yellow); His favourite drink cup was orange; he prefers to eat orange jelly babies. He was reported to be attracted to people with orange/ginger hair and his favourite dog was a Rhodesian Ridgeback which also has an orangey brown colour. When painting, or drawing he shows a preference to use oranges, greens and yellows. This child's preference curve can be seen in figure 7-5 below. The curve does not show a noticeable colour preference that is dependent on either lightness, saturation or hue, except for a colour of a mid-lightness and mid-saturation brownish hue. However, he selected orange as his favourite colour and during testing he was visibly happy to see the colour orange. Unfortunately, In the stimuli set for the colour preference task, there is not a good example of orange and yellow

(outside of those in the Macbeth Colour Checker Chart) a potential limitation when investigating a range of colour preferences.

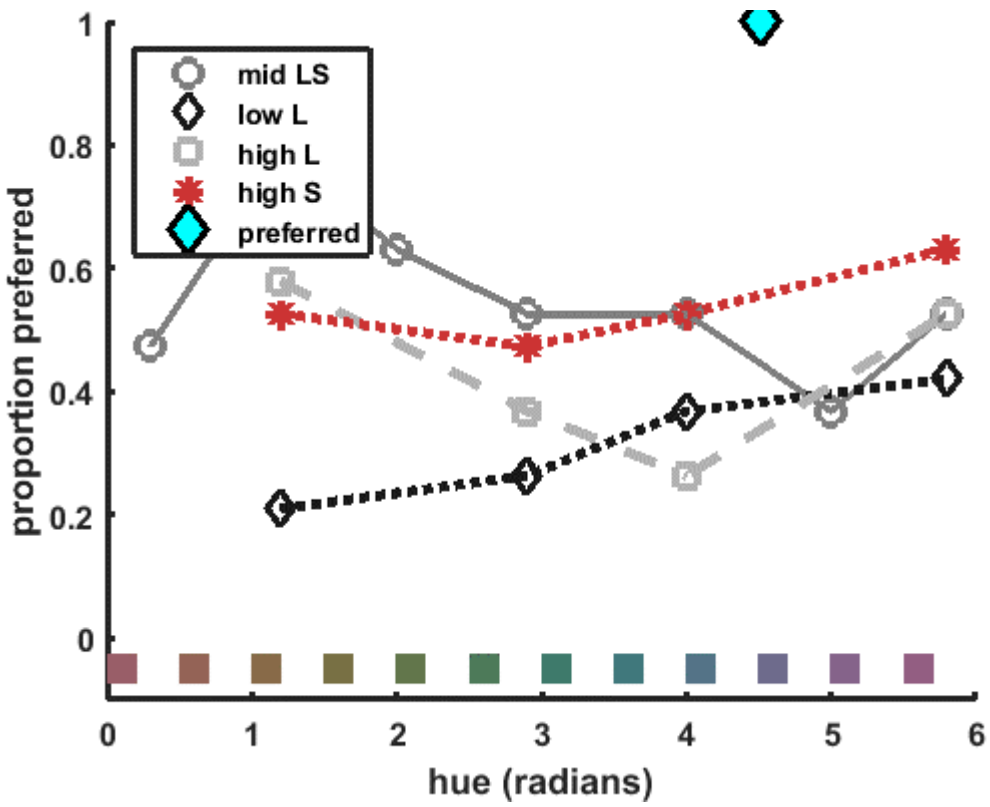



*Figure 7-5 - Preference curves for Williams Syndrome Case Study 1. Whilst there is no discernible pattern from hue, lightness or saturation variants, there is a clear preference for the self-selected favourite colour.*

The parent also reported that this child will identify things using colours rather than using the actual name of the object. For example, he will refer to crisp packets using the colour but not the flavour. Furthermore, he was reported to learn colour names at 7 years old, which is later than what would be expected of a typically developing child. His performance on the colour naming task in chapter 5 showed that he had relatively low concordance (low agreement) for overall colour naming (0.42), chromatic colours only (0.48) and achromatic colours only (0.23). The parent reported that he had difficulty naming blues and blacks. When looking at the corresponding individual colour patches this was not the case for the colours that had a high agreement that the colour was called blue. They had less agreement for the coloured patch at the blue colour boundary, but were still categorised as blue by most participants. For black, there was increased atypicality in naming darker colours, including use of the term mist. In fact, when naming the achromatic colours the term “grey” was not used, instead white, green or mist were used, indicating an lack of reliable colour naming for achromatic colours. Moreover “white” was used to name other chromatic colours were the individual lacked the colour term, for example calling browns white. These results from the naming of blue and black

colours confirm the parent's observation of difficulty naming for black and for non-prototypical blues. An additional example of a WS participant who showed peaks in colour preference curves that related to positive colour affected behaviours can be found in Appendix 5. The collection of WS participants who display a preference curve between a positive colour affected behaviour and colour preference. Another participant also shows another pattern related to their colour naming. Their reduced concordance rates for both chromatic and achromatic colours stemmed from a reduced colour vocabulary. This participant would inappropriately over generalise colour terms that they already know to colours that they are less certain about naming. This is a well-established pattern of vocabulary development in typical development. This suggests that the atypical colour naming seen in Chapter 5 may be represent a developmental delay for the development of basic colour terms.

*7.4.3.2 Negative Colour Affected– Williams Syndrome Case Study 2.*

Of the 26 children with WS in this study, 4 had parents who reported a negative colour affected behaviour. For example, Williams Syndrome Case Study 2 is a 10-year-old female with WS whose parent reported that her favourite colour is pink and that she dislikes green. The participant will choose pink things if given the option (e.g. clothes). She gets excited when being given something that is pink (clothes or toys). The parent also reports that she likes metallic colours, shiny silver marbles, but she loses interest in the marbles when their shininess goes. This suggests that it may not be metallic colours that the child likes but colours that appear to be shiny. Her behaviour also shows a dislike of greens. For example, she will give away colouring pencils that are green. She also dislikes wearing green coloured clothes and will ask to wear different clothing items. In the preference experiment, she showed a clear dislike of greenish hues. Conversely colours most analogous to pink (i.e. highly saturated and high lightness for reddish hues (see figure 7-6). There is a clear dip in preference for colours that are of greenish hues. Whilst colours closest to representing pink are preferred more, as well as her favourite colour was a pink which was selected most often. They were reported to have learnt colour names at 4 years old. Her performance on the colour naming task was in line with the average performance by the WS group for their overall, chromatic and achromatic concordance. This case highlights that not only positive colour-object associations' influence colour preference. In this case, there was a clear dislike of green that was also reported in colour affected behaviour.

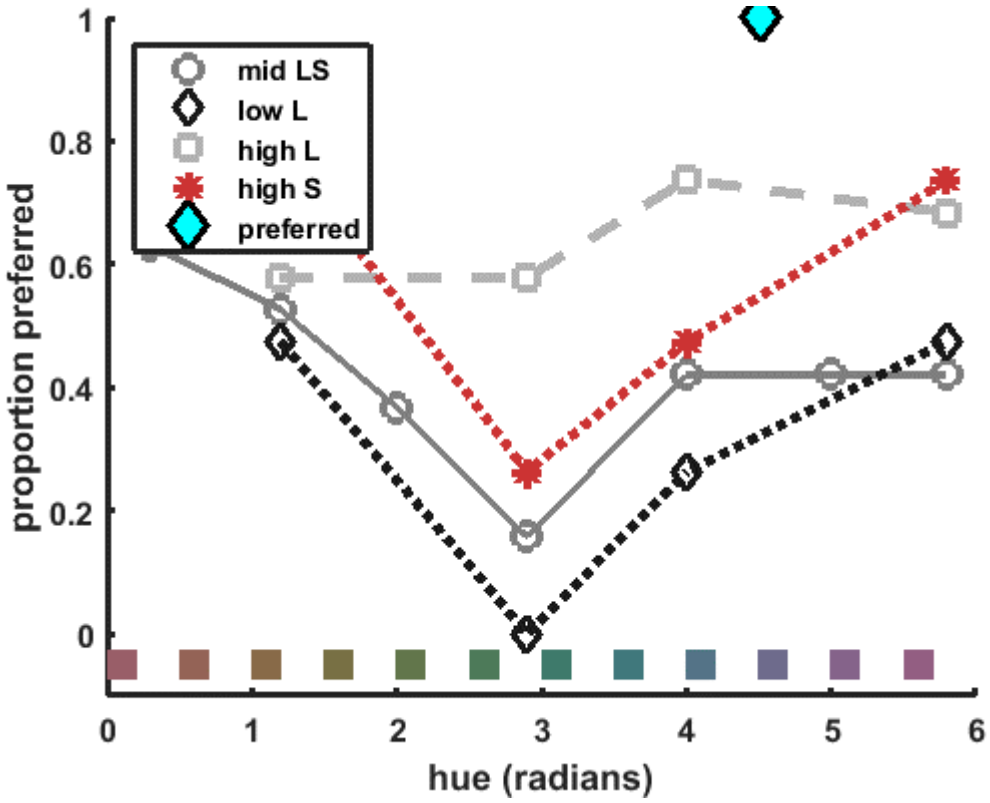



*Figure 7-6 - Preference curves for Williams syndrome Case Study 2.* There is a clear dislike of colours that have a greenish hue.

### *7.4.3.3 Other sensory associations: Williams Syndrome Case Study 3*

Williams Syndrome case study 3 is a nine-year-old male with WS. His parent reported that he is interested in emergency vehicles- many of which have blue flashing lights. This has extended beyond emergency vehicles to include other similar flashing lights (e.g. Christmas fayre, roadworks, car indicators). This fascination and excitement in relation to blue flashing lights will cause him to lose focus on whatever he is doing, but he will also become more anxious and tense because of the associated loud noise. His parent believes that, "*his reaction to colour is more driven by the other sensory experiences- in this case his reaction to loud noise, i.e. sirens tend to come with blue lights, but it is the noise of the sirens that drive his reactions*" not the visual sensory input of the colour. Similar associations between the presence and severity of repetitive behaviours (as measured by the RBQ) and sensory sensitivities, where sources of anxiety can become a repetitive interest that have been reported in WS using questionnaires (Janes et al., 2014; Riby et al., 2013). It is also an example of object associations driving colour preferences (Palmer & Schloss, 2010). This case study demonstrates additional qualitative evidence for a possible relationship between repetitive behaviours and sensory affected behaviours. Figure 7-7 shows his preference curve. There is hue dependent preference for colours of a medium level of lightness and saturation, where there is preference for clear for blue medium lightness and saturation and reddish hues. There is a hue-

dependent preference for lightness and saturation variants, where reddish hues of this variants are preferred more.

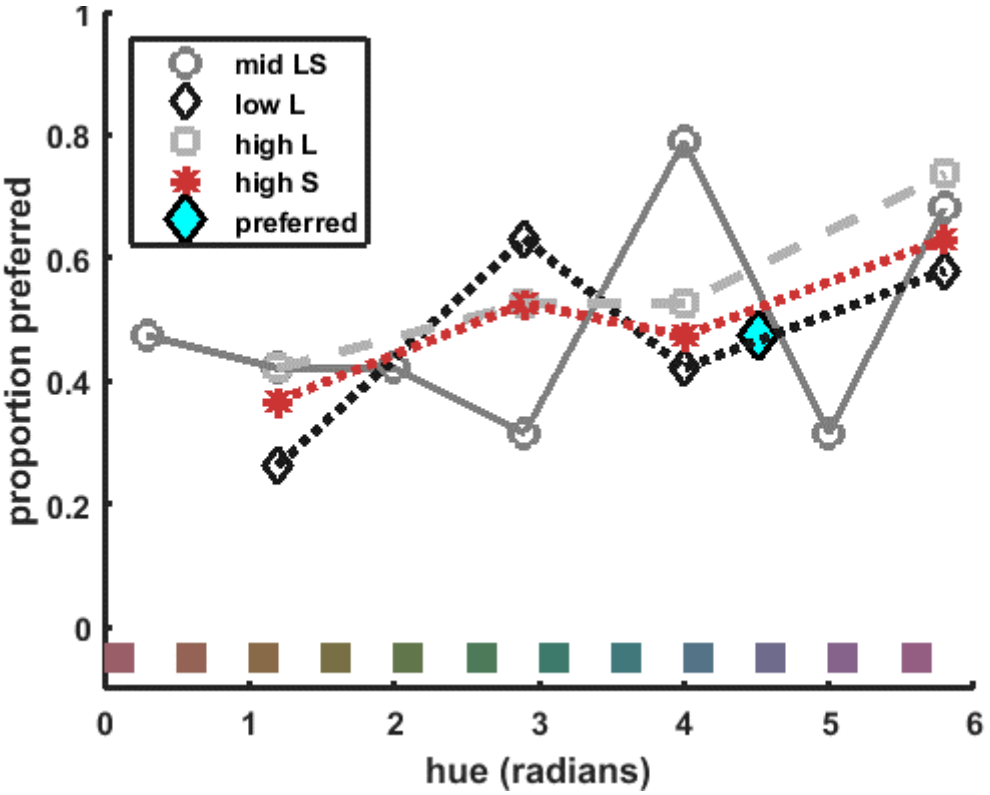



*Figure 7-7 - Preference curves for Williams Syndrome Case Study 3.*

His parents report him being able to use elaborate colour terms such as indigo or violet for blue and purple respectively. But there were no incidences of these words being used in the naming chapter, although this may be because there is not an appropriate exemplar of indigo/violet or that these names are not applied consistently. This case shows a different manifestation for the colour affected behaviours, namely through other sensory modalities or other sensory interests.

*7.4.3.4 Emotional Responses and Imaginative use of Colour – Williams Syndrome Case Study 4*

This was a 15-year-old male with WS, his parent reported that his favourite colour was blue and that reds would make him angry. For example, as a younger child (aged 7-9 years) he would get upset each morning as he dressed in his red school uniform, but this did not interfere with his functioning in school. He likes to draw and paint with blue colours and will avoid reds and browns if he can (such as colour of shoes). During the testing session, the participant made a comment about how he writing a story about a war between two factions. The “good” and the “bad” guys were separated by their colour, where the “good” guys dressed in blues and greens, but the “bad” guys were dressed in reds. The factions were fighting over worlds that all had different hues and saturations. On some worlds, the colours would be reversed. For example, grass would be blue or the sky would be green. When asked whether any worlds were red, the participant responded that this was where the bad

guys came from. His preference curve shows an increase either high lightness or highly saturated for blues, greens and reddish hues. By comparison, low lightness colours were least preferred.

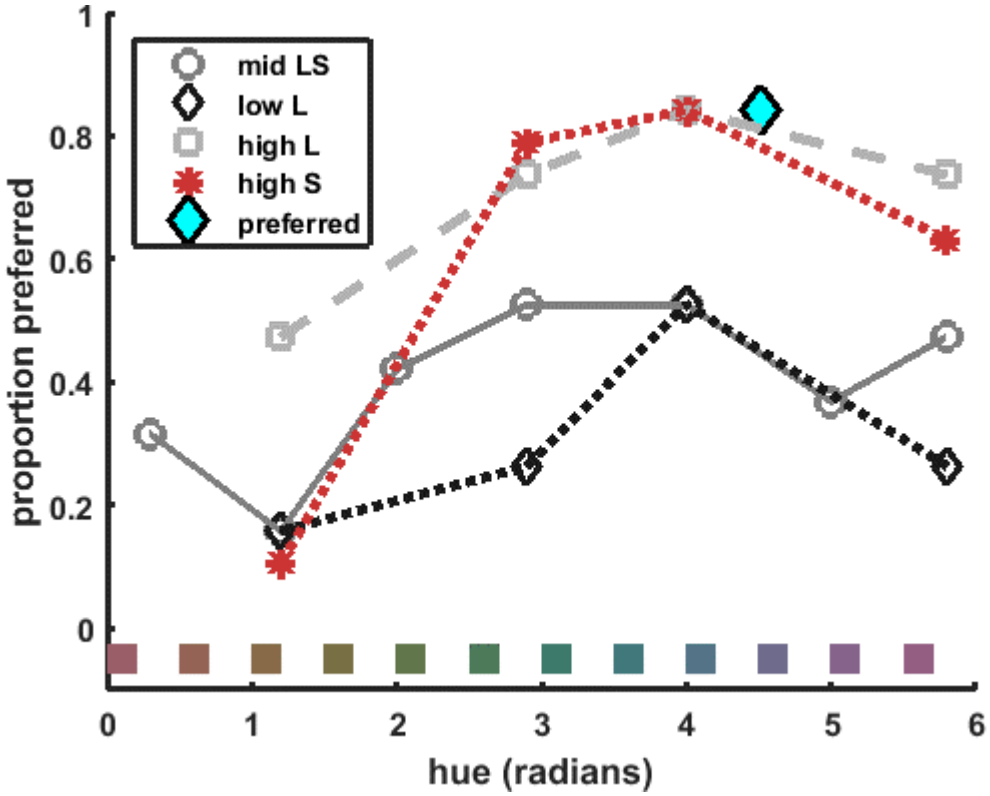



*Figure 7-8 - Preference curves for Williams syndrome Case Study 4. There is a preference for high lightness and high saturation variants for all hues, except for brownish hues. These relative peaks and troughs reflect the associations of "good" and "bad" in their story narrative.*

His parent also reported that he could name colours reliably at around 8 years old, and uses more elaborate terms for areas around favourite colours such as Teal, Turquoise, sea green. During the colour naming task, he described a close match as 'cyan'. However overall he showed similar concordance for naming of colour in line with the rest of the WS group for overall patches, chromatic and achromatic patches. This WS participant has shown that he can abstract the use of colour to categorise valence based on his own personal colour preferences (Figure 7-8).

### *7.4.3.5 Other Summaries from WS participants*

For the remaining eight WS participants, parents did not report any colour affected behaviours. Five out of eight participants showed clear colour preference patterns that were dependent on either hue, lightness or saturation variations. For three of these participants, the colour preference curves were in line with what the parent thought their child's favourite colour was. One child gave a reason for their favourite colour (i.e. it is Donny Osmond's favourite colour). For the other two; one WS participant's preference curve did not agree with their parent, whilst the final WS participant's

parent did not report a favourite colour. Only one of these WS participants who showed a colour preference but only one colour affected behaviour were reported to have a colour affected behaviour and their preference curve was in line with this. The remaining WS participants did not have a reported colour affected behaviour (see figure 7-9a). This suggests that object/action association is not the only association for colour preference. The WS participants who showed no clear colour preference pattern, their parent was also unlikely to report any colour affected behaviour. Some parents commented that their child was not affected by colour which was accompanied by flatter colour preference curves with no clear colour preference along hue, saturation or lightness variants (see figure 7-9b). This inter-individual variability in the preference curves and the presence/absence of colour affected behaviours may explain the relatively flatter preference curves seen at the group level for the WS group (see Chapter 4 results for more details).

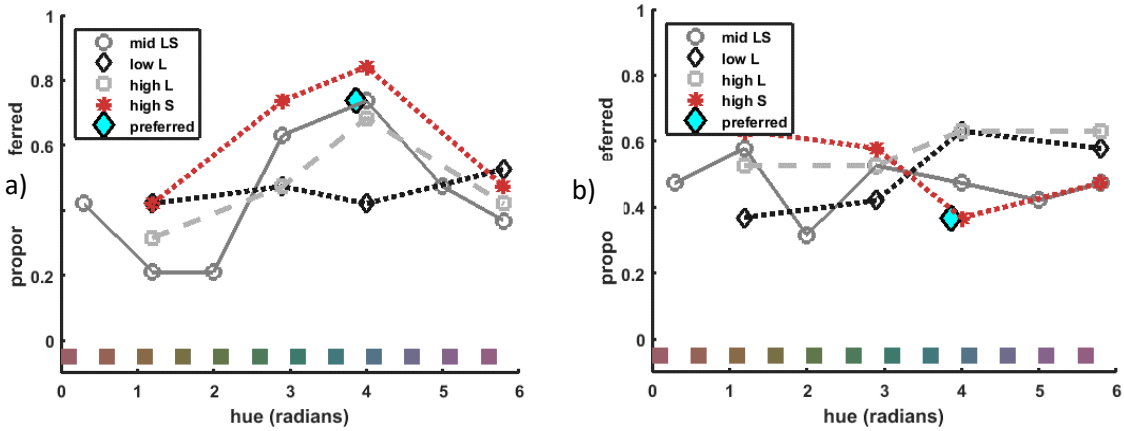



*Figure 7-9 – Example of colour preference curves for individuals who did not display a colour affected behaviour.* a) Preference curves for a participant with WS, who displays clear hue dependent colour preference in the absence of a colour affected behaviour and b) Another participant with WS, who shows no clear colour preference pattern whilst also displaying no colour affected behaviours.

### 7.4.4 Summary of Case Studies

In the WS group the lack of a colour affected behaviour was usually associated with colour preference curves with a less pronounced colour preference. It is important to note that even though the WS group showed no significant difference from the TD group for the frequency of colour affected behaviours, there were still individuals in the WS group who displayed several colour affected behaviours.

For the autism group, overall there were a larger number of reported colour affected behaviours. The individuals were also more likely to ascribe emotions onto colours and/or have emotional responses. One participant with autism reported to be synesthetic, where the colour they perceived multi-modally was also the colour that was most uncomfortable to them. These case studies highlight the wide inter-individual variability in responses to colour by participants from both groups.

### 7.4.5 Frequencies of Classification between groups

From the case studies, there is a consistent relationship between colour affected behaviours and relative colour preference patterns. To further investigate this possible relationship, the colour preference curves from all participants who had returned questionnaires (See Chapter 6 Methods section 6.2.1 for participant details) and completed the colour preference experiment (See Chapter 4) were compared to see the extent to this pattern continued across the whole sample (See table 1). Across all participants there were more matches than mismatches between hue colour preference curves and the reported colour affected behaviour. This was particularly pronounced for the TD and autism groups most of the actions and emotional responses to colour was also reflected in their hue preference curve. For the WS the proportion of matches and mismatches were more equal suggesting that there may be different association between colour preference and colour affected actions. Another possibility is that the colour preference colour set does not cover the colours that cause the colour affected behaviours (e.g. metallic or neon colours). A percentage of participants who were classified as mismatches were reported to have colours that were not in the colour preference stimuli set.

*Table 7-9 - Percentage of matches/mismatches between hue colour preference curves and colour affected behaviours and no colour affected behaviours for each group.* Raw numbers for each cell are shown in brackets.

| | Action | | | Emotion | | |
|---|---|---|---|---|---|---|
| Group | Match | Mismatch | No Action | Match | Mismatch | No Emotion |
| TD | 47.9 (23) | 12.5 (6) | 39.6 (19) | 16.7 (8) | 16.7 (8) | 66.7 (32) |
| Autism | 50 (10) | 30 (6) | 20 (4) | 32 (8) | 25 (5) | 35 (7) |
| WS | 20 (5) | 20 (5) | 60 (15) | 24 (6) | 32 (8) | 44 (11) |
| All Participants | 40.9 (38) | 18.3 (17) | 40.9 (38) | 23.7 (22) | 22.6 (21) | 53.8 (50) |

## 7.5 Discussion

Individuals from both the autism and WS groups were found to display colour affected behaviours. The manifestation of these behaviours and the colours that were involved varied widely between individuals in both groups. Despite this there was a common trend that the individual colour preference curves were largely congruent with the reported colour affected behaviour. The case studies also demonstrated that many of the colour affected behaviours took the form of the repetitive behaviour which could be illustrative of the either Sensory Motor behaviours or insistence on sameness.

When considering the combination of subject (as available) and informant descriptions of colour affected behaviours with the subjects' experimental performance there was also a mix of findings. For some individuals, there was little to no correspondence between participant performances on the chromatic discrimination or colour naming tasks. However, there were some striking results when comparing the results of the colour preference to colour affected behaviours. For the most part, individuals' colour preference patterns reflected the valence of the parent reported colour affected behaviour, i.e. if they showed a strong preference for blue, there would also be colour affected behaviours that were related to blue. This notion lends support for the ecological valence theory of colour preference (Palmer & Schloss, 2010). This theory suggests that colour preferences occur because they are inherently tied to the valence that is ascribed to objects (see Chapter 4 for wider discussion). However, there was also a sub-group of participants in both the autism and WS groups, who displayed clear colour preferences in the absence of an informant reported colour affected behaviour. This pattern was also common when for the TD group. The results from these participants who show colour preference patterns without any colour affected behaviours suggest that object/action-associations for colours are not the only factor when forming colour preferences.

Recent changes to autism diagnosis in DSM-5 have meant that unusual or hypo-/hyper-reactivity to sensory input are now classified within the restricted and repetitive behaviours domain (American Psychiatric Association, 2013). As outlined in chapter 6, most colour affected behaviours could be classified as a restricted or repetitive behaviour or an unusual sensory interest. Many of the behaviours reported in these case studies do illustrate different levels of severity as outlined in DSM-V (American Psychiatric Association, 2013). In the case studies the severity ranged from level 1: inflexibility of actions causing disruption to functioning (e.g. Williams Syndrome Case Studies 2, 3), level 2: Distress in changes to actions (e.g. Williams Syndrome Case Study 1) and level 3: extreme difficulty in coping with change (e.g. Autism Case studies 1, 2 and 3). The nature of colour affected behaviours in participants from both groups took two forms. The first was that the behaviour reflected a restrictive or repetitive behaviour, e.g. insistence on wearing clothes of a colour or insistence on the room being a colour. These behaviours in many instances led to increased impairments, for example disruption to school preparation routines or consideration of colour in the family's everyday life. Similar restricted behaviours have been reported in another case study of adolescent with autism (A. K. Ludlow et al., 2014). Interestingly examples of these repetitive behaviours were also found in the WS group, but were less likely to be found in the TD group. Although not part of the "classical" WS profile (see Chapter 1 section 1.2.1), repetitive behaviours have been estimated to be present in up to 86% of adults with WS (Davies, Udwin, & Howlin, 1998). There is also evidence that young children with WS do score higher than typically developing controls on measures display repetitive behaviours when assessed using gold standard diagnostic measures

such as the ADOS or ADI-R (Lincoln, Searcy, Jones, & Lord, 2007). Furthermore, they found that when there is a comorbidity of autism and WS within the same individual there is no difference in their restricted and repetitive behaviour scores on the ADOS. There are also similar scores for restricted and repetitive behaviours for individuals with WS and with either autism or pervasive developmental disorder – not otherwise specified (Klein-Tasman, Phillips, Lord, Mervis, & Gallo, 2009). The relative rates of these restricted and repetitive behaviours in the Lincoln and colleagues (2007) and Klein-Tasman and colleagues (2009) were 15% and 45% respectively. This suggests that restricted and repetitive behaviours are present and common occurrence in WS. The data from this study give further support for this notion in response to colours, with estimates of colour affected behaviours being above those found by Klein-Tasman and colleagues (58% vs 45%). Although that does not mean that these colour affected behaviours have the same underlying aetiology in autism and WS. Also unlike the Klein-Tasman study's results, in this study there are significantly more colour affected behaviours in the autism group compared to the WS group. This may be due to the differences in how restricted and repetitive behaviours are recorded by the ADOS and the questionnaire used in this chapter. The ADOS repetitive measures are based upon observable (usually play based behaviours, depending on module), whereas the behaviours reported in the questionnaires would not be part of an ADOS assessment, and given that the participants parent is completing the questionnaire there is more chance for such behaviours to occur than during an ADOS assessment. This difference in measure may account for the differences in these rates. The WS and autism participants are also older in this study compared to those in the Klein-Tasman et al (2009) and Lincoln et al (2007). It is unknown the extent to which restricted and repetitive behaviours change over time, particularly in WS. There are reports that such behaviours reduce in individuals with autism as they progress from childhood into adulthood (Esbensen, Seltzer, Lam, & Bodfish, 2009) and are more likely to reduce for higher functioning individuals (Leekam, Prior, & Uljarevic, 2011). It is also important to note that there are relatively small sample sizes, differences in chronological and recruitment method used mean that it is not clear whether this difference would exist with more participants.

The second type of colour affected behaviour was an emotional response to colour. In some cases (e.g. Autism Case Study 3), there was a dislike of white, because it felt, "empty" or an avoidance of colours because they appeared dull (Autism Case Study2). In some between subject cases different emotions would be ascribed to certain colours, though there was no consistency in which colours were given emotions. These types of behaviours were less common than colour affected actions, but were still present in a subgroup of individuals. Previous work has found that adults with autism are less likely to ascribe emotion or intention to moving shapes (Abell, Happe, & Frith, 2000; Castelli, Frith, Happé, & Frith, 2002), yet here individuals with autism are spontaneously ascribing emotion to

colours. However, it should be noted that it is unknown whether this is not a typical colour affected behaviour compared to more restricted and repetitive behaviours and that the individuals who assign different emotions to colour reflect a minority response. Alternatively, it is possible that this emotional response to colour represents colour itself as an "unusual sensory interest". The individuals who displayed these emotional responses to colour also had other colour affected behaviours that could be classified as restricted or repetitive interests, so it is unclear whether these emotional responses to colour are symptomatic of more severe colour affected behaviours. A final possibility is that these emotional responses to colour are learnt from culture (e.g. blue is sad) or interventions that aim to teach emotions through colour (e.g. Picture Exchange System).

The results of this study are like previous qualitative findings of sensory processing in autism. Previous research on children and adults with autism has shown that there were both positive and negative responses to different sensory stimuli (Kirby et al., 2015; Robertson & Simmons, 2015). Participants in both the autism and WS groups this study displayed both positive and negative examples of colour affected behaviours, e.g. liking or disliking colours in clothing and other objects. When a colour affected behaviour reported, they were more likely to be for either positive *or* negative rather than for both positive *and* for the same behaviour. Another factor that has been identified is that there could be a physical reaction to unpleasant stimuli. There was only specific example of this where a colour was reported to be experienced as a physical symptom. This was not described as a physical discomfort but as a synesthetic response (Case Study 312, also see below for wider discussion). The previous studies by Kirby and colleagues (2015) and Robertson and Simmons (2015) did not report a physical response specific to coloured stimuli. It is possible that colour itself may not induce a physical response compared to auditory stimuli of a frequency/pitch or bright lights. Participants in both the autism and WS groups who were reported to have colour affected behaviours usually the behaviour appeared to lead to some level of control of the sensory (colour) experience such as refusing to wear clothes of a colour. This is also in line with previous studies on children and adults with autism (Ashburner, Bennett, Rodger, & Ziviani, 2013; Dickie, Baranek, Schultz, Watson, & McComish, 2009; Robertson & Simmons, 2015). It is important to note that both autism and WS displayed these characteristics so it is unclear the extent to which these behaviours are specific to autism and WS or are reflective of atypical development. This suggests that responses to sensory stimuli is the same in children with WS as well as children with autism, meaning that there is not condition specificity in their qualitative responses to sensory stimuli as captured here, but that there is variability between condition in the response to types of sensory modality, e.g. auditory sensitivities are relatively more common in WS than autism, which also display considerable heterogeneity *within* each condition.

The types of colour affected behaviours also reflect some of the existing literature and clinical guidelines on sensory sensitivities in autism. The lack of a difference for colour affected behaviours relating to food between any of the groups is similar to a recent systematic review have shown that colour is not a primary factor concerning food selectivity or fussiness for children with autism (Hubbard, Anderson, Curtin, Must, & Bandini, 2014; Marí-Bauset, Zazpe, Mari-Sanchis, Llopis-González, & Morales-Suárez-Varela, 2013). Other factors such as texture, taste and appearance of food were the most important factors when addressing food related behaviours for individuals with autism (Marí-Bauset et al., 2013). In another study of children age between 3-13 with an autism diagnosis, colour specific related food behaviour has identified in approximately only 15% of total food related behavioural problems (Hubbard et al., 2014). The results of the current study found that other behaviours were more common such as playing with toys, wearing clothes or the colour of rooms compared to food affected behaviours. Insistence on sameness of clothing has been identified as a repetitive behaviours and is a question on some questionnaires of repetitive behaviours (Honey, Leekam, Turner, & McConachie, 2007; Honey, Rodgers, & McConachie, 2012). The results of this study identified that the colour of clothing was the most frequent colour affected behaviour across all three groups. Like food, behavioural responses to clothing can be driven by several different and often complementary factors other than or in addition to colour, such as texture or fit. Unlike the research focussing on food, there has not been a wider investigation of clothing as a problematic behaviour. This means that at present it is unclear the extent to which colour is influential in affecting behaviour relating to clothing over other factors. The colour of rooms is another behaviour that has been recognised as being common in autism. The NICE guidelines 142 and 170 specifically suggests that the colour of walls should be considered as a potential source of sensory sensitivities for children, adolescents and adults with autism (NICE, 2012, 2013). Specifically, the NICE guideline 142 for adults with autism recommends the use of low arousal colours, such as cream, to reduce the risk of colour affected behaviours. The data from this study showed that adolescents with autism were significantly more likely to be affected by the colour of the room than typically developing children of a similar mental age, supporting the notion that room colour should be considered as part of sensory sensitivities of individuals with autism. However, the range of colours and the types of reported behaviours included a mix of seeking and avoiding particular room colour schemes. The colours reported were also a mix of low arousal colours (e.g. white) and higher arousal colours (e.g. yellow). Interestingly one young person reported that white made him/her feel “empty” and distressed. For others, colours such as blue or purple were reported to be relaxing. The heterogeneity in response to different colours across individuals does not necessarily lend support to use of a single colour for adults with autism as suggested in the NICE clinical guideline 142. There is no systematic evidence to support the use of a room colour, and the results of this study would not support this opinion. Instead being aware that the colour of walls may affect the behaviour of

individuals with autism is more appropriate, as is suggested by NICE guideline 170. The results of this relatively small sample of children with autism and WS suggests that understanding the colour-emotion associations of an individual are more important than prescribing the use of a colour. Future research should focus on these colour-emotion associations to identify the best use of colour for individuals with autism.

Unusual sensory interests or hyper-/hypo-reactivity to sensory experiences have been linked to restricted and repetitive behaviours in both autism and WS. Yet these studies only use questionnaires. Wigham and colleagues (2015) built a model that explored the relationship between sensory sensitivities and repetitive behaviours for young adolescents with autism using the Short Sensory Profile and the Repetitive Behaviour Questionnaire. They found that the sensory hypo-reactivity was related to repetitive motor behaviours and non-motor repetitive behaviours, whilst hyper-reactivity was only associated with non-motor repetitive behaviours (Wigham et al., 2015). Similar associations between the presence of sensory processing atypicalities and the presence of repetitive behaviours have also been found in a separate sample of young adolescents with autism (Chen et al., 2009). For children and adolescents with WS increased sensory sensitivities has been associated with an increase in repetitive behaviours using the Short Sensory Profile and Repetitive Behaviour Questionnaire (Janes et al., 2014; Riby et al., 2013). Further analysis of correlations between the subscales of the two questionnaires used found that there were significant relationships between sensory seeking and repetitive behaviours. The authors suggest that individuals with WS who are hypo-sensitive are more likely to display repetitive behaviours to help regulate their hyposensitivity. There was no evidence to support this relationship in the data of the WS participants in this study, however there was one case study (Appendix 3) where the participant was reported to seek out a colour (blue) because this was associated with hypersensitivity to auditory stimuli (sirens on emergency vehicles). Although a single case, this also highlights the importance of looking at sensory sensitivities across different sensory modalities to see how and whether this affects behaviour. The lack of a relationship in this study could be because there is not a direct measure of hypo-sensitivity in the questionnaire used in this study. It is also possible that because the questionnaire used in this study focuses on general visual function and colour, whereas the Short Sensory Profile and Repetitive Behaviours Questionnaire do not specifically focus on colour but assess more general behaviours that these different focuses of each questionnaire may result in a discrepancy in the results. Given that in WS visual sensitivity has been reported to be less affected (Janes et al., 2014), it is possible that the relationship between hypo-sensitivity and repetitive behaviours is only seen when considering a broader range of sensory modalities.

One participant in the autism group reported the experience of colour-taste synaesthesia. There have been other case reports of different types of synaesthesia in adults with autism and studies

reporting a proposed link with savantism (Baron-Cohen et al., 2007; Bor, Billington, & Baron-Cohen, 2008; Bouvet et al., 2014). Recently estimates of the rate of synaesthesia in adults with autism have been reported at just under three times higher than in adult controls using self-report questionnaires, prevalence rates were 18.9% vs 7.22% respectively (Baron-Cohen et al., 2013). Another study also reported similar rates of occurrences (23% of participants) of synaesthesia in adults with autism using an experimental test of colour-grapheme synaesthesia (Neufeld et al., 2013), suggesting that the prevalence of synaesthesia is increased for adults with autism compared to chronologically aged matched controls. In the survey study by Baron-Cohen and colleagues (2013) colour was the most frequently reported perception domain reported to be involved in individuals who were reported to have synaesthesia in large sample of adults with autism. In the autism group colour-taste was the fourth most common type of synaesthesia reported with it occurring in 4.26% of adults with autism. This study only reported just one case of colour related synaesthesia in an adolescent with autism. As the main aim of this study was to assess colour affected behaviours not synaesthesia it is not possible to comment on prevalence rates of synaesthesia in autism. However, unlike previous case studies, how the individual copes and responds to the additional sensory input that comes with sensory processing was not reported. In this study the participant who self-reported himself as having synaesthesia was reported to have numerous behavioural problems associated with the colour red, which was the colour that tasted sweet to them. Furthermore, more there were emotional responses to red, which were sadness and anger. The participant also commented on how the colour red hurt their pain. For this individual, there were no other colours that they reported to have a synesthetic response to. Their colour preference for reddish hues was also lower than other hues. This case study indicates a clear impact on their behaviour and experimental colour preference performance for the colour associated with their synaesthesia. Given that this participant is of adolescent age, it is possible that there are no behavioural problems reported in the adult case studies because the adults have developed an appropriate coping strategies. Indeed, sensory sensitivities as measured by the Sensory Profile are suggested to decrease with chronological age for individuals with autism (Boyd et al., 2009; Kern et al., 2006; Shattuck et al., 2007). A second possibility is that other types of synaesthesia, such as grapheme-colour are less likely to cause sensory overload. Future research on autism and synaesthesia should focus on the behavioural responses associated with the synesthetic responses as well as other cognitive/neural functions, and identify how individuals with autism respond with their synaesthesia across the lifespan.

There are several limitations to this study. Firstly, the groups were not matched on gender, IQ or chronological age. Instead the TD and autism/WS groups were of an approximate mental age (see chapter 2). Previous research in children, adolescents and adults with autism has identified a relationship between IQ and sensory sensitivities where individuals with lower IQ are more likely to

have higher sensory sensitivities (Crane, Goddard, & Pring, 2009; Leekam, Nieto, Libby, Wing, & Gould, 2007). However, there was no correlation identified between colour affected behaviours and IQ in this study for any group. The groups also differed in their chronological age: the autism and WS groups were older than the TD group. It's important to note that colour affected behaviours are likely to decrease with age in the TD group suggesting that the differences found in this study are likely to increase if chronological age matched controls were used. There were no significant correlations between colour affected behaviours and chronological age in the TD group, it is unclear whether this reflects the reduced age range and that colour affected behaviours happen at a younger age than tested in this study or that there are generally very few colour affected behaviours in typical development. The recruitment method also needs to be considered. It is possible that the parents of participants who responded to the recruitment letters did so because their child's behaviour is affected by colour. Given that not all individuals with autism or WS were reported to have colour affected behaviours suggests that this is not the case exclusive. Nonetheless despite these limitations, this is the first demonstration of the nature of colour affected behaviours in autism and WS and highlights the importance of including colour when considering the sensory affected behaviours.

## 7.6 Conclusion

This chapter used case studies provided individual case study comparisons between the parent reported colour affected behaviours and the individual's experimental test results. Colour preference curves could largely be interpreted within the context of the individual's behaviour that was influenced by colour. The results also demonstrate how colour affected behaviours in autism and WS also reflect restricted and repetitive behaviours. Further research is needed to further clarify the nature of these preliminary results within the context of the manifestation of restricted and repetitive behaviours and how these behaviours relate to cognitive processes in individuals with autism or WS. This chapter also highlights the benefits of using mixed methodologies to progress our understanding of the nature of sensory processing (including colour vision) in both typical and in individuals with specific disorders such as autism and WS.

# Chapter 8 - General Discussion

This thesis investigated colour perception in children with developmental disorders (either autism or Williams syndrome) and those who are developing typically. Specifically, the aim of the thesis was to give a wider characterisation of colour perception, from low level perceptual discrimination (Chapter 3), to higher level responses to colour (Chapters 4 and 5) and behavioural responses (Chapters 6 and 7). A mixed methods approach entailing psychophysics and behavioural tasks, parental questionnaires and case studies was used to establish a comprehensive representation of colour perception. The General Discussion chapter will first collate the results of the four experimental chapters, before discussing the way that the data can inform the behavioural phenotypes of colour perception in typical and atypical development.

## 8.1 Summary of Results

The first section will bring together the results from the experimental chapters to give a representation of colour perception in children with autism or Williams syndrome relative to mental age equivalent typically developing children. Different aspects of colour perception were assessed across the experimental chapters, including perceptual discrimination (Chapter 3: FM100 and Chromatic Contrast Discrimination Test (CCDT)), higher order responses to colour (Chapter 4: Colour Preference and Chapter 5: Colour Naming) and behavioural responses to colour (Chapter 6: Visual Response and Chapter 7: Case Studies). Since all participants completed all tasks relationships between these different tasks will also be considered.

*Table 8.1 -– Summary of results across all experiments* Summarising the major results for the Autism and Williams syndrome groups relative to their mental age TD control groups.

| Response to Colour | Experimental Chapter | Experimental Task | Autism | Williams syndrome |
|---|---|---|---|---|
| Perceptual Discrimination | 3 | FM100 | No difference from TD group | No difference from TD group |
| Perceptual Discrimination | 3 | Chromatic Contrast Discrimination Threshold Test | Poor chromatic discrimination, specifically for the “blue-yellow” axis | No difference from TD group |
| Higher Response | 4 | Colour Preference | Reduced cone-contrast weights | Reduced strength of colour preference and reduced cone-contrast weights |

| | | | | |
|---|---|---|---|---|
| Higher Response | 5 | Colour Naming | Similar frequency of use for different types of colour terms* | Reduced concordance in the Williams Syndrome, specifically for achromatic and desaturated colours. |
| Behavioural Responses | 6 | Visual Response Questionnaire | Higher scores of atypicality in general vision | Higher scores of atypicality in general vision |
| Behavioural Responses | 6 and 7 | Colour affected behaviours | Higher frequencies of colour affected behaviours compared to TD group | No difference from TD in frequency of colour affected |
| Behavioural Responses | 7 | Case Studies | Colour preference related to colour affected behaviours | Colour preference related to colour affected behaviours |

* Only seven participants with autism completed the colour naming task.

### 8.1.1 Profile of Colour Perception in Autism

The autism group showed differences from the TD group (see table 8-1.) in their colour perception profile. For perceptual discrimination, there was poorer chromatic discrimination compared to luminance discrimination on the CCDT (Chapter 3). This reduction in chromatic discrimination was primarily driven by poorer discrimination for the “blue-yellow” colour axis. However, for higher uses of colour there were little differences between the autism and TD groups. Colour preferences in the autism group showed qualitatively similar patterns at the group level that were in line with same sex TD patterns across hue, saturation and lightness variations, but showed less reliance on cone-contrasts (Chapter 4) and a subset of participants’ colour naming also used appropriate terms to describe colours (Chapter 5). There were an increased number and severity of behaviours affected by colour in the autism group compared to younger TD children (Chapter 6). Furthermore, there was a clear relationship between the affected colour and the relative peaks and troughs of that individual’s colour preference (Chapter 7). However, there was no relationship between individual’s chromatic discrimination and performance on colour preference or naming and frequency of colour affected behaviours.

To sum up, the results show that there is atypicality for perceptual colour discrimination and at the behavioural level but not for higher uses of colour such as colour preference and naming. Since there is no relationship between chromatic discrimination and colour affected behaviours, the results suggest that there is no relationship between these atypicalites in chromatic discrimination and the presence of behavioural problems associated with colour. There was evidence for a link between colour preferences and affected behaviours, however it is difficult to assess the directionality of this association (see Chapter 6 section 6.2 for further discussion).

### 8.1.2 Profile of Colour Perception in Williams Syndrome

Compared to the autism group, a different profile of colour perception was identified in the Williams syndrome group. There was no difference in chromatic discrimination (Chapter 3). However, there were differences in higher uses of colour. Colour preference was found to be highly variable (Chapter 4) with some individuals with Williams syndrome show strong colour preferences, whilst others showed no clear colour preference. This amount of variability was not observed in either the TD or autism group. Although at the group level colour preference patterns across hue, lightness and saturation variants of sex equivalent between the WS group and TD controls. There was atypicality of colour naming, specifically there was less concordance between individuals in the Williams syndrome group for achromatic and desaturated colours (Chapter 5). Furthermore, there were also relationships between chronological age and verbal ability to the complexity of colour term used to name colours. Furthermore, the average age at which parents reported their child reliably learning colours was older (5 and half years) than what would be expected for a TD child (2-3 years). The frequency of colour affected behaviours did not differ from TD controls (Chapter 6). However, when individuals were reported to have colour affected behaviours these were also likely to be associated with performance on the colour preference task (Chapter 7). There was no relationship between chromatic discrimination and performance on any other task or behaviour. The results suggest that the colour perception profile for Williams syndrome consists of perceptual discrimination that is in line with their mental age, however there is atypicality in higher uses of colour. Finally, although at the group level, there was no difference in the frequency of colour affected behaviours, when such behaviours were reported they were more severe than in the TD group.

### 8.1.3 Comparison of Colour Perception Profiles between Autism and Williams Syndrome

This thesis used a cross-syndrome comparison approach to study colour perception. The previous two sections outlined the colour perception profiles in autism and Williams syndrome. This section will compare the two syndromes together to address issues on the nature of condition-specific or

condition-general atypicality. From the summaries above there is an atypical colour perception profile in both autism and Williams syndrome, but the atypciality differs between the two conditions. In the autism group, there was poorer chromatic discrimination but this was not the case for the Williams syndrome group. There were also differences in higher uses of colour. The Williams syndrome group were found to have less within group agreement on desaturated and achromatic colours, while the subset of individuals with autism showed typical colour naming. For colour preference, both groups showed typical sex patterns of behaviour across hue, saturation and lightness variations, although the Williams syndrome group displayed greater inter-individual variation in their colour preference. Finally, there were more colour affected behaviours reported in the autism group than in the Williams syndrome group. However, the behavioural manifestation of these colour affected behaviours was similar when they occurred in both groups, e.g. refusal to wear clothes or eat foods of a certain colour.

In addition, for all groups there was no relationship between colour preference and the colour involved in a colour affected behaviour. The results of this thesis provide evidence that colour perception is atypical in both autism and Williams syndrome but that this atypicality varies between the two conditions. However, despite these differences there are still similarities for colour preference, and a relationship between colour preference and colour affected behaviours. The next section will assess these different colour profiles in relation to the wider literature and theories of visual and sensory processing atypicalities in both autism and Williams syndrome.

### 8.1.4 Relation to Wider Literature

This section will consider the results reported in chapters 3 in the context of the wider literature on visual perception, approaches to the study of sensory processing and how the present research might inform knowledge about for example applied uses of colour (such as in NICE clinical guidelines or advice for educational settings).

#### *8.1.4.1 Basic Science*

##### 8.1.4.1.1 Relation to existing Perceptual Accounts of Atypical Sensory Processing

This section will focus on how the results of the thesis fit into existing perceptual theories of atypical sensory processing in autism and Williams syndrome. This will include the results of chromatic discrimination (Chapter 3) as they directly assess perceptual discrimination, unlike the other experiments which relate to higher order processing of colour.

##### 8.1.4.1.2 Enhanced Perceptual Functioning and Weak Central Coherence

Weak central coherence theory posits that individuals with autism have a bias towards processing the local elements at the expense of the global picture (Happé & Frith, 2006). The extension of this notion is that there is “enhanced perceptual functioning” for simple percepts (Bertone & Faubert,

2006; Mottron & Burack, 2001; Mottron et al., 2006). The results from this thesis do not support this theory, as there was poorer chromatic discrimination in the autism group relative to typically developing mental age controls. This result is similar to previous studies on autism which have found poorer chromatic discrimination using similar methods (Franklin, Sowden, et al., 2010; Hurlbert et al., 2011; Koh et al., 2010). Recently, it has been suggested that there is a "perceptual factor" that is involved in processing sensory information (Meilleur et al., 2014). Meilleur and colleagues argue that this is analogous to the notion of intelligence, but for sensory processing. However few studies have compared multiple different perceptual tasks, or when studies have they often have different results between tasks (Spencer et al., 2000; Spencer & O'Brien, 2006). This means that it is unclear the extent to which the "perceptual factor" varies within and between and within sensory modalities.

In Williams syndrome, a weak central coherence has been linked to a preference for different local processing of features on face processing and visuospatial tasks (Bellugi, Lichtenberger, Mills, Galaburda, & Korenberg, 1999; Bellugi et al., 1988; Deruelle, Mancini, Livet, Cassé-Perrotb, & de Schonen, 1999; Deruelle, Rondan, Mancini, & Livet, 2006). However, there is little evidence for enhanced perceptual functioning in Williams syndrome, with performance either being impaired or similar to mental age. In this thesis, there was also no evidence for enhanced perceptual for Individuals with Williams syndrome, as chromatic discrimination was found to be in line with their mental age.

##### 8.1.4.1.3 Dorsal and Ventral Stream Visual Processing

The dorsal stream deficit hypothesis proposes that functions of the dorsal visual stream are more vulnerable to atypical development in children with developmental conditions whilst functions of the ventral stream are more likely to be intact due to slower development of this pathway, and has been proposed as a possible reason for differential visual function in both autism and Williams syndrome (Atkinson et al., 2006; Atkinson et al., 1997; Spencer et al., 2000). Yet support for this hypothesis is mixed with dorsal and ventral functions being found to be atypical in both conditions. (Atkinson, 2000; Atkinson et al., 2003; Atkinson et al., 2006; Atkinson et al., 1997; Cowie et al., 2012; M. Martens et al., 2008; Simmons et al., 2009), see Chapter 1 section 3 for more details. In this thesis, chromatic discrimination was used as a marker of ventral stream function. Poorer chromatic discrimination in the autism group fits is not in line with findings of intact ventral stream functions, such as superior orientation or form discrimination (Dickinson et al., 2016; Spencer et al., 2000). Nonetheless this finding is in line with other studies using different chromatic discrimination (Franklin, Sowden, et al., 2010; P. Heaton et al., 2008). On the surface these findings appear at odds. It is possible that an excitatory/inhibitory imbalance caused by the neurotransmitter GABA inhibits chromatic discrimination (including the koniocellular pathway) (Mecarelli, Rinalduzzi, & Accornero, 2001), but enhance form/orientation discrimination (Edden, Muthukumaraswamy, Freeman, &

Singh, 2009). In addition disruption of binocular rivalry has also been proposed to be linked to the presence of autistic traits and performance on binocular rivalry (Robertson et al., 2013; Robertson et al., 2016). However, the orientation/form/chromatic discrimination studies and this thesis did not take direct measurements of GABA activity, nor have the same set of participants completed form/orientation/chromatic discrimination tasks. Furthermore, it is unclear what the role, and at what point in the visual system GABA influences chromatic discrimination. For these reasons, it is difficult to say whether the dissociation of ventral stream functions is due to imbalances in GABA. Nonetheless it remains an interesting mechanism for future studies to investigate.

Williams syndrome is typically characterised by profound visuo-spatial deficits and other dorsal stream functions showing developmental immaturity, by comparison the ventral stream functions are relatively intact (Atkinson et al., 2003; Atkinson et al., 2006; Atkinson et al., 1997). In this thesis, there was no difference in chromatic discrimination between the Williams syndrome group and mental age control group equivalents in this thesis. Previous research has found that chromatic discrimination in Williams syndrome is in line with mental age controls, but poorer than chronological age matched controls, suggesting developmental delay (Farran et al., 2013). However this task used the FM100 which this thesis demonstrates is related to chromatic discrimination (see also (Cranwell et al., 2015)). The findings of this thesis expand upon the study conducted by Farran and colleagues (2013) using a psychophysical task that is independent of ability and found that chromatic discrimination was in line with mental age controls. The finding of this thesis gives further evidence for ventral stream function in line with mental age for individuals with Williams syndrome.

##### 8.1.4.1.4 Neural Correlates of Sensory Processing

As outlined previously there is evidence for different neural responses to processing visual information in autism and Williams syndrome and for processing colour (see Chapter 1 section 3). Whilst it is important to note that no neuro-imaging method was used in this thesis, the use of Eskew colour space in the CCDT means that performance can be related to specific anatomical pathways (see Chapter 2 section 2.6.1). There was poorer discrimination in the autism group for the blue-yellow chromatic discrimination. The blue-yellow axis has been found to map onto the koniocellular layer in the LGN and projects to layers 2,3 and 4α in the primary visual cortex (Hendry & Reid, 2000). These layers have been reported to have increased density of neurons in histological studies of adults with Williams syndrome (Galaburda & Bellugi, 2000; Galaburda et al., 2002), yet similar histological and voxel-based-morphometry studies in adults and a three year old child who had autism have found typical structure of the primary visual areas (Buxhoeveden et al., 2006; Nickl-Jockschat et al., 2012). However, there was no reduction in chromatic discrimination in the Williams syndrome group. This leads to a dissociation between the data observed in this study and the observed structure of primary visual cortex in autism and Williams syndrome. It's possible that other factors, such as

atypical anatomical and functional connectivity of primary visual areas, which have also been identified in autism may instead be driving this difference (see Chapter 1 section 4 for more details). There is a dissociation between the underlying neural structure and expected visual function in the both the autism and Williams syndrome group. There is one study that has investigated neural processing of colour in adults with autism. Fujita and colleagues (2011) used visual evoked potentials to investigate neural responses to chromatic (red-green) and achromatic gratings. They found that there was a longer latency to the chromatic but not achromatic grating, whilst there was no difference in the peak amplitude for either grating (Fujita et al., 2011). The results are partially compatible with those found in this thesis. The lack of a difference for the achromatic condition is the same results as found in chapter 3. However, there is difference for the red-green colour axis in the Fujita study but not here, whilst the visual evoked responses to the blue-yellow axis were not recorded but there are differences on this chromatic axis in this study. It is important to note the differences in methodology, there are differences in spatial frequency of stimuli, age of participants (adults v children/adolescents). Furthermore, there is also no evidence linking performance on psychophysical chromatic discrimination tasks and visual evoked responses, therefore although this deficit in blue-yellow discrimination is found, without further methods it is difficult to identify where this difference occurs within the visual system. These weaknesses between studies can be addressed by adopting a mixed methods approach to studying sensory processing to enable direct links to be made between neuro-imaging methods, psychophysics and behaviour. The next section will explore how the mixed methods approach used in this thesis can be used to resolve previous issues between previous studies on sensory processing.

#### *8.1.4.2 Mixed Methods Approach to Studying Sensory Processing*

This section will explore further how the approach to studying colour perception in this thesis can inform better design for future studies of sensory processing in typical and atypical development. The results of the thesis also impact on the notion of how to study sensory processing. There is a wide ranging yet unconnected literature of sensory processing in both typical and atypical development, where typically a single method is used to study an aspect of sensory processing that is ultimately limited by the choice of method. For example, the Sensory Profile may capture general sensory affected behaviours but it does not inform about the qualitative experience that the individual has. These studies range from questionnaires, psychophysical studies, neuroimaging and experimental studies of sensory cognition. Many of these studies are about sensory processing but there is a lack of cohesion in terminologies, results between methods.

##### 8.1.4.2.1 Terminology

One of the difficulties in studying sensory processing across different fields is the lack of a unified set of terminology. An example of this is the mismatch between hypo-/hyper-sensitivity between

psychophysics and behavioural questionnaires (e.g. Sensory Profile). In psychophysics, these terms relate to under or over responsive to a stimulus at the level of perceptual discrimination. In behavioural questionnaires, hyper-/hypo-responsivity are defined in responses in terms of behaviours. Crucially whether these questions are deemed hyper or hypo is dependent on the output of factor analyses and have not been informed by or linked to actual processing of sensory information (see Chapter 1 section 1.1 and Chapter 6 section 2 for further discussion on terminology). For example, consider the use of the term "sensitivities". In psychophysics, this relates to the ability to detect and discriminate stimuli. However in questionnaires sensitivity (usually twinned with reactivity) this relates to the likelihood of an individual displaying a behaviour that is the result of a low threshold to sensory information (Dunn, 1999). Another example of this is the terminology mismatch between fields is in the use of hypo-/hyper-sensitivity between psychophysics and behavioural questionnaires (e.g. Sensory Profile).

Previous studies of sensory processing typically use only one method. Yet there are different aspects of sensory processing both within and between modalities. The mixed methods approach utilised in this thesis enables a holistic investigation of a more rounded study of colour perception, one aspect of sensory processing, across the same participants. Given that both autism and Williams syndrome are characterised by within-syndrome behavioural and cognitive heterogeneity (Little et al., 2013; Morris & Mervis, 2000; Porter & Coltheart, 2005; Waterhouse, 2013), cross sectional study of different aspects of sensory processing within or between different modalities may not appropriate as it is not possible to establish links between physiology and behaviour. It is important to capture this inter-individual variability for different aspects of sensory processing so that links between these different facets can be established. For example, in this thesis the autism group were found to have poorer chromatic discrimination, replicating previous results (Franklin, Sowden, et al., 2010; P. Heaton et al., 2008; Hurlbert et al., 2011), and increased colour affected behaviours in the same group (Chapter 6). It was found that colour preference was found to be related to the presence and nature of the colour affected behaviours but that chromatic discrimination was not. If these studies were conducted separately with different participants, then this association between behaviour and higher use of colour but not low-level perceptual discrimination would not have been identified. Furthermore, it could be wrongly interpreted that there is a relationship between chromatic discrimination and colour affected behaviours. It is important to assess these links between low-level perceptual discrimination, higher uses of this sensory information and the extent to which this impacts actual behaviour, in the case of this thesis the sensory modality was colour, but a similar approach could also be used for other sensory modalities. The use of a mixed methods approach in this thesis enables these associations between tasks and different aspects of sensory processing to be directly assessed, the application of this approach to the study of sensory processing would allow

for more comprehensive coverage of not just one aspect but possible underlying causes and associated between/within sensory modalities. While it is not possible to assess the direction of causality between the different methods, the use of a mixed methods approach has important ramifications for where to target potential interventions as well as assessing and advancing current theoretical models within the field. Few studies in visual perception in autism have used multiple methods, but when they are used they can reveal specific areas that may explain the location of atypicalities.

Colour offers a framework within which to do this because it can be studied at a perceptual, cognitive and behavioural level. Other work has attempted to do this, for example in autism establishing links between motion coherence and motor control (Milne et al., 2006) or between performance on motion coherence and biological motion tasks are also related to neural activity in V5 and severity of autism (Koldewyn, Whitney, & Rivera, 2011). Examples can also be found in studies on Williams syndrome that identify general visuospatial underlies specific rather than general motion coherence deficits (J.E. Reiss, James E. Hoffman, & Barbara Landau, 2005b). Yet very few studies attempt to link performance on such tasks to actual behaviour, or conversely parental questionnaires scores to experimental results within sensory processing, although mixed methods have been used in other areas such as anxiety or face processing (Dodd, Schniering, & Porter, 2009; Tager-Flusberg, Skwerer, & Joseph, 2006). Recent studies (Meilleur et al., 2014; Perreault, Habak, Lepore, Mottron, & Bertone, 2015) have attempted to link between different levels of visual function in autism within the same individuals by comparing low-level (e.g. perceptual discrimination) to mid-level (e.g. pattern matching). Meilleur et al (2014) found that performance on low-level auditory and visual perceptual discrimination was indicative of cognitive use of this information (block design/melody discrimination). Perrault and colleagues (2015) also found that the identification of radial frequency patterns by individuals with autism could be predicted by poor identification of luminance and that texture contour discrimination predicted performance on pattern identification, suggesting that abnormal low-level vision influences subsequent visual processing. However, neither of these studies attempt to link this to accounts of visual behaviours so it is unknown to what extent behaviours are affected by differences in low-level perceptual discrimination. A more holistic approach to studying sensory processing would facilitate a rounded representation of sensory processing and which factors (perceptual, cognitive, behavioural) contribute to atypical sensory response, and which factors are important for clinical applications (e.g. diagnostic criteria, interventions).

#### 8.1.4.2 Applied uses of colour

This section will apply the major findings of the thesis to various contexts. This includes the application of colour towards clinical, therapeutic and education.

##### 8.1.4.2.1 Clinical

One of the clinical applications of this thesis is to DSM-5 and the diagnosis of autism. Whilst the aim of this thesis was not to assess the validity of these categories, nonetheless the results of this thesis can be interpreted considering these categories. DSM-5 includes hyper-/hypo-reactivity to sensory input or atypical sensory interests as a diagnostic criteria for autism under the restricted and repetitive behaviours domain (American Psychiatric Association, 2013). However little is known about the nature of this atypical sensory processing and how these align with restricted and repetitive behaviours as a diagnostic criterion for autism. The results of this thesis suggest that there are atypicalities within the sensory domain of colour perception in adolescents with autism for perceptual discrimination (Chapter 3) and for colour affected behaviours (Chapter 6 and 7). The results of the parent and participant reported colour affected behaviours give some examples for both hyper-/hypo-reactivity to sensory input as well as unusual sensory inputs at various severity levels. It is important to note the distinction between hyper and hypo reactivity is also found for these colour affected behaviours. Similar case studies have also reported sensory affected behaviours reflecting restricted behaviours and insistence on sameness repetitive behaviours in adults and children with autism across multiple modalities (Kirby et al., 2015; A. K. Ludlow et al., 2014; Robertson & Simmons, 2015). Yet a wide variety manifestations of these behaviours, and it is unclear which behaviours should and how they would be represented within the DSM-V framework for restricted and repetitive behaviours domain. In this thesis, there was reduced chromatic discrimination in the autism group. There is limited research directly assessing either hyper-/hypo-performance on other perceptual discrimination across different modalities in autism (Marco, Hinkley, Hill, & Nagarajan, 2011; O'Connor, 2012; Simmons et al., 2009). The DSM-5 guidelines focus primarily on behaviour, but this ignores growing evidence of atypical perceptual discrimination in autism. However, it is important to note that there are not consistent findings between studies. For example in motion perception there is mixed evidence for motion coherence thresholds, but this can be explained by methodological and experimental power studies (see also Chapter 1 section 3) (Milne, Swettenham, & Campbell, 2005). This is a common picture among different visual functions and in other sensory modalities. This suggests that perceptual discrimination may not be useful as a diagnostic tool due to wide inter-individual variability and methodological variations. Furthermore, these findings are not linked to behaviour of the autism participants. Unlike most of these studies, this thesis assessed the link between perceptual discrimination and sensory affected behaviours within the same individuals, although there was no significant association between the two.

Assessment of the link between perceptual discrimination and sensory behaviours may lead to possible explanatory mechanisms for behaviours. Whilst this thesis only assessed this for colour, it is possible that such links exist between perceptual discrimination and behaviours in other visual functions (see Future Directions for Research for wider discussion).

In addition to DSM-5, the NICE guideline states for the need to use low arousal colours (e.g. cream) to be used in the environments of adults with autism (NICE, 2012) whilst other NICE guidelines for autism (e.g. CG170 for individuals with autism under 19) for use with children suggest that it is important to take into account environmental factors, of which colour is one (NICE, 2013). In the introduction, it was stated that there was no evidence to support such a specific use of colour as suggested, and the results from this thesis do not provide evidence for one colour. Colour preference results (Chapter 4) suggested that individuals with autism or Williams Syndrome displayed similar colour preferences to mental age typically developing controls, furthermore no child chose a low arousal colour as their favourite. However, when there is a colour affected behaviour these are more severe than in TD. Results from individuals with colour affected behaviours (Chapter 7) were not related to one specific type of colour, suggesting that there is not one colour which influences behaviour, which is in line with the more colour-general guidelines of CG170 rather than the colour-specific use of peach in the NICE guidelines for adults. Crucially, colours had both positive and negative influence on behaviours (see Chapters 6 and 7). The results of Chapters 6 and 7 this thesis support this notion as they demonstrate that 80% individuals with autism in this thesis had behaviours that were influenced by colour.

It has also been suggested that colour can be distracting to individuals in a classroom environment for children with Williams syndrome (Udwin et al., 1996). However, there was no difference in the frequency of colour affected behaviours between the Williams syndrome group and the TD groups suggesting that individuals with Williams syndrome being distracted by colour is the exception rather than the norm, certainly for a sample studied here.

#### 8.1.4.2.2 Education and Development

Another way in which colour has been applied is in Education. For example, colour naming is used as a marker of language development. In contrast to other linguistic categories, such as shape or objects which develop from 2 years, reliable colour naming develops later between 3 to 3.5 years for initial basic colour terms (Bornstein, 1985; Carey, 1978). This thesis found that colour naming was less consistent for achromatic or desaturated colours and developed at a later chronological age in the Williams syndrome group (Chapter 5). Despite language being characterised as a relative strength in Williams syndrome, language onset is often delayed and develops atypically (Brock, 2007; Capirci et al., 1996; Karmiloff-Smith et al., 1997). The atypical colour naming found in this thesis gives additional evidence to atypical language development in Williams syndrome for abstract concepts.

Previous research has identified delays in the development of spatial language (Mervis & John, 2010). Furthermore, it also has implications for the use of colour in school environments or interventions where there is a focus on colour (e.g. PECS). Unfortunately, there are not enough participants in the autism group (and young enough) to determine whether there are atypicalities in the acquisition of colour naming. Parents reported that on average children with Williams syndrome learnt colour terms at a similar age to typically developing children, however there was also subset of individuals who developed colour naming later than what would be expected in typical development (See chapter 5 for more in depth discussion)

## 8.2 Appraisal of Research

### 8.2.1 Strengths

A major strength of the thesis is that the same participants are used across tasks. This has enabled specific patterns of condition specific atypicalities in colour perception to be identified. In autism atypcialities were found for perceptual discrimination and behavioural responses to colour, whilst in Williams Syndrome there were atypicalities in higher uses of colour. Furthermore, links between different tasks revealed no associated between perceptual discrimination and other uses of colour. Related to this is the use of mixed methods to give a more comprehensive understanding of colour perception. The combination of quantitative and qualitative methods allowed for specific patterns to be assessed; for example, there was an association between colour preference and behaviours that were affected by colours. Such associations were only able to be established because the same participants and a mixed methods approach were used. Previous research on colour (and visual) perception in autism and Williams syndrome has either used different participants across a single colour experimental tasks (Farran et al., 2013; Franklin, Sowden, et al., 2010) or where the same participants have been used potential relationships between tasks has not been assessed (Farran et al., 2013; Franklin, Sowden, et al., 2008). Given that these links are assessed within this thesis, it represents a significant contribution to current knowledge of colour perception in autism and Williams syndrome from previous studies not only on colour perception but also on sensory processing and the importance of assessing links between perceptual discrimination and higher uses of this sensory information and behaviour. The thesis also follows the a recent trend of studies attempting to establish links between low-level visual perception and higher uses of this visual information (Meilleur et al., 2014; Perreault et al., 2015).

Another strength of the work in this thesis is the use of two clinical groups to enable specificity and condition-general issues to be addressed. Most previous developmental research usually compares aspects one clinical group against controls, yet between studies methods can vary leading to conflicting results which are not due to actual differences between the clinical and control groups. The benefit of this approach is that it enables condition-specific responses to be identified and

therefore not only furthers understanding of each condition but also typical and atypical development. In this thesis on different tasks there are dissociations between the autism and Williams syndrome groups. These dissociations display condition-specific atypicalities for various areas of colour (i.e. for perceptual discrimination, colour naming, and colour preference). It also allowed for similarities in responses to colour to be identified, e.g. when colour affected behaviours occur as examples of repetitive behaviours regardless of condition.

### 8.2.2 Limitations

One limitation of the research conducted in this thesis is the absence of a chronological age matched group for the autism and Williams syndrome groups. The differences in mental and chronological age in individuals with autism and Williams syndrome led to their chronological age being older than the participants in the mental age TD comparison group. In turn this discrepancy in CA may result in different task performance strategies, i.e. that there are chronological age-related compensation strategies that may have developed in the autism or Williams syndrome groups compared to their mental age TD controls. All the aspects of colour perception investigated in this thesis also show changes with chronological age. However, when comparing the results in the autism and WS groups on tasks in this thesis, with previous studies of tasks with similarly chronologically aged typically developing adolescents, both groups still show differences indicating that there is indeed differences colour perception in both clinical groups is not due to a different task strategy. In Chapter 3 chromatic discrimination thresholds for both the autism and Williams syndrome groups were significantly lower than what would be expected from their respective chronological ages from cross sectional studies of chromatic discrimination by Knoblauch and colleagues (2001). The sex-specific hue colour preference curves of the autism and Williams syndrome groups described in Chapter 4 were also similar to adolescent sex-specific hue preference curves from Ling and Hurlbert (2011), although the Williams syndrome group hue preference curves were still flatter. There is no comparative chronological age data for either the colour naming (Chapter 5) or the prevalence/behavioural phenotype of colour affected behaviours (Chapters 6 and 7). For the colour naming, whilst there is no adolescent data, adult colour naming is more consistent than the Williams syndrome group, suggesting that lower concordance of colour naming is atypical in Williams syndrome. The extent of colour affected behaviours in typically developing adolescents is unknown. Other repetitive behaviours reduce with age in typical development and it is possible that there could be some colour affected behaviours, for example adolescent males choosing not to wear pink clothing because pink is a “girls” colour, but other behaviours such as coloured rooms or food are less likely to continue into adolescence and adulthood. However, it is important to note that the phenotype of colour affected behaviours in the autism are Williams syndrome group have much

more profound impact on the day to day life of these individuals which is unlikely to be the case for male's refusing pink clothes.

The developmental trajectory method has been used to study developmental changes in clinical groups. Although the matched groups design is useful for identifying whether there is a difference between clinical and non-clinical groups on the experimental measure while controlling for either chronological or mental age, it does not describe when or how the differences occur. Given that autism and WS are both developmental conditions, the question of whether a difference with respect to TD children reflects either a deviancy or delay is crucial. For example whether there are differences between WS and Down Syndrome groups on number and vocabulary tasks differed depending on whether they are tested as adults or toddlers, i.e. there is within syndrome dissociation on task performance (Paterson et al., 1999). This highlights the importance of considering development itself when studying developmental populations and developmental conditions. The developmental trajectory approach places development itself as the primary focus of analysis. In comparison to the matched groups design this allows for greater focus on change over developmental time, i.e. how task performance develops over time regarding either chronological or mental age. The approach does this by using regression methods to link task performance with changes in chronological age or with a change in developmental ability (e.g. mental age) (Thomas et al., 2009). Thomas and colleagues (2009) suggest that this can be done either using multiple cross sections of TD children at different chronological ages or longitudinally studying the same participants across different chronological ages (the latter is a preference but comes with the downside of significant investments of time and money and is far less feasible). The method uses linear regression to determine any dependence of task performance on the desired developmental measure, for example, by assessing the dependence of chromatic discrimination on chronological age. Regression slopes and intercepts of task performance against chronological or mental age are compared between the TD and developmental condition groups. Different separate trajectories can be constructed for task performance across different cognitive domains. For example, developmental trajectories can be constructed based on ability (i.e. verbal or non-verbal ability) or chronological age with respect to task performance. The advantage of using a developmental trajectory approach is that it aims to establish links between performance with age (either chronological or mental). This provides a different type of characterisation of a clinical group, including whether there is a deviancy or delay in task performance. Differences between the intercept of task performance for TD and clinical groups suggest a different developmental onset for task performance, whilst a difference in gradient would suggest a different developmental rate (i.e. steep gradients are indicative of a faster developmental rate, compared to flatter gradients which would indicate slower developmental rate (Rice, 2004), see also (Thomas et al., 2009) for wider discussion. Although this approach requires

larger number of participants in the TD control group to compare the clinical group against. The developmental trajectories approach also assumes that the individuals within a group follow the same developmental trajectory. However, this assumption may mask heterogeneity within developmental conditions. For example there is variation between in infants and toddlers who develop autism in the onset of autism symptoms (Elsabbagh & Johnson, 2010; Johnson & Myers, 2007). Nonetheless it does allow exploration about aspects of the development of the specific skill under investigation and whether there is evidence of delay or deviancy in the developmental trajectories of both the autism and WS groups to an extent that is not covered in this thesis.

A second limitation relates to consistency of task performance. Consistency here relates to the same performance on a task in a short period as well as within an individual's development over their lifespan. For example, consistency of colour naming can be calculated at a single time point, i.e. how consistent is a particular coloured patch named after being presented multiple times, or after substantial time has been passed, i.e. does an individual give the same name given to the same coloured patch but presented a year apart. This question can also be asked of the colour preference chapter. Studies have found developmental changes for colour and other visual functions from young childhood into adolescence in both typical and atypical development (Knoblauch et al., 2001; Simmons et al., 2009). For example in autism there is evidence for improvements in identification of biological motion (Hubert et al., 2007; Murphy, Brady, Fitzgerald, & Troje, 2009) However for these studies on visual perception (not just motion perception) in autism, it is difficult to appraise and compare these studies due to different ability ranges, different characterisations of autism, matching on mental or chronological age and usually group matching rather than individual matching, which often lead to conflicting results (Milne et al., 2005; Simmons et al., 2009).

The studies on Williams Syndrome convey a different picture. There is a wide evidence for a dissociation between ventral and dorsal stream visual functions in Williams Syndrome (Atkinson et al., 2001; Atkinson et al., 2006; M. Martens et al., 2008). Despite poorer dorsal stream function at all ages relative to chronological age typically developing controls, for example there is still developmental change as adults with Williams syndrome outperform children with Williams syndrome on motion discrimination and biological motion tasks. when comparing children and adults with Williams syndrome will outperform young children with Williams Syndrome on motion discrimination and biological motion tasks (Atkinson et al., 2006; Reiss et al., 2005a). These results from other visual functions show that there are changes associated with chronological age but that they are still below that of individuals without Williams syndrome. Within the context of this thesis it is also important to note that repetitive behaviours, colour preferences and colour naming all change with chronological age (Esbensen et al., 2009; Ling & Hurlbert, 2011; Richler, Huerta, Bishop, & Lord, 2010). In relation to this thesis it is unknown whether the differences in colour perception in autism

and Williams syndrome continue into adulthood or whether they are consistent in the same individuals. There is minimal research on colour perception in adults with autism (one conference abstract) and no direct studies in Williams syndrome. An example of this changes in colour affected behaviours can be found in the Chapter 7, where there was a recent development of colour affected behaviours for an adolescent with autism. This participant's colour preferences reflect the colour that affected their behaviour, yet it is not known whether their colour preference would have been the same or different without this affected behaviour. This limitation is also applicable to the wider body of research on colour and visual perception, as well as typical and atypical development. Future research needs to move on consider both inter- and within-individual variability to gain a better grounding of how different individual differences affect performance on colour perception tasks.

## 8.3 Future Directions for Research

The research conducted in this thesis leads to some suggestions for future directions of research. The first of these is the use of a mixed methods approach to studying sensory processing to enable more comprehensive study of sensory processing and links between different aspects of sensory processing. For example, this thesis focused upon low-level perceptual processing, higher uses of sensory information and behavioural responses to colour, but this approach could be adopted for other sensory modalities and visual functions. Furthermore, adopting this mixed methods approach to sensory processing will also enable different measurements of sensory processing to be validated against each other and aid in this understanding. In this thesis, interviews with parents/participants with colour affected behaviours were reflected by performance in the colour preference experiment, as well as to validate the new bespoke questionnaire on colour affected behaviours. Regarding the colour preference result, this finding helps to further explain the theoretical understanding of colour preferences but also help to link experimental findings to the behavioural phenotype in those individuals. Future studies can expand on this notion for other sensory modalities and to assess the relationship between low-level sensory functioning and behaviour.

It is also important to consider variability across within and different sensory modalities. This thesis focused upon colour but it is not known, how colour perception relates to other visual functions of other sensory modalities. However, in order to do this multiple tasks need to be used. At present studies of visual perception assessing multiple visual functions in autism and Williams syndrome are uncommon. Previous examples of these studies have found different performance between motion/form coherence visual tasks in both autism and Williams syndrome (Atkinson et al., 2001; Atkinson et al., 1997; Spencer et al., 2000) or show poor performance across different visual tasks (Spencer & O'Brien, 2006). This highlights the importance of considering multiple visual functions,

especially in autism and Williams syndrome. Yet it is unclear, how colour relates to other visual modalities such as form, orientation or motion in the same individuals. Future studies should aim to make links between colour and other visual functions to establish a rounded visual profile in both typical and atypically developing populations. Furthermore, this such studies could extend the "*perceptual factor*" proposed by Meilleur and colleagues (2014) to assess general and specific visual function. A goal for future research should also be to further extend this to establishing the presence, or absence, of links between performance on such tasks, sensory affected behaviours and repetitive behaviours.

A second line of future research comes from the use of colour in a clinical setting. The results of the colour preference and discrimination provide several possible avenues for potentially improving the efficacy of coloured overlays and sensory rooms or toys, particularly for children where colour affects their behaviour. For example, the colour preference results suggest that particular coloured toys may be more popular for males and females with either autism or Williams syndrome. From chapter 6 and 7 there were also numerous colours that also had positive effects on the individual, such as reducing anxiety. However, there was no single colour that was found to have a positive effect, demonstrating the inter-individual variability within the group. Secondly, despite widespread use in SEN schools, it is unclear how well or for who these sensory based interventions work for. Since colour is predominately involved in these interventions, the colour tasks in this thesis could identify individuals who these interventions may be more likely to work for. Indeed, there were individuals in this sample who had used sensory rooms or coloured glasses and had coloured affected behaviours, but the small sample size makes it difficult to generalise beyond these few cases. Future studies would be able to look at this possible relationship and determine whether the presence or absence of colour affected behaviours affects the efficacy of such treatments.

## 8.4 Conclusion

The present research suggested that colour perception was atypical in children and adolescents with either autism or Williams syndrome relative to mental age controls, but this atypicality varied between conditions. In autism, atypical colour perception was characterised by poorer chromatic discrimination and higher incidents colour affected behaviours, but higher uses of colour (colour preference and colour naming) was in line with their mental age. Colour perception in Williams syndrome was characterised by atypical complex uses of colour (higher inter-individual variability and less within group agreement on naming colours, but their chromatic discrimination and the prevalence of colour affected behaviours was in line with their mental age. This thesis demonstrates the value of adopting cross-syndrome and mixed methods approaches to identify condition-specific atypicalities in colour perception.

# Appendices

## Appendix 1 – Parental Information Sheet

An example of the recruitment information sheets and consent forms used in this thesis. This variant is for TD children. Similar forms were sent out to the autism and WS participants with minor changes.

**Colour Perception Study Information Sheet**

Dear Parent/Guardian,

### Who is conducting the research study, and where is it being conducted?

We are researchers from Newcastle University investigating colour perception in children. We would like to invite your child to participate in our research study. We are delighted that **Head-teacher Name** of **School name** has agreed to allow us to invite you to take part in our research study.

### Why is this study being done?

Colour is important in everyday life; it helps us to decide what clothes we like to wear or the food that we may like to eat. Understanding how children see colours will help us to see how they may use colours in understanding emotions and social situations.

### Why have I been chosen?

You are being invited for your child to participate because she or he is in mainstream education and is aged between 4 and 8 years old.

### Do I have to take part?

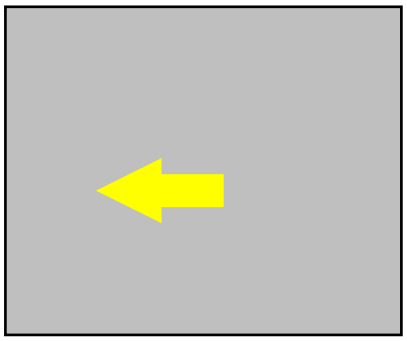
No. You can decide whether or not you would like your child to take part. If you agree to take part, and later change your mind, you can stop at any time. You don't have to give a reason.

### What do I have to do if I am in this research study?

If you agree for your child to participate in this study, please return the Permission Slip to your child's teacher by **Date**. Your child will complete 3 short one-to-one sessions with researchers. These sessions will take place on different days. All assessments are designed to be enjoyable for your child.

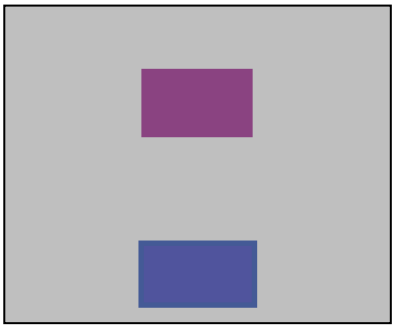
Colour Perception Assessments (two 30 minute sessions): We will assess your child's colour perception using computer-based, paper-based, and manual sorting tasks, that are designed as games. These tasks will investigate your child's ability to discriminate between different colours and his or her emotional responses to colour.

One game will look a bit like this:



Which way is the arrow pointing?

The other game will look like this:



What colour do you like best?

Behavioural Tasks (One 30 minute session): These will measure your child's non-verbal and language abilities. These will include simple tasks such as naming pictures, using coloured blocks to copy patterns and answering simple questions.

You will also be asked to complete two simple short questionnaires about the colour preferences and social behaviour of your child. (Total time 15 minutes)

## Can I talk to someone before agreeing to take part?

Yes, If you have any questions or would like to know more about this research study, please do not hesitate to contact Matthew Cranwell on **0191 208 3466** or by email at **m.b.cranwell@ncl.ac.uk**

If you and your child would like to take part in this study, then please fill in the form and return it back to your child's teacher. We will then arrange dates for your child to participate with their teacher, and will send you the questionnaires via your child's teacher, to be returned directly to us.

**Thank your for reading this letter!**

Yours sincerely;

Professor Anya Hulbert MD PhD (Professor of Visual Neuroscience), Matthew Cranwell BSc (PhD Student),

**Colour Perception Study Permission Slip**

Please include my child _____ (Child's Name) in this study.

Signature of Parent/Guardian:_____

If you feel your child is able to make an informed decision about taking part, please ask them to sign their name below in addition to the Parent/Guardian signature above.

Signature of Child: _____

Child's Date of Birth:_____

I understand that my child can stop the assessments at any time.

☐ Yes ☐ No

I understand that if the study is published there will be no information identifying my child in the publication.

☐ Yes ☐ No

I understand that all information will remain confidential and anonymous in line with the Data Protection Act. Personal information will not be kept beyond 10 years.

Yes ☐ No ☐

I would like to receive a report of my child's performance on standardized behavioural measures.

☐ Yes ☐ No

I would like the school to receive a copy of my child's performance on standardized behavioural measures.

☐ Yes ☐ No

Date:_____

## Appendix 2 – Colour Checker HSL Values

The colorimetric properties of the Macbeth ColorChecker Chart that was used in the colour preference experiment in Chapter 4.

| ColourChecker Patch | Hue | Saturation | Lightness |
|---|---|---|---|
| Dark Skin | 0.6166 | 0.719 | 66.6471 |
| Light Skin | 0.7065 | 0.5343 | 105.2377 |
| Blue Sky | 4.2172 | 0.6667 | 83.4424 |
| Foliage | 1.988 | 0.7083 | 73.4373 |
| Blue Flower | 4.5996 | 0.633 | 88.5271 |
| Bluish green | 2.892 | 0.5938 | 113.6132 |
| Orange | 0.6616 | 1.4506 | 100.9587 |
| Purplish blue | 4.5205 | 1.5516 | 69.1119 |
| Moderate red | 0.1631 | 1.4188 | 82.7358 |
| Purple | 5.1148 | 0.9872 | 51.0749 |
| Yellow green | 1.7090 | 0.9775 | 88.8437 |
| Orange Yellow | 0.9832 | 1.1765 | 111.3010 |
| Blue | 4.6035 | 2.3429 | 50.6872 |
| Green | 2.2605 | 1.1336 | 86.5040 |
| Red | 0.2062 | 2.2285 | 66.2644 |
| Yellow | 1.2883 | 1.0903 | 124.6702 |
| Magenta | 0.5296 | 0.4879 | 80.3667 |
| Cyan | 3.8622 | 0.996 | 83.1918 |
| White | 2.1555 | 0.0905 | 148.7848 |
| Neutral 8 | 2.251 | 0.086 | 121.0732 |
| Neutral 6.5 | 2.1449 | 0.0659 | 96.6155 |
| Neutral 5 | 2.4209 | 0.0955 | 79.9917 |
| Neutral 3.5 | 2.5522 | 0.0858 | 57.5594 |
| Black | 2.7694 | 0.0564 | 43.993 |

## Appendix 3 – Colour Preference Hue Comparisons

### Autism Analysis

The main effect of hue was further explored using Bonferroni corrected post hoc t-tests. These revealed that the reddish hue was significantly less preferred than Greenish hue, $t$ (34) = 4.91, $p$ < 0.001, and Bluish hue, $t$ (34) = 4.45, $p$ < 0.001. Brownish hues was also significantly less preferred than Greenish Hue, $t$ (34) = 4.09, $p$ < 0.001. Reddish hue was significantly more preferred than greenish hue, $t$ (34) = 3.7, $p$ < 0.001 and Purplish, $t$ (34) = 3.77, $p$ < 0.001.

Further post hoc t-tests revealed that significant differences between colours between sexes. Males were found to significantly prefer brownish hues, $t$ (34) = 2.98, $p$ < 0.005, and reddish hues, $t$ (34) = 3.78, $p$ < 0.001 relative to females. Whilst females significantly preferred colours purplish hue, $t$ (34) = 4.23, $p$ < 0.001 relative to males.

### Williams Syndrome Analysis

Further exploration of the main effects of hue and hue by sex interactions. To explore this post hoc t-tests with a Bonferroni corrected p-value were conducted for comparisons between the different hues. Reddish hues was significantly less preferred than both greenish hues, $t$ (51) = 3.4, $p$<0.001, and, $t$ (51) = 3.68, $p$<0.001. Brown hues was also significantly less preferred than greenish, $t$ (51) = 3.51, $p$<0.001, and C4, $t$ (51) = 3.42, $p$<0.001. Post hoc t-tests found that brown hues was preferred significantly more by males, $t$ (50) = 3.69, $p$<0.001, whilst colour purple hues were preferred significantly more by females, $t$ (51) = 5.46, $p$<0.001.

## Appendix 4 – Parental Questionnaire Version 1

**Section A:** Visual responses and sensitivities. *Please circle best answer.*

1. Does your child like bright lights?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

2. Do bright lights bother your child (even after having enough time to adjust to them)?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

3. Does your child like to be near bright lights?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

4. Does your child try to get away from bright lights?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

5. Does your child enjoy watching spinning objects?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

6. Does your child prefer to be in the dark?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

7. Is your child happy to be in the dark?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

8. Does your child become frustrated when trying to find objects in competing backgrounds (for example, a cluttered drawer)?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

9. Does your child have difficulty in putting puzzles together (compared to same age children)?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

10. Does your child cover their eyes or squint to protect their eyes from light?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

11. Does your child look carefully or intensely at objects/people?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

12. Does your child really like to look at one special object?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

If sometimes, most of the time or always, what is the object?

_____

13. Does your child have a hard time finding objects in competing backgrounds?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

14. Does your child have difficulty in identifying moving objects against a background?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

15. Does your child notice visual changes in a room (for example turning on/off a light)?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

16. Is your child bothered by visual changes in a room?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

17. Does your child have difficulty reading words from a book or computer screen?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

18. Is your child able to colour within the lines?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

19. Does your child have difficulty writing on a line?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

20. Does your child have difficulty coordinating actions (e.g. catching a ball)

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

21. When watching television or on the computer does your child sit near to the screen?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

22. Does your child have difficulty seeing things near to them?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

23. Does your child have difficulty seeing things that are far away from them?

*Not at all* *Very occasionally* *Sometimes* *Most of the time* *Always*

**Section B:** Colour preference. Please circle true or false for each statement. If *TRUE* or *SOMETIMES TRUE* please write in the colour.

24. My child has a favourite colour. *True* *Sometimes true* *False*

My child's favourite colour is

_____.

25. My child really dislikes a particular colour. *True* *Sometimes true* *False*

The colour my child dislikes is

_____.

26. Certain colours make my child feel sad. *True* *Sometimes true* *False*

These colours are

_____.

27. Certain colours make my child feel happy. *True* *Sometimes true* *False*

These colours are

_____

28. Certain colours excite my child. *True* *Sometimes true* *False*

These colours are

_____.

29. Certain colours help to relax my child. *True* *Sometimes true* *False*

These colours are

_____.

29. My child avoids certain colours. *True* *Sometimes true* *False*

These colours are

_____.

30. My child likes particular colour combinations. *True* *Sometimes true* *False*

These colour combinations are

_____

31. My child DISLIKES particular colour combinations. *True* *Sometimes true* *False*

These colour combinations are

_____

32. My child likes to eat food that is a particular colour. *True* *Sometimes true* *False*

The food colour my child likes to eat is

_____.

33. My child does NOT like to eat food that is a particular colour. *True* *Sometimes true* *False*

The food colour my child does NOT like to eat is

_____.

34. My child likes to be in a room that is a certain colour. *True* *Sometimes true* *False*

The room colour my child likes is

_____.

35. My child does NOT like to be in a room that is a certain colour. *True* *Sometimes true* *False*

The room colour my child does NOT like is

_____.

36. My child likes to wear clothes that are a certain colour. *True* *Sometimes true* *False*

The colour of clothes my child likes is

_____.

37. My child does NOT like to wear clothes that are a certain colour. *True* *Sometimes true* *False*

The colour of clothes my child does NOT like is

_____.

38. There are toys that my child really likes because of their colour. *True* *Sometimes true* *False*

The toy colour that my child really likes is

_____.

39. There are toys that my child really does NOT like because of their colour. *True* *Sometimes true* *False*

The toy colour that my child really does NOT like is

_____.

40. My child uses a particular colour or colours more than others

when colouring-in pictures. *True* *Sometimes true* *False*

These colours are

_____.

41. My child uses a particular colour or colours more than others

when drawing or painting. *True* *Sometimes true* *False*

These colours are

_____.

42. If there are other instances where the colour of something makes your child happy or upset, please describe.

_____

_____

_____

**Section C:** Colour naming. *Please answer each statement by circling the appropriate choice, or writing in.*

43. Does your child use unusual colour names/terms? *Yes* *Sometimes* *No*

If yes, in what way are the colour names unusual?

_____

_____

_____

44. Do your child's colour terms disagree with the terms normally used (e.g. they say "brown" instead of "green")?

_____

45. Are your child's colour terms more elaborate than usual (e.g. they say "light magenta" instead of "pink")?

_____

46. Are your child's colour terms less elaborate than usual (e.g. they say "red" instead of "magenta" or "burgundy")

_____

47. Does your child reliably label colours correctly? *Yes* *No* *Sometimes*

If yes, at what age did your child begin to reliably label colours correctly?

Age:_____

48. Does your child have difficulty in naming colours (e.g. takes a long time to name colours)?

*Yes* *No* *Sometimes*

49. Does your child have difficulty in describing the colours of objects accurately?

*Yes* *No* *Sometimes*

**Section D:** Family history of colour blindness.

50. Are there (or have there been) any incidences of colour blindness in your family? *Yes* *No*

51. If yes, how was/is the person related to you?

_____

_____

**Section E:** Child's language. Please circle the appropriate answer, or write in response.

52. Is your child spoken to in any languages other than English? *Yes* *No*

If yes, what language other language are they spoken to in?

_____

53. Does your child speak any languages fluently other than English? *Yes* *No*

If yes, what other language do they speak?

_____

**Section F:** Medical history **(OPTIONAL)**

54. Has your child been diagnosed with any behavioural problems? *Yes* *No*

55. Does your child have any assistance to help their vision? (e.g. glasses) *Yes* *No*

If yes, what assistance have they been prescribed?

_____

56. Has your child ever had to wear an eye patch to help their vision? *Yes* *No*

Thank you for your help. If you have any further comments please add them below.

## Appendix 5 – Parental Questionnaire Version 2

**Section A:** Sensitivity to vision.

For section A, answers please circle the appropriate answer in the Last 6 Months column. The letters correspond to following categories: N=**N**ever, O=**O**ccasionally, S=**S**ometimes, F=**F**requently, A=**A**lways. If you child has shown the behaviour but **not** in the last 6 months, then please indicate this in the Lifetime column. Please write your child's approximate age in years and months when they showed this behaviour (where possible).

| | Question | Last 6 Months | | | | | Lifetime |
|---|---|---|---|---|---|---|---|
| 1. | Does your child like bright lights? | N | O | S | F | A | |
| 2. | Do bright lights bother your child (even after having enough time to adjust to them)? | N | O | S | F | A | |
| 3. | Does your child try to get near bright lights? | N | O | S | F | A | |
| 4. | Does your child try to get away from bright lights? | N | O | S | F | A | |
| 5. | Does your child enjoy watching spinning objects? | N | O | S | F | A | |
| 6. | Does your child prefer to be in the dark? | N | O | S | F | A | |
| 7. | Is your child happy to be in the dark? | N | O | S | F | A | |
| 8. | Does your child have difficulty in putting puzzles together? (Compared to same age children) | N | O | S | F | A | |
| - | Is this because of the movements involved? | Yes | | | | No | Yes<br>No |
| 9. | Does your child cover their eyes or squint to protect their eyes from light? | N | O | S | F | A | |
| 10. | Does your child look very carefully or intensely at objects/people? | N | O | S | F | A | |
| 11. | Does your child really like to look at one special object? | N | O | S | F | A | |
| - | If sometimes, frequently or always what is it? | | | | | | |

| | | | | | | | | |
|---|---|---|---|---|---|---|---|---|
| 12. | Does your child have a hard time finding objects in competing backgrounds? | N | O | S | F | A | | |
| 13. | Does your child have difficulty in identifying moving objects against a background? (e.g. a bird flying or moving car) | N | O | S | F | A | | |
| 14. | Does your child notice visual changes in a room? (e.g. turning on/off a light) | N | O | S | F | A | | |
| 15. | Is your child bothered by visual changes in a room? | N | O | S | F | A | | |
| 16. | Does your child have difficulty reading words from a book or computer screen? | N | O | S | F | A | | |
| 17. | Is your child able to colour within the lines? | N | O | S | F | A | | |
| - | Is this because of the movements involved? | Yes | | | | No | Yes<br>No | |
| 18. | Does your child have difficulty writing on a line? | N | O | S | F | A | | |
| - | Is this because of the movements involved? | Yes | | | | No | Yes<br>No | |
| 19. | Does your child have difficulty in coordinating actions (e.g. catching a ball) | N | O | S | F | A | | |
| - | Is this because of the movements involved? | Yes | | | | No | | |
| 20. | When watching television or on the computer does your child sit near to the screen? | N | O | S | F | A | | |
| 21. | Does your child have difficulty seeing things near to them? | N | O | S | F | A | | |
| 22. | Does your child have difficulty seeing things that are far away from them? | N | O | S | F | A | | |

**Section B:** Colour preference. Please select all the statements that apply.

23. My child has a favourite colour. True

False

My child's favourite colour is _____.

I know this is my child's favourite colour

because_____

My child consistently uses the same name for his/her favourite colour True

False

The name (or names) my child uses for his/her favourite colour is

_____

24. Are there colours that your child really doesn't like? True

False

- If yes, what is it?

_____

- I know my child dislikes _____

because_____

25. Does your child like to eat food that is a certain colour? True

False

- If true, what is it? What does your child do?

_____

26. Does your child dislike eating food of a certain colour? True

False

- If true, what is it? What does your child do?

_____

27. Does your child like being in a room that is a certain colour? True

False

- If true, what colour is it? How does your child react?

_____

28. Does your child dislike being in a room that is a certain colour? True False

- If true, what colour is it? How does your child react?

_____

29. Does your child insist on wearing clothes of a certain colour? Yes No

- If yes, what is it? What do they do?

_____

30. Does your child refuse or really not like wearing clothes of a certain colour? Yes No

- If yes, what is it? What do they do?

_____

31. Are there toys that your child really likes to play with because of the colour of the toy rather than the types of toy?

True False

- If true, what colour is it? What is it that your child does with the toy that makes you think it is to do with the colour of the toy?

_____

_____

32. There are toys that your child refuses or really doesn't like to play with because of their colour of the toy rather than the type of toy?

True False

- If true, what colour is it? What is it that your child does with the toy that makes you think it is to do with the colour of the toy?

_____

_____

33. Can you think of any other times when the colour of something seems to have been particularly important to your child? Did this reaction to colour seem to make your child happy or cause your child to be upset or distressed.

Please describe the situation:

_____

_____

- When was this? How frequently did it happen and how long did it last? How did your child react on these occasions?

_____

_____

_____

34. When your child colours in pictures, do they use a particular colour more than others?

Yes No Sometimes,

- If yes or sometimes, what colours do they use?

_____

35. When drawing or painting does your child use some colours more than others?

Yes No Sometimes

- If yes or sometimes, what colours do they use?

_____

36. Does your child have any unusual colour names or terms for particular colours?

Yes No Sometimes

- If yes, what are the colour names or terms that your child uses?

_____

37. Does your child ever disagree with the terms normally used for one or more colours (e.g. they say “brown” instead of “green”)?

Yes No Sometimes

- If yes or sometimes please give examples.

_____

39. Does your child ever use terms that are more elaborate than usual (e.g. they say “light magenta” instead of “pink”)?

Yes No Sometimes

- If yes or sometimes please give examples.

_____

40. Is your child ever less elaborate than usual (e.g. they might say “red” instead of “magenta” or “burgundy”)

Yes No Sometimes

- If yes or sometimes please give examples.

_____

41. Is your child able to label colours reliably and correctly?

Yes No Sometimes

- If yes, at what age did your child achieve this skill consistently without making mistakes?

- Age (In years and months, if possible):_____

- If no, are there particular colours that your child does not label correctly (e.g. primary colours such as red, green, blue, yellow, or more complex colours such as mauve, beige, turquoise?

_____

_____

42. Does your child have difficulty in naming colours (e.g. takes a long time to name colours)?

Yes No Sometimes

- If yes or sometimes, which colours?

_____

43. Does your child have difficulty in describing the colours of objects accurately?

Yes No Sometimes

- If yes or sometimes, which colours?

_____

44. Do certain colours help to relax your child?

Yes No Sometimes

- If yes or sometimes, which colours? How does your child react to these colours that show they are relaxed?

_____

_____

45. Do certain colours excite your child?

Yes No Sometimes

- If yes or sometimes, which colours? How does your child react to these colours that show they are excited?

_____

_____

46. Do certain colours make your child feel sad?

Yes No Sometimes

- If yes or sometimes, which colours? How does your child react to these colours that show they are sad?

_____

_____

47. Do certain colours make your child feel happy?

Yes No Sometimes

- If yes or sometimes, which colours? How does your child react to these colours that show they are relaxed?

_____

_____

48. Does your child avoid certain colours?

Yes No Sometimes

- If yes or sometimes, which colours? How do they avoid them?

_____

_____

49. Are there any colour combinations that your child likes?

Yes No Sometimes

- If yes or sometimes, which colours? How do you know that your child likes these colour combinations?

_____

_____

50. Are there any colour combinations that your child dislikes?

Yes No Sometimes

- If yes or sometimes, which colour combinations? How do you know your child dislikes these colour combinations?

_____

_____

**Section 3:** Familial Details

51. Are there (or have there been) any incidences of colour blindness in your family?

Yes No

52. If yes, how was/is the person related to you?

_____

**Section 4:** Child's language

53. What languages are spoken to your child at home and at school?

_____

54. What is the usual spoken language of your child? Yes No

_____

55. Does your child speak more than one language? Yes No

If yes, what other languages do they speak?

_____

**Section 5:** Medical History (OPTIONAL)

56. Has your child been diagnosed with any behavioural problems or mental health difficulties?

Yes No

If yes – What other diagnosis

_____

_____

57. Does your child have any assistance to help their vision? (e.g. has your child had an eye test and been prescribed spectacles/glasses)

Yes No

58. If yes, what assistance have they been prescribed? (If known)

_____

59. Has your child ever had to wear an eye patch to help their vision?

Yes No

Thank you for your help and filling out the questionnaire.

If you have any further comments relating to your child's colour vision and behaviour or their vision and behaviour and how this relates to other sensory types (e.g. hearing, movement and smell) then please add them below.

_____

_____

_____

_____

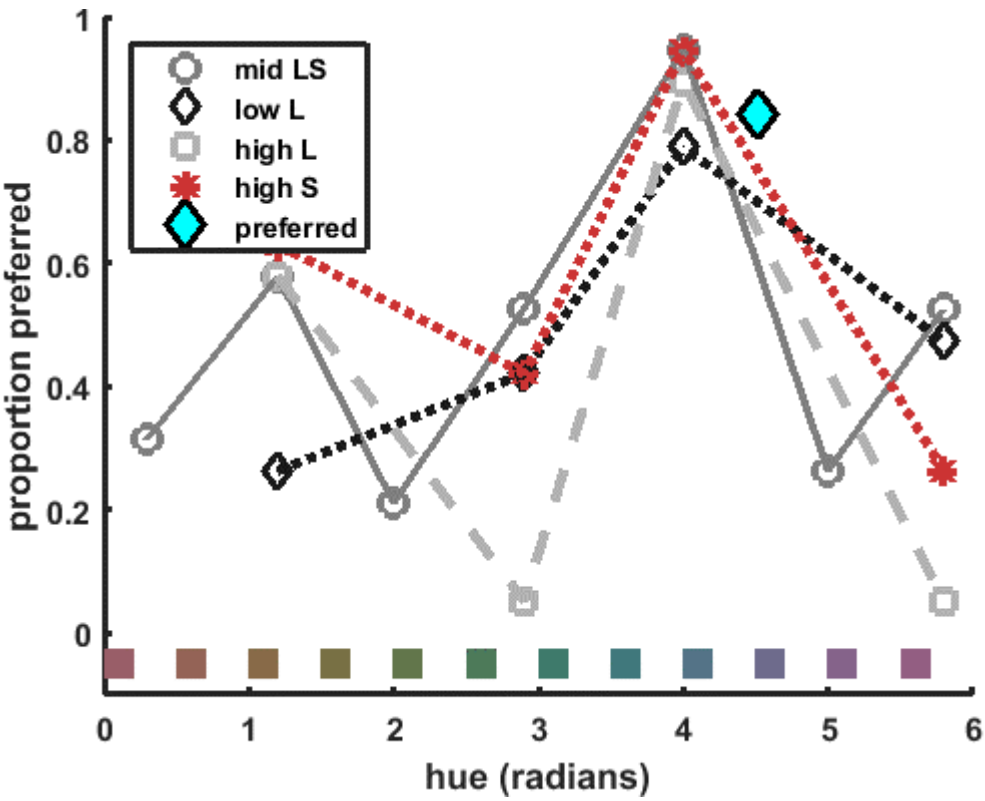
_____

_____

## Appendix 6 – Additional Case Studies

### Extreme Hue Preference

Williams Syndrome Case study 4 is a 14 year old male. His parents report that his favourite colour is blue. This extends to his behaviour being affected by this. His parents reporting that "everything has to be blue". This includes personalised objects such as phone and Nintendo DS covers, but also to other things such as his bedrooms colour (including bedding). The participant also stated, "I don't like anything green coloured". But conversely also liked a videogame character called Link who dresses in green. The colour preference results show a clear peak for bluish hues, regardless of manipulation but not necessarily a dislike of green (with the exception of greens of a high lightness). This participant shows another example of how an individual's responses to colour can be mediated by object associations.

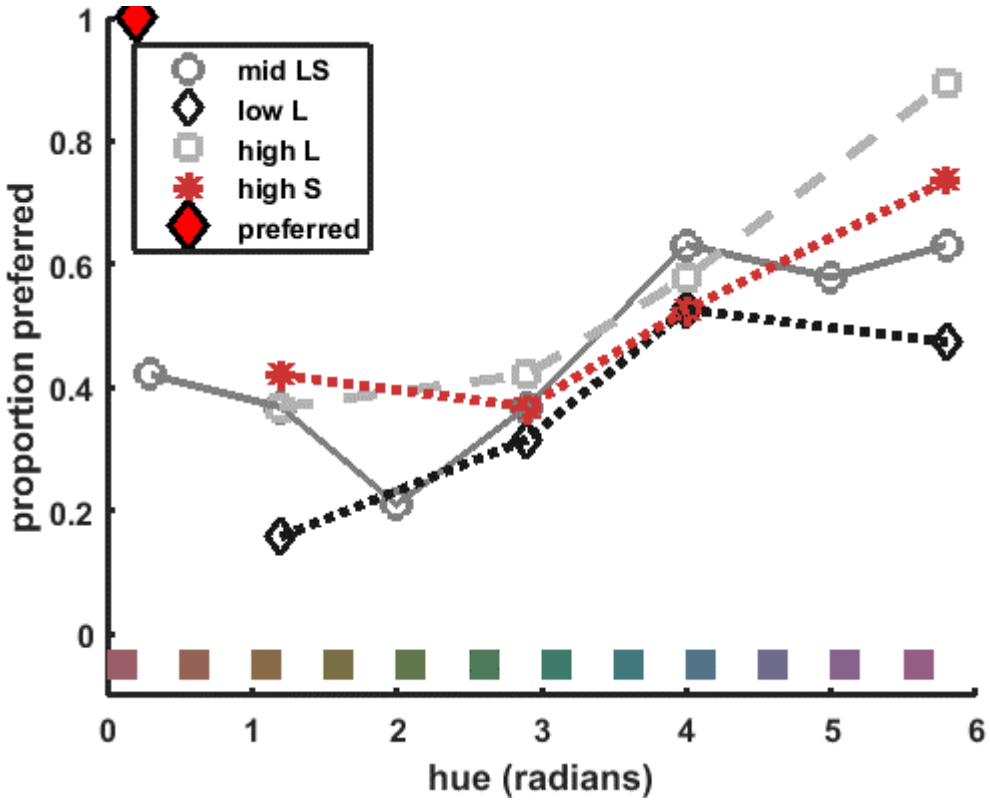



*Appendix Figure - 1 - Colour Preference curve for Williams Syndrome Case Study 4.* There is a clear preference for bluish hues, regardless or saturation or lightness level.

### Colour Insistent Behaviours

Williams Syndrome Case Study 5 is a 7 year old male. His parent reported that his favourite colour was red but that he did not have any colours that he didn't like. He was reported to show various different colour affected behaviours (clothing and food). For example where he insists on wearing a red jumper for school, and he will not wear the school's usual jumper which is blue. This results in difficulty in getting ready for school in the morning. When eating food, he will only eat applies if they

if they have some red on them. His preference curve largely dependent on hue. There is a relative peak for the colours of reddish hue for all variants. His favourite colour was the Primary Red from the colour checker chart and was chosen on all presentations. Thus being another example of colour preference results qualitatively reflecting colour affected behaviours.




*Appendix Figure - 2 - Colour Preference curve for Williams Syndrome Case Study 5.* There is a preference for reddish hues, regardless or saturation or lightness level, in line with the report colour affected behaviours reported

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