Development and validation of a parent-completed screening checklist for early movement abnormalities indicating a high risk of development of cerebral palsy

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Abstract

Introduction: Each year around 1800 UK children are diagnosed with Cerebral Palsy (CP). Of these, 40-50% are deemed 'low risk' at birth, relying on their parents and primary health care professionals (PHCPs) to identify concerning features and seek referral, ultimately leading to diagnosis. Reports suggest delays to diagnosis are occurring within primary care referral.

Aim: Identify the cause(s) of delays in the referral of infants with emerging motor difficulties to secondary care. Develop new tool(s) to reduce delays.

Methods: This study was carried out in three phases.

Phase 1: Online survey of parents and carers of children with CP about their earliest concerns and experiences of the referral and diagnosis process. Thematic analysis identified the earliest concerns and the Andersen Model of Total Patient delay categorised where delays are occurring. Phase 2: Scoping review of motor screening tools for infants aged 0-6 months. This identified how the contents of the tools were developed, if parents were included in their development, and how the content relates to early parental concerns. Phase 3: Iterative interviews with key stakeholders while developing a new tool for identifying concerning features.

Results: Phase 1: 255 respondents reported more concerns than those routinely reported in the literature. Delays related to symptom awareness, parental confidence, and watch and wait approaches. Phase 2: 42 tools identified. One tool included a parent of a child with CP in the development process. No tools identified all identified parental concerns. Phase 3: Two informational resources were developed: a short hard-copy resource to raise awareness in new parents, and a long online resource to provide further information.

Discussion: Reported delays suggest parents' experience difficulties in help-seeking, and parents and PHCPs lack symptom awareness. New resources aim to rectify this. Further research is needed to refine, validate, and identify the impact of new resources.

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Table of contents

List of Tables

Lists of figures

Chapter 1. Introduction

This thesis explores the early identification of infants with Cerebral Palsy (CP) within the community. It will investigate the reasons for delays in CP diagnosis, the difficulties parents have in reporting their concerns, and the limitations of currently available motor screening tools for use within primary care. It will also seek to develop information sheets for new parents around the earliest signs of CP and examine the need for more generalised information around typical infant development and behaviour.

In this chapter, I will outline what Cerebral Palsy is and how this umbrella term describes a range of impairment types, a range of impairment topographies, and the impact CP has on function. I will then discuss why diagnosis maybe delayed due to factors including current practices, the lack of specific CP biomarkers, the different outcomes that can occur after infant brain injuries, and the need to consider alternative diagnoses before a CP diagnosis can be given. Next, I will explain why early diagnosis is important, not just for the infants' outcome, but for the parents' mental health. I will then discuss alternative ways infants within the community could be identified, starting by explaining what currently happens, before identifying the opportunities for early motor screening that are currently being missed. Following this, I will then discuss how a screening tool could be used within primary care to identify infants' early emerging movement difficulties, such as CP, before discussing how parental concerns could be used as an alternative to the currently available screening tools.

1.1 What is Cerebral Palsy?

Cerebral palsy (CP) is an umbrella term for a group of permanent posture and movement disorders due to non-progressive damage to the developing brain, often accompanied by associated impairments, such as visual, learning, speech, and intellectual impairments, epilepsy, and secondary musculoskeletal problems (Rosenbaum *et al.*, 2007). CP is the most common physical disability of childhood. It has worldwide prevalence of 2.11 per 1000 live births (Oskoui *et al.*, 2013) and is estimated to affect approximately 17 million people globally. Across high income countries, such as the UK, the prevalence of CP is 1.6 per 1000 live births (McIntyre *et al.*, 2022). Within England and Wales, it was estimated that 22,100 children aged 3-15 years had a diagnosis of CP in the year 2020 (Glinianaia *et al.*, 2017). CP is

a lifelong condition with implications for daily living, quality of life and self-esteem (Russo *et al.*, 2008). Due to the heterogeneous nature of CP, it is categorised by the type of motor impairment, the affected limb topography, and by the level of function the individual has.

1.1.1 Impairment type

Individuals with cerebral palsy may have Spastic, Dyskinetic and/or Ataxic impairment types. Spastic CP is the most common form of CP, representing around 75-86% of CP cases (Johnson, 2002; Westbom, Hagglund and Nordmark, 2007). The Surveillance of Cerebral Palsy in Europe (SCPE) collaborative group defines Spasticity as 'increased tone and pathological reflexes' (Cans *et al.*, 2007, p. 36). Increased tone in the context of Spasticity refers to an increased resistance in movement that is velocity dependent. A Spastic 'catch' (a sudden increase in muscle activity in response to a fast, passive movement) is felt at some point after starting the movement. 'Pathological reflexes' refers to either hyper-reflexia, and changes in pyramidal reflexes, such as the the Babinki sign[1] in their affected lower limbs or Hoffmans[2] in the affected upper limbs. Spasticity in the lower limbs can lead to '(1) internal rotation of the hip; (2) hip adduction; and (3) equinus foot, the combination resulting in a 'scissored' position' (SCPE, 2002; Cans *et al.*, 2007, p.36). Spasticity in the upper limbs can result in flexion in the elbow, wrist, and fingers.

Dyskinetic CP is reported to be the most common form of CP within term-born, appropriate-size-for gestational age children, who suffered adverse perinatal events (Himmelmann *et al.*, 2009). The reported prevalence of Dyskinetic CP differs between CP registers, with between 3-15% of CP being recorded as Dyskinetic (Johnson, 2002; Himmelmann *et al.*, 2005; Parkes *et al.*, 2005; Serdaroğlu *et al.*, 2006; Andersen *et al.*, 2008). Dyskinetic CP is characterised by 'involuntary, uncontrolled, recurring, and occasionally stereotyped movements. The primitive reflex patterns predominate, and the muscle tone is varying' (Cans *et al.*, 2007, p.36) (SCPE, 2002). Individuals with Dyskinetic CP perform involuntary, repetitive movements. Dyskinetic CP is further defined by two subcategories, dystonic and choreo-athetotic. The dystonic subgroup refers to those who demonstrate abnormal postures, due

[1] The Babinski sign is a reflex that occurs when the sole of the foot is stroked firmly with a blunt object. Children up to 2 years old and those with changes in their pyramidal reflexes will respond by lifting their big toe and fanning their other toes out.

[2] The Hoffman sign is a reflex that occurs when the middle finger nail of an individual is flicked by another person. Individuals who have changes in their pyramidal reflexes with react by flexing their index finger and thumb quickly and involuntarily right after their middle finger is flicked.

to sustained muscle contractions and hypertonia (abnormally high tone). In contrast, the choreo-athetotic subgroup are defined by their 'rapid, involuntary, jerky, often fragmented movements' (Cans *et al.*, 2007, p.36) and their 'slower, constantly changing, writhing, or contorting movements' (Cans *et al.*, 2007, p.36). However, individuals may demonstrate both dystonic and choreo-athetotic characteristics.

Ataxic CP is reported to make up 4-6% of all CP cases (Johnson, 2002; Serdaroğlu *et al.*, 2006; Andersen *et al.*, 2008). Ataxic CP is defined by the 'loss of orderly muscular coordination, so that movements are performed with abnormal force, rhythm, and accuracy' (Cans *et al.*, 2007, p.36). For example, an individual with Ataxic CP may go to point at an object and over or under shoot their goal.

However, the different types of impairment are not stand-alone diagnoses. Overlap between impairment types can occur within individuals, for example Spasticity is reported to occur in around 69% of Dyskinetic CP cases (Westbom, Hagglund and Nordmark, 2007). Overlapping impairments can present themselves as affecting the same limb or different limbs, such as Spasticity affecting the left arm and dyskinesia affecting the right arm.

1.1.2 Topographical limb impairment and function

Classifying CP by the topography of the limbs affected is also key, as the limbs impaired between individuals with CP also vary. CP can affect all four limbs, trunk and neck (quadriplegia) or may affect just one limb (monoplegia). Individuals with both legs impaired, but no arm involvement are categorised as having diplegia. Additionally, individuals may only be affected on one side of their body. An individual affected only in their left arm and leg, or their right arm and leg, is categorised as having hemiplegia. Individuals with three limb or four limb CP are referred to as having tetraplegic and quadriplegic CP respectively. The use of topographical descriptions is useful for identifying the potential functional difficulties the individual may have.

However, despite describing impairment type and the limb topography, the amount of function each individual with CP has also varies and thus must also be described. The gold standard for measuring functional impairment is the Gross Motor Function Classification System (GMFCS). First developed by Palisano *et al.* (1997), the latest version of the GMFCS categorises motor function in children aged <2 years to 18 years into 5 levels based on age

appropriate gross motor tasks, such as manipulating objects, sitting, crawling and walking (Palisano *et al.*, 2008; Rackauskaite *et al.*, 2012; Gudmundsson and Nordmark, 2013). Level 1 describes individuals with the least amount of impairment, while level 5 describes individuals with the highest amount of impairment. Examples of these levels are shown in Figure 1.

The importance of using functional impairment, topography and impairment type to described an individual's CP is highlighted in Gorter *et al.* (2004). Gorter *et al.* (2004) demonstrated that topographic involvement and type of impairment both significantly ($p<.001$) influenced functional level on the GMFCS in 657 children aged 1-13 years. However, within each topographical group, and within each impairment type, children scored across the GMFCS spectrum. Thus, impairment type, topography and functional impairment are all required to accurately describe an individual's CP.

1.1.3 Associated impairments

As well as motor impairments, individuals with CP often have additional associated impairments in cognition, communication, behaviour, and sensation, and may suffer from epilepsy and secondary musculoskeletal problems. This includes 3 in 4 being in pain, 1 in 2 having intellectual disability, 1 in 4 being unable to talk, 1 in 4 having epilepsy, 1 in 4 having behaviour disorder, and 1 in 5 having a sleep disorder (Novak *et al.*, 2012). The presence of associated impairments has been shown to be positively correlated to the severity of the motor impairments (Beckung and Hagberg, 2002; Himmelmann *et al.*, 2006; Novak *et al.*, 2012; Horber *et al.*, 2020). Studies have described these associated impairments as having the potential to be more disabling than the motor impairments associated with CP (Beckung and Hagberg, 2002; Vidart d'Egurbide Bagazgoïtia *et al.*, 2021) (2-4). Due to these reasons, screening for associated non-motor impairments is recommended as part of CP assessment.

Overall, CP is an umbrella term for a group of movement disorders caused by damage to the developing brain, often accompanied with impairments in non-motor domains. To define an individual's CP diagnosis, their motor impairment type(s), their topographical limb impairments, and their level of motor function are needed. Although an individual's CP diagnosis can be easily defined, the route to diagnosis is not so clear.

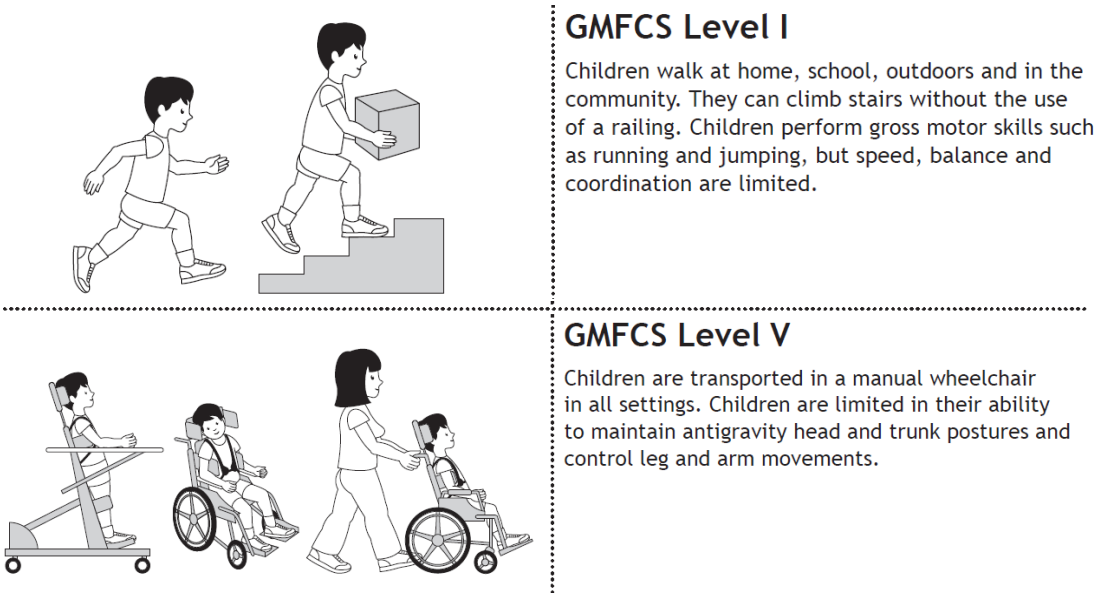


Figure 1 Examples of the Gross Motor Function Classification Scale levels 1 and 5 for children aged between 6 and 12 years. Images and descriptions taken from the 'GMFCS E & R between 6th and 12th birthday: Descriptors and illustrations' form on www.canchild.ca. Descriptions by Palisano et al. (1997). Illustrations by Reid, Willoughby, Harvey, and Graham, The Royal Children's Hospital Melbourne. GMFCS: Gross Motor Function Classification Scale.

1.2 Delays in Cerebral Palsy Diagnosis

1.2.1 How is Cerebral Palsy diagnosed currently?

Diagnosis of CP is made on clinical grounds through history-taking, examination and often also informed by neuroimaging findings. Outcomes after early brain injury are variable, with a traditional approach having been to watch and wait, and then determine whether a child fits a clinical diagnosis of cerebral palsy once established clinical signs of this are present (McIntyre *et al.*, 2011; Basu, 2014). For example, despite perinatal stroke (a stroke occurring within or before the first 28 days of life) being a major cause of CP, 50% or fewer of infants who suffer a perinatal stroke go on to develop CP (Golomb *et al.*, 2008). Similarly, not all infants who go on to receive a diagnosis of CP demonstrate early signs or risk factors for CP.

Registries across the world currently indicate a mean age at diagnosis of 19 months for CP (McIntyre, *et al*. 2011), consistent with empirical clinical UK experience. Multiple calls for earlier CP diagnosis have been published and earlier diagnosis has been demonstrated to be possible (McIntyre *et al.*, 2011; Novak *et al.*, 2017b; te Velde *et al.*, 2019). For example, the Australian Cerebral Palsy Register Group (2018) reports that 21% of children on their register, with prenatally or perinatally acquired CP, were diagnosed within the first 6 months

of life, while 50% of children on the register were diagnosed within the first year of life. The rest of this section will look at the factors that result in delays to diagnosis.

1.2.2 Identifiable biomarkers

Currently there are no clinically used biomarkers that accurately predict CP; however it is possible to identify infants at risk of developing CP through clinical risk factors (McIntyre *et al*., 2011). Damage to the brain, which results in CP, can occur before birth (congenital CP) or shortly after birth (acquired CP). The causes of congenital CP are largely unknown; however, the Centers for Disease Control and Prevention (CDC) have defined a list of risk factors to help identify infants at high risk of congenital CP. The CDC congenital CP risk factors include: low birthweight (<5 ½ pounds or 2500g at birth), premature birth (born at <37 weeks gestation), multiple births, use of assisted reproductive technology infertility treatments, infections during pregnancy, infant jaundice, birth complications (such as placenta detachment and uterine rupture) and maternal medical conditions (such as thyroid problems or seizures) (CDC, 2017, National Institute of Neurological Disorders and Stroke (NIH), 2018). In contrast, acquired CP is often caused by infections of the brain, injury to the brain, and problems with blood flow to the brain (such as stroke) during infancy, with preterm and/or low birthweight infants being at greater risk for developing CP (NIH, 2018, CDC, 2017).

However, the risk factors listed above are not specific for CP. Around 40-50% of infants with CP do not demonstrate the risk factors described at around the time of birth, while around 18% of individuals with CP have no clear etiological explanation for their CP (Shevell, Majnemer and Morin, 2003). Infants who do not show risk factors for CP around the time of birth are naturally categorised as at 'low-risk' for having CP. This low-risk category also includes infants who were born moderate-late pre-term (after 30^{+0} weeks gestation) but were not identified to have: a brain lesion associated with developmental problems or disorders, demonstrated through neuroimaging; a grade 2 or 3 hypoxic ischemic encephalopathy; bacterial meningitis; or herpes simplex encephalitis, within the neonatal period (NICE, 2017b). These low-risk infants return home with their families, only for the signs of CP to emerge later. These infants therefore rely on their families, caregivers, and Primary Health Care Professionals (PHCPs) to identify and report any emerging signs for the referral process for diagnosis and therapy to begin.

In contrast, infants identified as 'high risk' for CP are provided with enhanced developmental support and surveillance for up until 2 years corrected age by a multidisciplinary team (NICE, 2017b). As such, these infants receive 3 'follow up visits' with a focus on infant motor development at 3-5 months, before 12 months, and at 2 years corrected age. During these visits parents are encouraged to raise any concerns they may have, infants are checked for signs and symptoms of developmental disorders, including CP, and if a problem is suspected or identified, further investigation is carried out as well as referring the infant to the appropriate pathway.

Research has shown that infants referred for diagnosis from primary care are referred significantly later than their counterparts who were followed up in secondary and tertiary care clinics (Hubermann *et al.*, 2015; Boychuck *et al.*, 2020). A retrospective case-notes review by Boychuck *et al.* (2020) places the age gap at the time of diagnosis between those referred from primary (median age= 14 months) and secondary (median age=3.5) care at 10.5 months.

Biomarkers that could potentially be introduced into clinical practice in the future include genetic and epigenetic markers. Familial clustering of CP has been reported in Australia, Sweden, and Norway, with the relative risk of CP between full siblings reported between 4.8% and 9.2% (Hemminki *et al.*, 2007; O'Callaghan *et al.*, 2011; Tollånes *et al.*, 2014). Mounting evidence suggests that 10-30% of individuals with CP have some form of genetic component, however, genetic sequencing has also demonstrated high heterogeneity in the genes that could be responsible for CP onset (Moreno-De-Luca, Ledbetter and Martin, 2012; McMichael *et al.*, 2015; Oskoui *et al.*, 2015; Fahey *et al.*, 2017; Zarrei *et al.*, 2018; Rosello *et al.*, 2021; Savasana *et al.*, 2021; Chopra *et al.*, 2022). Until a consensus is reached as to which genes are responsible for CP, or for CP subtypes, genetic testing is unlikely to become a part of CP screening.

Epigenetics refers to the process of managing gene expression without altering the DNA sequence. Several studies have investigated epigenic changes in individuals with CP. Jiao *et al.* (2017) and Mohandas *et al.* (2018) investigated epigenetic differences in monozygotic twins in which only 1 twin was diagnosed with CP. Both studies identified differences in the epigenetic expression of genes within the monozygotic twin pairs from blood samples. In terms of the level of epigenetic differences, Jiao *et al.* (2017) demonstrated that the twin

pairs still had significantly similar epigenetic patterns across the whole genome (Pearson's r's=around 0.98). Furthermore, machine learning based approaches have demonstrated epigenetic screening to have good sensitivity (95%) and specificity (94.4%) for CP in new-borns (n=44, n=23 with CP) (Bahado-Singh *et al.*, 2019), and high sensitivity (100%) but low specificity (40%) in adolescents (n=43, n=22 with CP) (Crowgey *et al.*, 2018). However, as Romero *et al.* (2021) highlights, epigenetic research in CP is still at an early stage and the effects of different factors, such as the site of sample collection, have on result outcomes still require research.

1.2.3 Different outcomes after brain damage

Neuroimaging is classed as a gold-standard tool in predicting CP (Ashwal *et al.*, 2004; Bosanquet *et al.*, 2013; Novak *et al.*, 2017b). Magnetic resonance imaging and cranial ultrasound scans have both been demonstrated to have high sensitivity and specificity for identifying neurological abnormalities predictive of CP. Predictive abnormalities include white matter injuries (such as cystic periventricular leukomalacia), cortical and deep grey matter lesions (such as basal ganglia lesions), and atypical brain development (such as polymicrogyria). Studies have demonstrated that often the type and timing of the lesion are predictive of the affected limb distribution and the severity of the CP (Krägeloh-Mann and Horber, 2007; Reid *et al.*, 2015; Himmelmann *et al.*, 2021).

However neuroimaging only gives some idea as to whether the infant as at risk of having CP. Around 11-29% of children diagnosed with CP demonstrate 'normal' and/or non-specific neuroimaging results on MRI, not indicative of CP (Bax, Tydeman and Flodmark, 2006; Benini, Dagenais and Shevell, 2013). These infants cross all clinical CP subtypes and range across the severity scale. Furthermore, infants with similar brain lesions do not always develop CP or the same form of CP (Pierrat *et al.*, 2001; Golomb *et al.*, 2008). Therefore, even after neuroimaging clinicians must wait to determine if the infant has emerging signs of CP.

1.2.4 Other conditions may be mistaken for Cerebral palsy

The symptoms of CP are non-specific and many conditions present similar symptoms and signs. Examples of similarly presenting conditions include Dopa responsive dystonia, a genetic disorder that causes involuntary movements that improve when the patient takes L-Dopa medication (Appleton and Gupta, 2019). It is important to ensure the correct diagnosis

is given, especially since some similarly presenting conditions can be treated, and the infant's family can be given more accurate information about their infants' prognosis. Additionally with the advancement of genetic and epigenetic research into CP, parents of infants with genetic conditions that cause CP or cause presentations similar to CP (also known as CP mimics) can be given genetic counselling.

Delays in diagnosing CP can come from trying to ensure the correct diagnosis is given. CP has no clear specific biomarkers, and even when a brain lesion is detected that does not mean the infant will later present with the CP phenotype. The signs of CP overlap with other motor disorders which emerge as the brain develops, and so these other conditions must be considered before the CP diagnosis can be given. However, these delays prevent early interventions from occurring that could have beneficial impacts on the infants' brain development and the parents' mental health.

1.3 Why is early diagnosis important?

1.3.1 Brain development

The development of the brain is a long and intricate process. During the fifth week to the fifth month of gestation a process called cellular proliferation occurs, in which neural stem cells located within the walls of the vesicles, known as the ventricular zone, proliferate at an exponential rate (Bear, Connors and Paradiso, 2007). The rapid proliferation of neural stem cells results in the production of daughter cells (Bystron, Blakemore and Rakic, 2008). These newly formed daughter cells either remain within the ventricular zone to continue replicating, or they migrate to form the neocortex. The migrating cells, called neuroblasts, form structures from the inside out, with the subplate being formed first and layer I (the outermost layer) being formed last. The process of migration is completed during the third trimester (Bystron, Blakemore and Rakic, 2008). Once having reached their positions in the neocortex, the neuroblasts undergo differentiation, each forming an axon and dendrites. This is followed by synaptogenesis, in which the neurons form synapses with other neurons. However, around half of these neurons and synapses are later eliminated from the brain either due to cell death or activity-dependent withdrawal. Cell death occurs due to competition between innervating neurons for synapses with their target neuron (Blanquie *et al.*, 2017). Similarly, activity dependent withdrawal is where the least active neuron of

competing neurons is withdrawn, resulting in only the most active synapses remaining (Blanquie *et al.*, 2017).

However, when an early brain lesion occurs, as with CP, the topography of the brain is disturbed. Brain lesions cause damage to the neurons. Depending on the level of damage the neurons experience they may remain or may undergo immediate cell death (Berger, Garnier and Jensen, 2002; Truttmann, Ginet and Puyal, 2020). However, after damage occurs to a neuron, the neuron's activity level is typically reduced. If the neurons become less active they may be eliminated, resulting in an atypical topography (Eyre *et al.*, 2001). As the brain matures the body parts that correspond to the atypical topography begin to show reduced variation in their movements.

The reduced variation in these infants' movements can be explained through Neuronal Group Selection Theory (NGST). Developed by Gerald Edelman, NGST explains that behavioural exploration of all motor possibilities creates afferent feedback to the nervous system (Hadders-Algra, 2000a; Hadders-Algra, 2010). The feedback allows for epigenetic changes to shape the nervous system, as seen during activity dependent withdrawal. NGST splits development into two phases, the primary and the secondary. The primary phase is characterised by variations in movement that produce afferent information that do not feedback to adapt behaviour to environmental constraints. As such, infants in the primary phase will continue to produce varied movements. In the secondary phase, the nervous system uses the afferent feedback to select the motor behaviours that best fit the situation. This results in the nervous system selecting the most effective motor patterns, reducing the variation in movement with practice. Although the secondary phase always follows the primary phase, each phase does not occur synchronously across the brain.

When an early brain lesion occurs there are two main implications within NGST. The first is that the motor repertoire of the infant is reduced, resulting in more stereotypical movement patterns (Hadders-Algra, 2000b; Hadders-Algra, 2010). The loss of neurons after a brain lesion reduces the neural pathways available to the infant. As such the reduced number of neural pathways reduces the variations in neural pathways the infant may use, and therefore reduces the variation in their motor repertoire. Reduced variation in motor movements has been recorded in infants who have suffered brain lesions. In particular, movement variation is a key factor for determining if an infant's movements are normal or

abnormal on Prechtl's General Movements Assessment (GMs), a gold standard tool for identifying infants with CP (Einspieler *et al.*, 2004). The second implication occurs during the secondary phase. As the infants' motor repertoire is reduced, their ability to select the most effective motor strategy is also reduced (Hadders-Algra, 2000b). As a result it is likely that these infants will have to try more movement variations to determine the best strategy to use, resulting in their development becoming delayed, a known feature of CP.

Animal research has demonstrated that early targeted therapy can help the nervous system to retain a more typical structure and improve the functional ability of the affected limbs. Animal research has focused on promoting affected limb use in kitten models after induced unilateral perinatal stroke (Martin *et al*., 2011, Martin *et al*., 2007, Friel *et al*., 2012, Salimi *et al*., 2008). Although these studies have consistently demonstrated 'therapy' to improve function outcomes, Friel *et al.* (2012) also assessed the effect of age at the time of therapy on the outcome. After inducing unilateral strokes between Postnatal Weeks (PW) 5 and 7, Friel *et al*. (2012) split the kittens into early (PW 8-13) and late (PW 20-24) training groups. Both training groups received the same reach training and constraint of the unaffected limb. The constraint, regardless of age, restored the corticospinal tract connections and the motor map of the affected area. However, the early constraint group also had an increase in the number of contralateral spinal interneurons relative to ipsilateral, resulting in a more normal wiring pattern, and a reduction in the control impairments of the affected limb, demonstrating 'therapy' to be more effective when carried out early.

Early translational studies have shown promising results across CP topologies. A recent systematic review by Damiano and Longo (2021) highlighted 6 randomised control trials of early motor interventions for infants aged 0-3 years with or at risk of CP. Three of the trials had significant positive effects on the infants' outcomes, all of which promoted voluntary movement with targeted goals. Eliasson *et al.* (2018) promoted increased movement of the infants' more-affected arm through Baby Constraint Induced Movement Therapy (CIMT) in infants with unilateral impairments. During therapy the infants were required to wear a mitt on their less-affected hand to prevent the infant from using it for grasping. The infants were then presented with toys to promote grasping and exploration with the more-affected hand. Chamudot *et al.* (2018) also used CIMT and compared it with promoting bimanual movements in infants with unilateral impairments. Infants in the CIMT group were also

required to wear a custom mitt on their less-affected hand during therapy and were presented with toys that promoted unilateral grasping and play with the infants' more-affected hand. In contrast the bimanual therapy group were not required to wear mitts and were presented with toys that promoted use of both hands at the same time. Chamudot *et al.* (2018) found both therapies to equally improve the infants' hand and gross motor function. In contrast to Chamudot *et al.* (2018) and Eliasson *et al.* (2018), Morgan *et al.* (2014) included infants with unilateral and bilateral impairments. Their GAME therapy promoted increased movement through collaborative goal setting between the parents and therapists based on the infant's abilities (Morgan *et al.*, 2014; Morgan *et al.*, 2016). Furthermore, part of the therapist role was to teach the parents strategies they could use at home which would promote the infant reaching their goal. Although not identified by Damiano and Longo (2021), Holmström *et al.* (2019) also found significant improvements in motor ability when infants with bilateral and unilateral impairments were presented with the Small step intervention. Small step consists of 3 components: mobility, hand use, and communication. Although the components were presented in a rigid structure over 5 weeks, the goals of each component were set through collaboration between the parents and therapists. However, it should be noted that all three of these studies used small numbers of participants.

Unlike the above studies, the three studies identified by Damiano and Longo (2021) as not having significant positive effects did not provide specific training goals. Stark *et al.* (2016), trialled whole body vibration stimulation alongside standard care. Mattern-Baxter *et al.* (2020) trialled low and high intensity treadmill training in infants 14-32 months. Although treadmill training only targeted walking ability, the intervention remained the same for each infant, with no modifications occurring based on the infant's ability. Hielkema *et al.* (2020) trialled a family centred programme named Coping with and caring for infants with special needs (COPCA). COPCA has two components, a family and education component, and a neurodevelopment component based on NGST. Unlike the education components of GAME and Small step, families receiving COPCA were observed and only given suggestions on how to interact with their infant rather than given instructions based on collaborative decisions (Akhbari Ziegler, Dirks and Hadders-Algra, 2019). The COPCA study took this approach in the idea that it would enhance family coping strategies and would allow families autonomy in exploring possibilities to challenge their infant to self-produce motor behaviour. However, it

is likely that education component was unsuccessful due to it's hands-off approach, rather than the hands-on approach used in GAME and Small step.

Overall, early diagnosis can allow for the implementation of early therapy that targets specific goals. In turn, early therapy can have significant impacts on infant outcomes, including reducing the severity of their CP. However, early diagnosis has also been demonstrated to have a positive impact on parental mental health, which in turn can influence infant development.

1.3.2 Parental mental health.

Following their child's diagnosis of CP parents often report grieving the life their child could have had. Although the process varies between individuals, parents report feeling a mix of emotions, such as sadness, for the lost opportunities they were expecting to have with their infant, and relief as their feelings of uncertainty were lifted, that are intensified just after receiving the diagnosis and at each of the infant's major life events (Whittingham *et al.*, 2013). These experiences align with chronic sorrow theory, described as 'the periodic recurrence of grief-related emotions associated with an ongoing disparity between desired and current reality due to a loss experience' (Eakes, Burke, & Hainsworth., 1998). The timing of the diagnosis can also impact on parents' outcomes. Delayed diagnoses of CP, especially when the parents have raised concerns, can cause parents further dissatisfaction with how the diagnosis is made, and makes some parents feel angry, shocked, and helpless about how their concerns had been handled (Baird, McConachie and Scrutton, 2000; Huang, Kellett and St John, 2010; Williams *et al.*, 2021). Furthermore, these parents are at higher risk for poor psychological outcomes compared to parents of typically developing infants, such as depression and anxiety (Pinquart, 2018; Scherer, Verhey and Kuper, 2019; Barreto *et al.*, 2020).

Parental mental health can influence the interactions between the parent and infant with potential impacts on the infant's development over time. For example maternal anxiety can decrease maternal sensitivity and engagement towards their infant (Riva Crugnola *et al.*, 2016). As the mutual responsiveness between the parent and infant influences the infant's cognitive and motor development, maternal anxiety can reduce the mothers responsiveness to their infant which in turn reduces the infant's cognitive and motor development speed (Landry, Smith and Swank, 2002; White-Traut *et al.*, 2018).

However, early psychological interventions that provide support to parents can improve outcomes. Dickinson *et al.* (2020) systematically reviewed the efficacy of psychological interventions compared to standard care in parents whose infants were diagnosed with or were at risk of a neurodevelopmental disability. The meta-analysis demonstrated psychological intervention within the first year of the infant's life to have significant short and long term impacts on parental depression, anxiety, stress, and trauma. Irwin, Jesmont and Basu (2019) looked more specifically at interventions aimed at parents of children with CP. Despite few studies in this area, and heterogeneity in their results, the meta-analyses demonstrated psychological intervention to significantly improve parental mental health.

This chapter has shown that early diagnosis and early intervention are key to giving infants with CP the best possible outcomes. It has shown that targeted, goal directed early motor intervention can significantly improve motor outcomes of infants by utilising the plasticity of the developing brain. Finally it has shown that psychological therapy for parents can not only improve parental mental wellbeing but their interactions with their infants, and subsequently their infant's development. I will now outline the current screening programs that seek to maximise early diagnosis and early intervention. This next section will focus purely on the practices within the UK; this is because there are differences in how screening is carried out across the world (Kim, 2022). For example, in the USA, developmental surveillance is recommended to occur every 2-3 months across the first 18 months of life (Centers for Disease Control and Prevention (CDC)Council on Children With Disabilities *et al.*, 2006; 2021). In Estonia infants receive monthly monitoring (National Center on Education and the Economy, 2021), whereas in the UK, the first standardised motor development appointment occurs at 9-12 months (NHS, 2020b).

1.4 How can screening be used to minimise delays in identification of Cerebral Palsy

1.4.1 Current screening programs

In the UK there are currently two types of screening carried out; clinical follow up and standard developmental screening. Clinical follow up is only for infants identified as having major risk factors for future conditions. More specifically for infants with major CP risk factors, the NICE (2017) guidelines recommends these infants undergo regular follow up between birth and 2 years of age. However, the content of follow up appointments is not

standardised, with guidelines suggesting that the appointments be tailored to the needs of the infant.

In contrast to clinical follow up, all UK infants undergo standard developmental screening. Standard developmental screening is carried out by Health Visitors or a member of their team either in the parents' home or in a GP surgery, baby clinic or children's centre. Although appointments occur from birth until the infant is 2 years old, there are only 5 standardised postnatal visits that aim to provide parents with additional information about their infant and to carry out diagnostic tests: 5-8 days, 10-14 days, 6-8 weeks, 9-12 months and 2-2½ years of age (NHS, 2020b). The first standardised appointment with a developmental assessment focus is at 9-12 months. The 9-12 month appointment and the 2-2½ year appointment both typically use the Ages and Stages Questionnaire 3 (ASQ3)(Squires and Bricker, 2009) to assess infant development.

The current developmental screening program has two main issues. The first is of missed opportunities for earlier screening in standard developmental screening, and the second is for the types of screening tools being used.

1.4.2 Missed opportunity

The developmental screening program misses the age 3-4 month period in which parents of infants with emerging CP begin to develop concerns (NHS England, 2016). Although some Health Visitor Services do carry out a 3-4 month visit, such as The Newcastle upon Tyne Hospitals NHS Foundation Trust (2018), Bridgewater Community Healthcare NHS Foundation Trust (2017) and Harrogate and District NHS Foundation Trust (2018), this additional visit is not universal across the UK. So an assessment, to identify developmental issues, could be introduced at 3-4 months as part of the routine vaccination appointments infants already receive.

1.4.3 Screening tools

The ASQ3, is currently used in standard development screening. The ASQ relies on the development of typical motor milestones, which are known to vary within and between populations. For example, the ability of infants to lift their head whilst in the prone position typically emerges between birth and 2½ months (Adolph, Karasik and Tamis-LeMonda, 2010). Thus, assessments using typical motor milestones require more than one assessment

to determine if the infant is delayed or within normal variation. As such, given the current screening programme, assessment at 6-8 weeks is too early for many milestones. So referral could only occur after 9-12 months. Alternatively, the ASQ could be used at the 3-4 month age mark, however a second appointment would still be needed for a timely referral to occur. Furthermore, the open-ended questions on the ASQ would not identify all CP limb distributions. For example the ASQ asks 'Does your baby use both hands and both legs equally well? If no, explain:' Questions such as the one described, would only be indicative in cases of developing unilateral CP, in which only one side of the body is affected. CP subtype prevalence studies demonstrate unilateral CP (including monoplegia) to account for only 29.2-32% of the total CP population (Johnson, 2002a, Yeargin-Allsopp *et al*., 2008, Mongan *et al.,* 2006).

Another pre-existing assessment or questionnaire for ubiquitous screening could be introduced with a potential to increase rates of early referral. In 2018, Kjolbye *et al*. reviewed the existing validated motor function tests that were suitable for use by a GP (taking <15 minutes to complete). They described 5 tools (Alberta Infant Motor Scale [AIMS], Harris Infant Neuromotor Assessment [HINT], ASQ3, Brigance Infant and Toddler Screen [BITS] and the Early Motor Questionnaire [EMQ]). Kjolbye *et al*. (2018) considered the AIMS, HINT and BITS as too time consuming or not economically viable, thus finally recommending only two assessments; the ASQ3 and the EMQ. However, the EMQ has similar issues to the ASQ3. Its scale was developed from typically developing milestones (Libertus and Landa, 2013, Squires *et al*., 2009, Knobloch *et al*., 1980) which leaves it vulnerable to instability between populations (van Heerden *et al*., 2017, Alvik and Grøholt, 2011). The EMQ open-ended questions also lack precision. For example, the EMQ asks; 'When sitting on your lap or in a highchair playing with toys, do you notice your child is able to successfully hold on to a small object such as a ring or stick?' An infant developing CP is likely to have fisted hands, thus if a parent were to place a toy in the infant's hand, it is likely the infant would 'successfully' hold on to the toy due to not being able to let go and therefore would present as typical on that question. However, it is also likely that the child would not be able to grasp or let go of the toy unaided. Alternatively, the child may be able to successfully hold on to toys with their less affected hand. Thus, by only asking about 'successful' holding, the EMQ misses other fine motor abilities indicative of impaired motor abilities. It is likely that parents and HCPs would answer this question with more flexibility in

their interpretation. However, this flexibility may be absorbed by the scoring system. The EMQ uses a 5-point Likert scale, beginning at -2 ('sure that child does NOT show behaviour') to 2 ('Sure the child shows this behaviour and remember a particular instance'). Points -1 and 1 describe the infant as probably or probably not showing the described behaviour, while point 0 is 'unsure whether child could do this or not'. Parents witnessing their infant 'successfully' holding a toy due to being unable to un-fist their own hands may use one of these less definite categories. Alternatively, parents may use the 'Comments and concerns' section at the end of the EMQ to describe their infant's ability to hold objects. Therefore, the ability of the EMQ to identify infants with emerging CP may come down to the parent's interpretation of the question and response scale.

An alternative tool that could be used is Prechtl's General Movements Assessment (GMs). GMs assesses the infant's early movements for subtle movement abnormalities using observation (Einspieler *et al*., 2016, Ferrari *et al*., 2004, van Iersel *et al*., 2016). These abnormalities are often detected in infants who subsequently develop CP (Karch *et al*., 2012, Chen *et al*., 2013, Chen *et al*., 2015, Guzzetta *et al*., 2010, Einspieler and Prechtl, 2005). GMs is the gold standard screening tool used to follow up high-risk infants at 3-4 months of age with excellent sensitivity (98%) and specificity (94%) and is used in some UK clinical follow up programmes (Morgan *et al*., 2016a). However, studies investigating the effectiveness of GM in the general population have demonstrated GMs to have low sensitivity and predictive power for CP (Bouwstra *et al.*, 2010; Bennema *et al.*, 2016), identifying as few as 12 CP cases out of 100 CP cases (Bouwstra *et al.*, 2010; Bennema *et al.*, 2016). Therefore, screening the general population in primary care with the GMs would be inefficient.

To overcome the limitations by the recommended tools, a new tool could be developed. Although the onset of CP predictive signs varies between individuals, signs, such as neonatal seizures, can appear within 24 hours after birth (Garfinkle and Shevell, 2011). As such, the tool should allow parents and health care professionals to raise concerns about infant development at any of the infant's appointments, not just the 9-12 month appointment. However, apart from the use of the ASQ3 from 9 months, there is no other recommended screening tool for primary health care professionals to use in the UK. A new tool that is not based on motor milestones and incorporates the opinions of key stakeholders could be developed for use in the general population.

1.5 Parent concerns

An alternative approach, which could support early identification of motor difficulties, is by developing an assessment based on early parental concerns that could be an alternative to and/or complementary to the currently available assessments. Parents are usually the first to notice early symptoms of developmental difficulties in their infant and have been described as 'lay epidemiologists' by Arksey (1994) due to their profound knowledge of their own child. From focus groups carried out by the Newcastle research team I have been part of, parents reported noticing symptoms from as early as three weeks;

> (At) three weeks, you couldn't dry under his arm properly because you couldn't get his arm up. Six months on we got a diagnosis. (Basu *et al.*, 2015)

However, despite recognising and reporting these symptoms, some parents did not get a diagnosis or referral until much later;

> She wasn't following her developmental milestones like my first child did. Erm, and I repeatedly kept going back to the GP and saying, "Look, there's something not right. She, she can't walk length, for a long period. She can't..." She was delayed in everything but no one took you serious. And it was only 'til she went into nursery school and that, and obviously they do their checks as well to fit in on the child's development file, that they found, er, her fine motor skills weren't right. And then sent off a letter to the GP and that was only then people started taking my- myself seriously. (Basu *et al.*, 2015)

Such findings are reflected in the literature by studies covering parent's experiences of the referral and diagnosis process in a range of childhood illnesses (Arksey, 1994; Dixon-Woods *et al.*, 2001; Lauritzen, 2004; Ostergaard, 2005; Finnvold, 2009; Sundaram, Day and Kirk, 2009; Usher-Smith, Thompson and Walter, 2013; Clarke *et al.*, 2014; Hubermann *et al.*, 2016). A common theme between these studies is of parents feeling that GPs dismissed their concerns. However, it should be noted that parents do not always develop concerns even when their child does have emerging difficulties and therefore do not report their child's symptoms (Lauritzen, 2004). Additionally, part of a GPs role is to gatekeep access to secondary services, due to limited numbers of specialists and as a way to control healthcare expenditure (Loudon, 2008). By developing a screening tool which utilises the concerns that parents raise and the signs that specialists use in treatment and diagnosis, both primary

health care professionals and parents may be supported in beginning to recognise the earliest symptoms of CP. Recognition of the earliest CP signs by both parties may then begin a conversation that results in the infant's early referral to a specialist.

However, to my knowledge there is no literature on the first concerning observations parents make when their infant is developing CP. Therefore, this thesis will look to rectify the lack of data, by surveying parents who have a child with CP asking about the abnormalities that they first noticed and the red flags that parents presented to their child's health care professionals. It will then determine if parental concerns have been included in the development of any current screening tools for early motor development, before carrying out interviews on the development of a new tool to help identify infants in primary care with unidentified CP.

1.6 Aims

1. Develop a list of early concerning observations made by parents whose infant was later diagnosed with Cerebral Palsy
2. Determine if parental concerns have been used in the design and development of currently available screening tools for early motor development.
3. Develop a tool from the above list with key stakeholders, that will help to identify infants within primary care with CP.

1.7 Outline of the thesis

This thesis will fulfil its aims through the following further five chapters.

Chapter 2 will present the findings of a caregiver survey on the earliest concerns they had of their infant's development and their experiences of referral and diagnosis process for CP. This chapter will show that low risk infants do experience delays to referral and diagnosis compared to high risk infants in the UK. It will also show that parents and caregivers identify the same and additional signs of CP compared to those reported in the literature.

Chapter 3 will present the pathways and delays occurring within the primary care referral pathway for CP. This chapter will show that delays occur due to parent and caregivers' appraisal and help-seeking behaviours, and due to delays within the diagnosis process. However delays to treatment do occur within the CP population after a diagnosis has occurred. It will also show that parent and caregivers' experiences of primary care are

determined by PHCPs acknowledging their concerns, the PHCPs' awareness of CP, and problems with the referral itself.

Chapter 4 will present the findings of a scoping review into the methods used to develop currently available screening tools. This chapter will show that only five currently available tools for screening motor development included parents in the development of the tool. Only two included parents in the development of the theoretical contexts of the tool, and only one included a parent of a child with CP. It will also show that the majority of the included tools developed their items from the same core sets of literature.

Chapter 5 will present how representative of the caregivers' concerns the items on currently available screening tools are. This chapter will show that the most commonly used items describe motor milestones and that no single tool identifies all of the concerns parents identified in Chapter 2. It will show that the tools aimed at parent completion do not address issues around tone, whereas tools aimed at Health Care Professionals (HCP) completion do not address parental concerns. Additionally, it will show that that tools aimed at parental completion typically use lay language, similar to that used by parents and caregivers making the tools accessible. Whereas tools aimed at HCP completion tend to use medicalised language that is not immediately compatible with the language used by parents and caregivers.

Chapter 6 will present the findings of a series of iterative interviews with key stakeholders on the development of a new screening tool. This chapter will outline how two information sheets were developed, the changes that occurred between the interviews, and the rationale for making these changes. It will also describe the four issues that emerged: 1) The types of tools parents want, such as information sheets rather than questionnaires, as well as how and when they want to access them, such as being given them by their Health Visitor during a Health Visitor appointment. 2) How best to improve parental understanding of atypical movement, such as the use of images and explaining what typical development looks like. 3) How much information is enough for parents to identify their infant's atypical movements without overwhelming them or causing false concerns. 4) Managing disagreements between participants across the interviews.

Chapter 7 will provide an overall conclusion of the thesis. It will explain the evidence for candidacy at the level of the parent and then at the level of the HCPs. This will be followed by a discussion of the implications for future research, my reflections, and suggestions for future research.

Chapter 2. Parent and Caregivers earliest concerns and experiences of Cerebral Palsy

2.1 Introduction

Despite previous research, there are still many unknowns when it comes to primary care referral for suspected Cerebral Palsy (CP) in the UK. These include what the earliest concerns parents have are, and how they relate to the CP signs reported in the literature. Do the primary care referral delays for CP reported elsewhere also occur in the UK? This chapter will aim to answer these questions to determine the earliest concerns and the current status of the UKs primary care referral system for CP. To do this an online survey asking about the earliest concerns and the referral experiences of parents and caregivers of children with CP was carried out. The survey received a good response rate, with a total of 240 responses included in the analysis. The outcomes identified that primary care referral delays are occurring within the UK. Parents and caregivers reported three types of concerns: day to day observations, motor milestones, and troubling medical history. Notably, compared to the symptoms reported in the literature, parents and caregivers identified the same and additional CP signs.

Aims

1. Determine if a delay in referral of infants subsequently diagnosed with CP occurs within primary care
2. Describe the earliest concerns caregivers have while their infant's CP is emerging

2.2 Methods

Caregivers of children with CP were invited to take part in an online survey. Ethical approval was granted by the Faculty of Medical Sciences Research Ethics Committee, part of Newcastle University's Research Ethics Committee.

2.2.1 Participants

Inclusion Criteria

a. Parent or caregiver of an infant with CP.
b. Informed consent
c. Ability and willingness to complete the survey

Exclusion Criteria

a. Parents or caregivers who did not look after the child in question before the child received a diagnosis of CP

b. Parents and/or caregivers of a child/adult who does not have a formal diagnosis of CP

c. Parents and Caregivers who reside outside of the UK

To allow for a ±5% sampling error and a confidence level of 95%, I aimed to recruit one parent or caregiver from 378 families with a child with CP (Welch and Comer, 1988).

2.2.2 Design

I undertook a cross-sectional web-based survey of parents and/or caregivers of infants who were diagnosed with CP.

2.2.3 Materials

The survey consisted of items covering the child's demographics, the earliest concerns caregivers had regarding their child's development, who reported the concerns, to whom the concerns were reported, the caregivers' experience of the referral and diagnosis process, and caregiver demographics. The survey was made up of multiple choice and free text items. Free text items were used for topics such as earliest concerns and experiences to reduce bias. All questions were forced response. However, participants were made aware that they may choose not to answer the questions on their experience of the referral and diagnosis process by entering 'N/A'. A copy of the survey is in Appendix A.

The survey was reviewed by the UK charity Scope (scope.org.uk), this was because of their extensive experience of working with families of children with disability across the UK as well as having a specialist CP programme which have worked with other research groups. It was piloted in 22 individuals known to the team from a variety of educational levels (in order of pilot testing, 6 researchers, 3 clinicians, 7 postgraduates, 3 undergraduates, and 3 college educated students). Pilot testers accessed the survey online through their computer or phone and were asked to provide written feedback on the accessibility and sensitivity of the items, if any items were leading, grammatical or spelling errors, time taken to complete the survey, as well as any technical issues they experienced. The survey was then piloted within

3 parents of children with CP known to the team. Both stages of pilot testing were carried out in an iterative manner until 2 testers raised no additional comments.

To assess the severity of each child's CP the Gross Motor Function Classification System Family Report Questionnaire (GMFCS) (Palisano *et al.*, 1997; Palisano *et al.*, 2008) was included within the child's demographics section. The GMFCS is a five-level ordinal classification system for determining the level of impairment an individual's CP has on their everyday life. A score of 1 indicates limited effects on everyday life, such as being able to walk without limitations. A score of 5 indicates severe limitations on everyday life such as requiring extensive assisted technology and physical assistance. The GMFCS is validated for children aged 2-18 years and is split into 4 questionnaires based on the child's age (2 years to <4 years; 4 years to <6 years; 6 years to <12 years; 12 years to 18 years). The GMFCS gives caregivers a written description of each GMFCS level appropriate to their child's age. After reading through the caregivers are asked to choose the description which most closely describes their child's ability. Before starting the GMFCS, caregivers were asked which age category their child is currently in (<2 years; 2 to <6 years; 6 to <12 years; 12 to 18 years; 18 years +). As the GMFCS is not valid for children <2 years, these caregivers were not required to complete items relating to the GMFCS. Caregivers whose children were 18+ years were presented with the 12 to 18 years GMFCS questionnaire as it has been shown to be reliable in adults (Gorter *et al*., 2011).

2.2.4 Procedure

Participants were recruited using e-flyers (shown in Appendix B) through UK based charities: Bobath (bobath.org.uk), Cerebra (cerebra.org.uk), CP UK (cerebralpalsy.org.uk), Heel & Toe (heelandtoe.org.uk) and Scope (scope.org.uk), Parent Carer forums and through social media posting. Once participants completed the survey, they were asked to share the survey link with their friends and followers on social media to encourage snowball sampling. (Kosinski *et al.*, 2015; Marengo, Giannotta and Settanni, 2017; Devlin, 2018).

The survey was hosted online using Boston Online Surveys. Participants went through the survey in the order shown in Figure 2. If participants did not consent to take part, they were taken to a 'thank you' page and exited the survey.

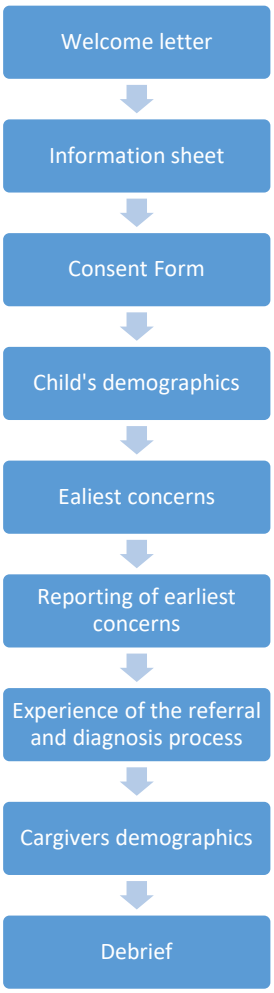


Figure 2 Order of the survey sections that participants accessed

The survey was open between 5/6/2019 and 15/11/2019. In that time the survey was accessed 2,328 times with 266 full responses given (11.43%). 11 responses were excluded due to the participant not being UK based (N=4), miscellaneous or likely erroneous reported limb involvement distribution, such as just the neck being affected (N=4), and no information given about the earliest concerns they developed (N=3).

2.2.5 Quantitative data analysis

Quantitative data analysis was carried out using IBM SPSS statistics Version 24. Due to large differences in sample sizes, non-parametric statistical analysis was used in the analysis of:

1) Delays in receiving referral to therapy and receiving a diagnosis compared between primary care and secondary care. To determine the time between receiving a diagnosis and receiving a referral for therapy, the age in which the referral to therapy was given was subtracted from the age at diagnosis. As this data was collected categorically the categories were converted into months before the subtraction. The categories: 12 months-17 months;

18 months – 23 months; 2 years; 3 years; 4 years; and 5 years, were converted to 12 months; 18 months; 24 months; 36 months; 48 months; and 60 months respectively. The category 6+ years was excluded from the analysis due to it having no upper age limit.

2) Differences between infants whose first signs were identified in primary care and secondary care. The differences were limb involvement distribution and CP severity. They were assessed using a Chi-squared test.

3) Differences between infants who received an immediate or a delayed referral from primary care. The differences assessed were age at the time the first concern was raised, the limb involvement distribution, and CP severity. The infant's age at the time of the first concern was assessed using Mann Whitney U tests, while limb involvement distribution, and CP severity were assessed using Chi-squared tests.

4) To determine if raising a specific concern influenced the speed of the referral within primary care. The concerns caregivers raised were identified through thematic analysis, as described in *qualitative analysis* below. Once the thematic analysis was complete, the frequency with which caregivers raised each concern was calculated for the immediate and delayed referral groups using the crosstab query function within NVivo 12 (Version 12.6.0.959; QSR International). The frequency with which concerns were raised between the immediately referred group and the delayed referral group were then assessed using Fishers exact tests.

Due to small sample numbers (n=6), infants with monoplegia were not included in analysis of limb involvement distribution. Infants under 2 years were not included in analysis involving GMFCS as GMFCS is only validated for infants aged 2 years and over.

Cases where it was not clear who raised the concerns or if the concern was raised in primary or secondary care (n=8) were not included in analyses comparing primary and secondary care.

2.2.6 Qualitative data analysis

All qualitative analysis was carried out in NVivo 12 (Version 12.6.0.959; QSR International) after the free-text qualitative data was pseudonymized. All responses underwent thematic analysis using a realist, inductive, semantic approach (Braun and Clarke, 2006) to identify the earliest caregiver concerns. The themes identified for the earliest concerns were then

mapped out against the categories used by Garfinkle *et al.* (2020), shown in Appendix C. Although thematic coding was carried out by one researcher (JB), the themes were discussed across the research team.

2.3 Results – Referral delays

This web-based survey of caregivers of children with CP demonstrates significant delays in referral from primary care for diagnosis and therapy compared to infants identified in secondary care. This section will describe the sample the survey was carried out in, the delays reported between caregivers whose infants were identified as at risk in primary and secondary care, as well as factors that may influence the time of referral.

2.3.1 Participants

255 responses were analysed. 240 responses (94.1%) were from Mothers and the respondent median age was 39 years (Range 20-73 years). Respondent demographics are shown in Table 1.

The median age category of the children described was 6-11 years, the most frequently reported limb involvement distribution was Hemiplegia and the modal GMFCS score was 2 (Table 2). 56.3% of the infants were identified as presenting concerning features in primary care (Table 3). 1 infant was identified by their school teacher who reported their concerns to secondary care. It was unclear as to where concerning features were identified for 8 infants. 34.1% of the sample was diagnosed by the age of 1 year, 69.4% by the age of 2 years, and 87.1% by the age of 3 years. 1 infant was diagnosed after 6 years of age.

2.3.2 Delays in referral and diagnosis

Infants identified in primary care were significantly older (Primary care median age category = 12-17 months; $U=4536$, $p<.001$, $z=-5.5$) when they first received therapy compared to their counterparts identified in secondary care (Secondary care median age category = 8 months). Infants identified in primary care were significantly older (median age category =18-24months; $U=5356$, $p<.001$, $z=-4.1$) when they received their diagnosis compared to their counterparts identified in secondary care (median age category =12-17 months). However, there was no significant difference ($U=6974$, $p=.454$, $n=244$) in the time between a diagnosis and being referred on for therapy between infants identified in primary (mean rank =125.56; median age= 2 months) and secondary care (mean rank =118.83; median age = 4 months)

These results demonstrate that infants referred from primary care are older than those identified in secondary care when they receive therapy and diagnosis. As there is no significant difference in the delay between receiving a diagnosis and being referred for therapy between the groups, it can be deduced that the delay is occurring within the primary care referral process.

2.3.3 Differences between infants identified in primary and secondary care

Several factors may result in a delay in the referral to secondary care from primary care. These include the affected limb distribution and the severity of CP.

The limb involvement distribution was significantly different between those whose symptoms were first identified in primary and those identified in secondary care. Those identified in primary care were significantly more likely to have hemiplegic or diplegic CP compared to infants whose symptoms were identified in secondary care, whom were more likely to have quadriplegia and triplegia CP ($\chi^2_{(3, 239)}$ =23.2, p<.001), Table 4.

Similarly, the severity of the CP the infants were later diagnosed with was significantly different between those whose symptoms were identified in primary and those identified in secondary care. Those identified in primary care had less severe CP (Modal GMFCS =2) than those identified in secondary care (Modal GMFCS=5, $\chi^2_{(4,232)}$ =28.2, p<.001), Table 5.

These results demonstrate that infants whose symptoms were first identified in primary care have fewer limbs involved and to have less severe CP than infants whose symptoms were identified in secondary care. However, not all infants whose symptoms are identified within primary care receive an immediate referral, therefore the next step was to investigate if the same patterns occurred between those who did or did not receive immediate referral from primary care.

2.3.4 Differences between infants identified in primary care who did or did not receive immediate referral.

The median infant age caregivers first reported their concerns in primary care was 6 months (n=136). However, caregivers whose infants did not receive an immediate referral reported their concerns significantly earlier (median infant age = 6 months, n= 69) than those who did receive an immediate referral (median infant age = 8 months, $\chi2_{(17,118)}$=29.8, p=.028).

Relationship to infant	
Mother	240
Father	8
Grandmother	6
Other family member	1
Median age (range)	39 years (20-73 years)
Highest level of education	
GCSE level or equivalent	41
A Level or equivalent	67
University degree	147
Ethnicity	
White European	248
Asian other	1
Black African	1
Black Caribbean	1
Indian	1
Other	3
Employment status	
Employed full time	65
Employed part time	85
Full time carer	63
Full time homemaker	22
Unemployed and looking for work	2
Unemployed due to health	3

Retired	1
Full time student	2
Maternity/Paternity leave	4
Other	8
Marital status	
Married/Civil partnership/Co-habiting with long term partner	209
Divorced/Separated	16
Single	28
Widowed	2

Table 1 Respondent demographics

Limb distribution and severity were not significantly different (p=.512, p=.485 respectively) between infants who did or did not receive immediate referral.

The results demonstrate delays occurring in primary care referral. Infants whose symptoms are identified in primary care, experience delays in referral to secondary care for therapy and diagnosis. However once referred to secondary care there are no additional delays between receiving a diagnosis and receiving therapy compared to infants whose symptoms were identified in secondary care. Within primary care, the earlier in their infant's life a caregiver reports their concerns the more likely their infant will not receive an immediate referral. Although differences occur in limb distribution and severity between those identified in primary and secondary care, these differences did not explain why infants whose symptoms were identified in primary care did or did not receive immediate referral. The next reason for immediate or delayed referral to be tested is the nature of the concerns raised by caregivers within primary care.

CP type	*Total (n)*	*Percentage of the sample (%)*
Hemiplegia	118	46.3
Quadriplegia	75	29.4
Diplegia	36	14.1
Triplegia	20	7.8
Monoplegia – Lower limb	5	2.0
Monoplegia – Upper limb	1	0.4
Total	255	
Age group		
Under 2 years	17	6.7
2-3 years	61	23.9
4-5 years	45	17.6
6-11 years	77	30.2
12-17 years	34	13.3
≥18 years	21	8.2
Total	255	
GMFCS		
Under 2 years	17	6.7
1	63	24.7
2	67	26.3
3	46	18.0
4	27	10.6
5	35	13.7
Total	255	

Table 2 Children's demographics

Concerns raised in	*Concerns raised by (n)*			
	Caregiver	*Health Care Professional*	*School teacher*	*Unclear*
Primary Care	135	6	0	0
Secondary Care	56	49	1	1
Unclear	0	0	0	7

Table 3 Infants identified in primary and secondary care by their caregiver or health care professional.

Cerebral Palsy Type	*Concerns raised in Primary or Secondary care (n)*		
	Primary	*Secondary*	*Total*
Hemiplegia	79	39	118
Diplegia	24	12	36
Quadriplegia	23	46	69
Triplegia	8	11	19
Total	134	108	242

Table 4 Limb involvement distribution against identification of the infant in primary or secondary care

GMFCS	*Primary care (n)*	*Secondary care (n)*
Under 2 years	7	9
1	42	20
2	45	22
3	26	20
4	11	16
5	5	25

Table 5 Frequency of gross motor function classification scores of infants identified within primary and secondary care

2.4 Caregiver reported concern results

In response to the web-based survey for caregivers of children with CP, caregivers reported their earliest concerns about their infants' development. The concerns fall into three overarching areas; *Day-to-day observations*, consisting of caregiver concerns regarding infant development that were not based on milestones or the infant's medical history. *Motor milestones*, consisting of delayed milestones and milestones being met in an atypical way. The third area, *Troubling medical history*, consists of concerns around their infant's medical history. This section will begin by discussing the content of the concerns. Day to day observations will be discussed first as they consist of the caregivers' observations and are seemingly the least influenced by medical discourse. Motor milestones will be discussed next as although influenced by the medical discourse, some caregivers provided more lay descriptions of what they observed. Troubling medical history will be described last due to the use of medical language rather than lay descriptions of their observations by caregivers. This section will then end by providing a quantitative analysis of the frequency with which the concerns were reported between the infants who received immediate or delayed referral.

2.4.1 Day-to-day observations

Day-to-day observations consist of caregiver concerns regarding infant development that were not based on milestones or the infant's medical history. They focused on general overarching issues such as parental instinct and feeding difficulties, as well as specific aspects, such as tone.

Parent instinct: Some caregivers reported having a 'gut feeling', 'instinct' or just knowing that something was not right with their infant, despite their infant having 'no physical signs' (M071) that they could report.

Temperament: Caregivers also described their infant's temperaments as either 'unsettled', 'too settled', or as situational. Unsettled infants were described as seeming to be distressed most of the time and were 'very difficult to clam [*sic*]' (M207) or 'comfort'. They were described as needy, and their crying made their caregivers feel that they were unable to put the infant down. However, some noted that their infant only became distressed in specific situations, such as dressing, tummy time and baby massage. They described their infants as 'not tolerating', or 'being extremely uncomfortable' when in these situations. Some also

noted that their infant only settled if they were laid down in a particular position, such as on their side. In contrast, some infants were reported to be 'too quiet', 'too good', 'very passive', 'too easy', and did not react to sounds around them, which caused the caregiver concern. However, one caregiver reported that she 'wasn't worried because this baby slept and ate so much better than my first' (M062) suggesting being too settled could also prevent some caregivers from developing concerns.

Sleep: Caregivers described their infants as tired, unable to sleep, having sleep issues, or that sleep was problematic or poor. Sleep issues were reported to occur during day and night. While one caregiver noted that their infant was still unable to sleep through the night at 7 years of age, another noted that because their infant slept well they had not initially been worried about their infant.

Feeding Difficulties and Physical Development: Feeding difficulties were also reported to occur throughout the first year of life. Infants were described as feeding for extended periods of time, struggling to 'suck' or 'swallow' milk and struggling to latch on, with both breast and bottle feeding. Infants 'coughed', 'gagged' or 'choked' when swallowing liquids, pureed foods, food with lumps, and solid foods and suffered from reflux, were unable to eat without being sick, or had a food intolerance or allergy. Some caregivers reported they were given interventions to improve feeding including; nasogastric tube, gastrostomy, cup feeding, and a squeeze bottle with a special teat designed for babies with cleft palates. Infants with delayed physical development were also described as 'not gaining weight' or 'failing to thrive' regardless of whether they were described to have feeding difficulties.

Eye Gaze: Eye movements were described in some infants as struggling to fix and follow, not looking at the caregiver, or being unable to maintain eye contact. Infants were described as looking vacant, or that their eyes would wander or roll. One infant was described as being unable to move their eyes independently of their head. Some caregivers stated that their infant had medical conditions such as nystagmus, ptosis and squint, or one eye that turned inwards.

Movement: Some caregivers observed that their infants did not use two hands to hold larger objects (bimanual movements). Some infants were described as not use their hands to pull their feet to their mouths or chest or to 'Never brought two hands together' (M065) (midline

movements) even when the caregiver tried to help them. They used a range of terms to describe infant movement quality, the lack of strength in their movements and their general lack of movement, shown in Table 6. They noted that these qualities became more apparent when in the bath, during play, or while their infant was waking. Some also noted that the concerning feature in their infant's movement quality was only present for a couple of months before it then disappeared, or they noticed that it developed over time.

Asymmetrical movements: Caregivers reported that asymmetrical movements were observed between the infant's hands, legs, and whole body. They outlined how the infant did not use, rarely used, or tried not to use one of their arms for tasks such as picking toys up. For example, an infant was described to 'look at something to his left but reach over with his right.' (M028). Some caregivers labeled the hand asymmetries as an 'early hand preference'. Similar reports were made of asymmetries in an infant's kick, with them noticing this when 'in the bath or swimming' (M068), on a playmat, or in a bouncer or chair.

Asymmetries across the whole body were also described as the limbs on one side of the body not moving as much, with some labelling their description as a 'dominant side'. Some infants who were reported to have a dominant side had an early head preference to the same side. Caregivers described noticing their infant's dominant side while the infant was laid on the floor or while playing.

Reactions and Reflexes: Caregivers stated or described a range of reflexes that their infants either had or did not have or were delayed to use, such as the step reflex. The startle reflex was the most frequently reported concerning reflex, with infants being easily startled and/or very or severely startled. They noticed the startle reflex when something approached their infant's affected side or startle-like movements when asleep. One caregiver described startling movements as a whole body flexion followed by stretching of the infant's limbs.

However, some reported that their infants showed 'no reaction' (M224) to any sounds around them or when their affected limbs were touched. Some caregivers reported their infant having 'brisk knee reflexes', though this is almost certainly a feature described to the caregivers after their infant was medically examined.

Terms used to describe movement quality	*Terms used to describe lack of strength*	*Terms used to describe lack of movement.*
Jittery	Weak limbs	Hardly moving
Twitchy	Weak core movements	Physically inactive
Writhing	Lack of or little strength	Not moving their limbs
Un-coordinated	'Shaking under his weight' (M033)	Making no effort to move
Non-fluid		
Cyclic		
Constant stretching		
Un-controllable		

Table 6 Terms used by caregivers to describe movement

Posture: Caregivers also reported concerns around the posture of the infants, in relation to each of the head, limbs and torso. Most often caregivers described their infants' hand(s) being clenched or not opening. The clenching was described as being 'like that of a stroke victim' (G266) and caused one infant to 'cut the palm of his hand with his fingernails and got [*sic*] an infection.' (M021). Arm posture was the next most frequently reported postural concern and was often reported alongside the infant's hand posture. They described their infants' arm(s) as 'always tucked into her chest' (M129) or 'held arm in a bent position' (M078). Reports about head posture and leg posture occurred at similar rates. Although head posture was typically described as the infant 'looked to one side and couldn't move his head to the other side' (M037) leg posture was described in multiple ways. Examples include that their 'legs would cross or scissor' (M252), or looked 'awkward' (G242) or 'odd whilst laying in the bath' (G242). Unlike the posture of hands and arms, foot posture was reported separately to leg posture. Feet were described as 'turning in' or 'out', as well as being 'always curled up' (M077). In contrast body orientation was rarely reported but was consistently reported as either 'arching backwards' or 'weird'. One caregiver added that at

2.5 years her sons 'pelvis completely twisted from lack of intervention with his muscles tensing up' (M261)

Tone: Caregivers described changes in tone through a variety of terms, or by describing what the infant could not do. An example follows:

> When I attended a baby group and mums with younger babies could clap their baby's hands together, when I tried I couldn't get his hands anywhere near each other to imitate clapping. […] When changing his nappy I was unable to open his legs so had to start using pull ups (M168).

This example highlights that caregivers may not use key words to describe their observations of tone but instead use comparisons to other infants, state what the infant cannot do, or describe the coping mechanisms they have developed, such as switching to pull up nappies.

Caregivers noticed their infants' tone during everyday situations such as nappy changing, dressing, picking their infant up, and during play. Some noticed their infants' tone was mixed, others that it increased over time.

In contrast, caregivers who described decreased tone reported their infants to be floppy, some labelled this as 'low tone' or 'hypotonia'. They noticed their infants' floppiness from birth or when bringing the infant home for the first time. One caregiver noted that after every change in height or weight the infant would become more floppy.

2.4.2 Developmental Milestones

Some caregivers reported that their infants had delayed milestones, or met their milestones in an atypical way which caused the caregiver concern.

Delayed milestones: Caregivers who described their infants' milestones as delayed often reported that just the milestone was delayed, for example delayed crawling, with no other explanation as to if or when the milestone was achieved. Due to the lack of description the delayed milestones reported are presented in Table 7.

Atypical Sitting: Some caregivers described their infants as flopping, sliding, slipping, tilting or slouching while sitting in a stroller, a bouncer, a high chair, on their bottom, or on a caregiver's knee. Some also reported that their infant began 'W sitting' (F150).

Caregiver Reported Delayed Milestone
Head control
Reaching and grasping
Babbling and Speech
Smiling
Sitting
Rolling
Crawling
Standing
Walking
Depth perception
Dribbling (Infant never stopped)
Fine motor skills
Jumping
Self-feeding
Social Skills

Table 7 Milestones described by caregivers of children with Cerebral Palsy as being delayed

Atypical Crawling: Upon learning to crawl, some infants' crawls were described as 'not normal' or 'not typical', with their crawl styles described as 'army' or 'commando' crawl or as back or bum 'shuffling'. Some caregivers described their infants as 'pulling', 'dragging', 'propelling' or 'pushing' themselves with their unaffected limb(s) while their affected limbs were described to not be 'symmetrically reciprocating' the crawl, being 'dragged behind', 'trapped underneath' the infant's body, or to just be 'kicking'. When an infant still used their affected arm, caregivers noted the affected hand was used as a fist or the back of the affected hand would be placed on the floor rather than the palm

Atypical Rolling: Others noticed that their infants could or 'would only roll over in one direction' (M166), resulting in the infant getting stuck as they 'couldn't turn the other way'

(M166). Some also noticed that their infants' rolling pattern was different to other infants, but did not provide a description.

Atypical Standing: Infants who learnt to stand atypically 'pronated' or 'tip-toed' on their affected foot, some were described to fall over a lot or fall backwards when holding on to something. One caregiver noticed that their infant's balance decreased with each growth spurt in height or weight. Some infants 'would not', 'could not', or 'were unable to' weight bear through one or both of their legs, and would 'retract [their] legs up into fetal position' to avoid weight bearing. One caregiver also noted that their infant was unable to use their 'legs properly' (M262) when trying to pull to stand.

Atypical Walking: Some infants who progressed to walking held their affected arm(s) inwards or did not move their affected arm while walking. Some bent their knees or turned their leg(s) inwards, with their thighs held tightly together. Infants were described to tip-toe, or were unable to put their affected foot down flat. The infants' affected foot/feet were described as twisted, turned/pointing inwards or outwards while walking.

Infant gaits were described as looking funny, unusual, clumsy, ape like, or as wide, with some infants dragging their affected foot behind them resulting in the infants looking as if they were limping, or they would walk upstairs by placing both feet on each step.

The infants' progression after beginning to walk was described as slow and took a long time to get better. Infants fell over a lot and were unable to take more than a few steps at a time for several months. Caregivers explained their infants falling as being due 1) to the infant's affected leg staying behind, 2) poor balance, or 3) that there was no reason they could identify.

2.4.3 Troubling Medical History

Some caregivers reported their infant's medical history as their main concern. Often these concerns related to complications or conditions around the time of birth that were identified in hospital, such as 'hydrocephalus'. They also used medical language to describe their earliest medical based concerns, such as;

> Hypoxic brain injury at birth caused by placental abruption, cord wrap and placental insufficiency, requiring [##] minutes of resuscitation, therapeutic hypothermic cooling (M121).

It is likely that the caregivers learnt the terminology from interactions with Health Care Professionals treating their infant resulting in their lexicon becoming more specialized.

2.4.4 Comparison of reporting frequency between immediate and delayed referral from primary care

Across the concerns raised in day-to-day observations and motor milestones only two significant differences in reporting frequency were identified. *Movement,* from day-to-day observations, was reported significantly more frequently by those who received a delayed referral (29.0%) compared to those who received an immediate referral (9.1%, p=.004). In contrast *atypical walking* was more frequently reported by those who received an immediate referral (24.2%) than those who received a delayed referral (8.7%, p=.019).

Overall, the results demonstrate that caregivers report a variety of concerns based on their day to day observations of their infants, on their infants meeting their developmental milestones, and on their infants medical history. However, despite the range of concerns, only two concerns are shown in different frequencies between those who do or do not report receiving immediate referral from primary care.

2.5 Discussion

2.5.1 Referral delays

This study determined that UK infants with CP identified within primary care, on average, experience longer delays in referral compared to their counterparts identified within secondary care. This replicates the findings of both Hubermann *et al.* (2015) and Boychuck *et al.* (2020) who found a 6 month difference in referral ages between primary and secondary care when looking at medical notes. This study also found that caregivers who noticed and then raised their concerns earlier were more likely to receive a delayed referral compared to caregivers who raised their concerns later. This is likely due to later reporting parents raising concerns around more concrete signs such as failure to start walking. However, without the GPs viewpoint the reasons for this delay can only be speculative.

2.5.2 Caregiver concerns and how they relate to the literature on CP signs

This study identified three overarching themes in the types of concerns that caregivers raise to Primary Health Care Professionals (PHCPs). Caregivers described their earliest concerns around their day-to-day observations of their infants, developmental milestones, and troubling medical history. Notably, the concerns raised by caregivers are reflective of the

signs of CP reported by clinicians. Garfinkle *et al.* (2020) in their scoping review split early CP signs into three categories, 'Early features from clinical history', 'Early features from questioning and examination of developmental milestones' and 'Early features from the neurological examination'. Of the CP signs collated by Garfinkle *et al.* (2020) the caregivers described all of them except 'high-pitched or weak cry' from clinical history and 'postural reactions' from neurological examination. Although some neurological examination items were raised, such as brisk knee reflexes, these items were typically not identified by caregivers, as to be expected.

The caregivers did provide additional concerns which were not previously identified by Garfinkle *et al* (2020): atypical sitting, bimanual movements, changes in balance with growth, dragging of limbs while crawling, eye gaze, facial drooping, gut feeling/instinct, midline movements, poor balance, and the posture of the arms and feet. Caregivers highlighted the situations and activities where they noticed these concerns, such as when the infant was in water, during play, when laid down, and when sat up. Furthermore, they used a range of lay terms to describe their concerns, none of which were highlighted in the scoping review. Therefore, the results of this study demonstrate the ability of caregivers to be able to recognize their infants' earliest signs. It also adds new potential signs of early CP that could be investigated for their accuracy in early screening, highlights the range of terms caregivers use when raising concerns and the specific situations that should be considered, such as bathing.

2.5.3 What are the potential causes for delayed referral? - The lack of a key symptom.

A potential reason for delayed referral is that the concerns reported by caregivers are non-specific and therefore may not cause PHCPs to initially recognise the concerns as symptoms as early CP. Research into the way GPs handle cases with non-specific vs specific symptoms demonstrates a need for key symptoms, or 'red flags' to be presented which distinguishes that condition from others. For example, Molassiotis *et al.* (2010) retrospectively interviewed cancer patients about their experiences from the initial change in their health to receiving a diagnosis. Participants who reported to their GP that they had found a lump as their concern, were typically referred quickly with a cancer diagnosis. However, those who did not report a lump, reported their initial interaction with a GP resulted in being given a misdiagnosis or a treatment that was later deemed inappropriate. They suggest that the red

flag of a lump was needed for the GPs to recognise the symptoms as being predictive of cancer.

Although Molassiotis *et al.* (2010) did not include the perspectives of GPs, Usher-Smith, Thompson and Walter (2013) retrospectively interviewed the GP and the family of children recently diagnosed with Type 1 diabetes. Usher-Smith, Thompson and Walter (2013) highlighted a range of reasons which can make diagnosing conditions in children difficult such as: the subtlety and vagueness of symptoms; the individual not presenting as expected for the condition; that most children they see have self-limiting illness'; and that they do not want to impart unnecessary worry and anxiety on to parents. In terms of the concerns reported by our sample, the symptoms are often vague, overlap with standard variation within typical development, and overlap with self-limiting conditions. However, unlike CP, infants at the lower end of typical variation and those with milder developmental delays are able to catch up with the peers without intervention. When caregivers raise early concerns there is a possibility that referral is delayed due to CP having no key red flag symptom, making CP difficult to identify from typical development or self-limiting conditions. Although reporting concerns around atypical walking did increase the chance of the infant receiving an immediate referral, this was only reported by 24% of those who received an immediate referral from primary care. Additionally walking occurs relatively late in development, with typical variation in learning to walk occurring up until 18 months corrected age.

2.5.4 Utilisation thresholds

In addition to the lack of a key symptom, the caregiver's perceived utilisation threshold may also be a cause for delay. The utilisation threshold is a concept first outlined by Michiels-Corsten, Bösner and Donner-Banzhoff (2017). It refers to the way, that because of knowledge gained through continuity of care with specific people, GPs can become aware of the factors that influence their patient's decision to seek help and tailor their diagnosis process accordingly. As such, individuals with lower utilisation thresholds are thought to seek care earlier than patients with high thresholds. This aligns with this study's finding that caregivers who raise concerns earlier are more likely to experience delays in referral from primary care. Michiels-Corsten, Bösner and Donner-Banzhoff (2017) reanalysed interviews of 12 GPs talking about their diagnostic reasoning across a total of 295 consultations. In particular, the GPs described their low threshold patients to be 'anxious' and 'sensitive', and

their high threshold patients as 'withdrawn'. The GPs described struggling to take their low threshold patients seriously, as they could become irritated by them, would doubt that there would be a severe disease outcome so would stress the benign course of their patient's symptoms and give reassurance to their patients. In contrast, when interacting with patients with high thresholds, GPs reported elevated awareness and concentration, they put more time and effort into their diagnostic work, they reported making more effort to identify a potentially serious disease and were more likely to refer the patient to specialists.

The concept of utilisation thresholds suggests that multiple factors about the specific patient influence how GPs perceive the patient's threshold, with perceived high threshold patients receiving more direct treatment than patients with perceived low thresholds. Although Michiels-Corsten, Bösner and Donner-Banzhoff (2017) data looked at consultations with patients rather than with parents, similar attitudes towards parents utilisations thresholds are described by Clarke *et al.* (2014). Clarke *et al.* (2014) retrospectively interviewed parents of children diagnosed with Leukaemia and their child's GP about the diagnosis process. The GPs reported that they drew on the contextual information they knew about the family to determine if to take the concerns seriously at that time. Concerns from parents deemed 'sensible' were given greater concern than from parents deemed 'neurotic' or 'worriers'. Although other research has not so candidly reported GPs using such contextual cues to determine if to take parental concerns seriously, studies into delays within paediatric primary care have highlighted parents being called 'worriers' or 'overreacting' (Hinton and Kirk, 2015) or parents and GPs disagreeing about the seriousness of the infants' symptoms (Dixon-Woods *et al.*, 2001; Usher-Smith, Thompson and Walter, 2013). Therefore, in addition to the lack of key symptoms and the non-specific nature of the symptoms, the factors that influence the way GPs perceive the parent's utilisation threshold may also be causing delays.

2.5.5 Disclosure

Delays may also be occurring due to the information disclosed within the appointment. Multiple studies have highlighted that adult patients do not always report all of their symptoms to their PHCP. For example, Paskins *et al.* (2018) found that in a sample of 190 over 45 year olds, 22.6% of the sample failed to disclose a symptom they had previously identified in the waiting room as wanting to discuss with their GP just minutes later.

Although Paskins *et al.* (2018) did not address the reasons for non-disclosure, others have reported reasons such as; the patients ability to explain the complexity of their illness (Peters *et al.*, 2009), time constraints (Peters *et al.*, 2009; Houwen *et al.*, 2017), the patient determining the symptom to be most relevant to discuss (Shaw *et al.*, 2001; Bugge, Entwistle and Watt, 2006), and the behaviour of the GP (Bugge, Entwistle and Watt, 2006; Houwen *et al.*, 2017; Houwen *et al.*, 2019). When concerns are not disclosed, further delays can occur. For example, Shaw *et al.* (2001) found that some of their participants 'just mentioned' concerns during regular check-ups, and if the GP did not respond the patient would not broach the subject again. As the GP did not comment on the concern, these patients waited until their next appointment to raise their concern a second time, resulting in a delay.

2.5.6 Clinical need to see the patient

However, in the paediatric literature on a specific clinical issue, respiratory tract infection, we know that what the clinician observes influences treatment decisions. Cabral *et al.* (2019) found that in paediatric respiratory tract infections 34 of the 56 cases evaluated in the study were advised to continue home care, ranging from the 'watch and wait' approach to detailed care instructions, regardless of how the parents presented their concerns. 15 of the parents who took part implied wanting antibiotics during their consultation, however only 2 received antibiotics. Yet for the 11 cases where antibiotics were prescribed, the clinician based their decision on their own clinical observations, such as yellow phlegm, rather than the potential diagnosis given by the parents. Horwood *et al.* (2016) found similar results when interviewing PHCPs about their experiences and decision making around children with respiratory tract infections. Prescriptions were often given out based on the children's symptoms that were observed by the PHCP. Some prescriptions were given if they felt the parent would not return or would have access issues, or that the parent had already presented multiple times with the same concerns. However, when parents pressed for antibiotics which may not have been clinically warranted, the PHCPs used a range of strategies to prevent prescribing them. Although these studies focus specifically on respiratory tract infections, we know that needing to observe symptoms first-hand is central to a lot of clinical decision making. The caregivers in this survey described how their infants' signs only appeared in specific situations, such as in the bath, which would not be observable within the GPs office. The combination of CP symptoms emerging over time, symptoms only occurring in specific situations and the need for first-hand observations at a time where

symptoms are not the most evident may explain why the earlier caregivers report their concerns the less likely they are to receive an immediate referral. A potential solution is to ask caregivers to video their infants' concerning behaviours, however this excludes those without knowledge or access to such technology.

2.5.7 Lack of CP awareness in PHCPs

These delays may also be occurring due to GPs lack of awareness of the importance of early CP referral. Freedom of information requests submitted by Action CP in 2016 and 2018 identified that CP specific training for PHCPs is not standardised across the UK, is infrequent, and that often it would be included with generic disability training (Action CP, 2016; Action CP, 2018). Although the content of these training courses was not described, it is clear that PHCPs are being provided little training on early CP signs and the importance of early referral. Combined with the lack of key symptoms, non-specific concerns, utilisation thresholds, disclosure of concerns, and a clinicians preference to rely on their observations, a lack of training and awareness suggests that there are likely multiple factors resulting in delay.

2.5.8 The impact of COVID-19 on CP referral from primary care

It should be noted that 4 months after the survey closed, March 2020, the UK entered lockdown to deal with the COVID-19 pandemic. At the start of lockdown, the UK government asked PHCPs to carry out consultations over the telephone or through video, and by April 2020 90% of GP consultations were occurring remotely (Murphy *et al.*, 2020). Between 21st March 2020 and 5th June 2020 the number of contacts infants under 1 year old had with GPs had decreased by 29.3% on the previous 4-year average in the same time period (Foley *et al.*, 2022) and routine referrals for children and young people were reported to have fell by 89% compared to pre-covid levels (Morris and Fisher, 2022).

Although the direct impact of the pandemic and the switch to remote GP consultations on early CP identification has not been investigated, the effects on other conditions and GP practice in general have been reported. While interviewing UK GPs, Archer *et al.* (2021), Borek *et al.* (2021), and (Murphy *et al.*, 2021) all found that by not being able to see the patient in person GPs felt less able to assess subtle symptoms and signs, and therefore felt that they had to take more risks in their decision making by trusting what they were told by patients. Archer *et al.* (2021) also found that some GPs felt that they had lost their 'gut

feeling' about a patient presenting with something serious. The GPs picked up on patients becoming more reluctant to seek help due to the risk of catching COVID-19 and due to public health information advising they stay at home (Archer *et al.*, 2021; Borek *et al.*, 2021). This lead to some GPs beginning to believe that the 'watch and wait' approach may not be appropriate in every case due to not knowing how late patients are presenting. Despite this, routine referrals across the NHS e-Referral service, although increasing, have not returned to pre-pandemic levels (British Medical Association, 2022). It is likely that different elements of these changes in practice also extend towards infants with emerging CP and may have further exacerbated the delays to referral.

However primary care services were not the only services affected. To cope with the number of COVID-19 patients, large numbers of paediatric secondary HCPs were redeployed to adult services resulting in negative impacts to the care pathways. This resulted in delays ranging from delayed or missed routine outpatient appointments to delayed surgery. During the pandemic, the delays in outpatient appointments resulted in functional deterioration and deterioration of comorbidities in children with physical neurodisabilities, including CP, already in care pathways (Cadwgan *et al.*, 2021; Arichi *et al.*, 2022). Because of these delays there still remains a backlog to care. Evidence submitted to the House of Commons Health and Social Care Committee by the Royal College of Paediatrics and Child Health (RCPCH) suggested that over 267,000 children and young people in 2021 were waiting for treatment in the UK (Health and Social Care Committee, 2021; RCPCH, 2021). However, delays to treatment can cause further negative consequences, which in turn can require their own treatment. As such, infants referred for emerging motor difficulties, synonymous with CP, are likely being affected by this backlog, resulting in later therapy opportunities and, as a result, poorer outcomes. Developing materials that help identify infants with emerging motor difficulties may not only improve identification and referral within primary care but may allow for streamlining of referrals to ensure infants at risk receive therapy in a timely manner.

2.5.9 Limitations

This study has two main limitations: a non-representative sample, and a lack of input from GPs. 97% of the sample identified themselves as white European, whereas only 81.7% of the English and Welsh population declared themselves as having white ethnicity in the 2021

census (Office for National Statistics, 2022). Every year the GP patient survey is conducted within the UK to allow patients to feedback about their experiences and the services they have received (Ipsos MORI, 2022). The GP patient survey has continually shown patients from any Asian background, any mixed background, and any other ethnic group to report more 'poor' experiences with their GP practice than those from white or black/African/Caribbean backgrounds (Ipsos MORI, 2022). Studies using the GP patient survey data (Mead and Roland, 2009; Kontopantelis, Roland and Reeves, 2010) have shown ethnic minorities are significantly less able to get an appointment on the same day or within 2 days of asking or to get an appointment with a particular GP, compared to white patients. Additionally, they are significantly less satisfied with their GPs opening hours and being able to get through to their GP surgery on the phone. PHCPs have also reported that patients from minority ethnic backgrounds can have different cultural expectations and understandings of the UK health care system and that they may have language difficulties which act as further barriers to them accessing medical care (Robinson *et al.*, 2022).

The included sample is over represented with university degrees (57.6%) compared to the general population (33.8%) (Office for National Statistics, 2023). Parental education typically predicts the income within the household, and therefore is often used as an indicator of socioeconomic status (SES) (Davis-Kean, Tighe and Waters, 2021). Individuals with low SES often experience barriers to participating in research due to factors such as; feeling unqualified to take part, negative financial impact, and requirement for additional carer time to aid participation (National Institue for Health and Care Research, 2020). Also the quality of health care they experience is typically worse and they often have poorer health literacy (QualityWatch, 2020). As such, it is unlikely that these findings fully represent the language and experiences of those from lower SES.

Additionally, the modal age for the children reported about was 6-11 years, with 22% of the sample responding about a child aged over 12 years. Primary care practices have changed over the last two decades, including the development and implementation of care pathways and treatment guidelines (National Institute for Health and Care Excellence, 2017a; Action CP, 2018). As such, the responses may not be reflective of the current process. By not having a proportional representation of the current UK population this study may not fully represent the first concerns parents develop in the UK.

The unrepresentativeness of the sample maybe explained through the recruitment strategy and the inherent biases in online survey research. Recruitment was carried out through social media posting by the research team, charities, and parent carer forums. Unlike the research team and the charities, the parent carer forums posted onto private social media pages and, in some cases, emailed the survey to their members as part of a newsletter, making the parent carer forums gatekeepers. Gatekeepers are defined as individuals who control access to a privately controlled space, such as an institute or a forum (Singh, 2016). In these spaces the gatekeeper's permission is required for research to occur. Research has shown gatekeepers to skew samples as they may decide to inform everyone in that space about the research or only inform a select few (Lamprianou, 2022). Additionally, gatekeepers may also limit who is able to access the space based on certain criteria (Singh, 2016). Although parent carer forums are supposed to be for parents and carers of children with special needs and/or disabilities, they may have criteria that stop some individuals from joining. Because I am unable to see who the parent carer forums shared the recruitment link with, I cannot be sure that they did not decide to exclude anyone who may have been eligible.

Online survey research can result in several biases in the sample and the data: self-selection, response, and recall. Self-selection bias describes when participants can choose if they take part in a study and the final sample is demographically different to the population. Studies into self-selection bias have identified that those who are more likely to take part in online surveys have a greater involvement in the topic of the study, such as a greater interest or concern about the topic or the results (Cranford *et al.*, 2008; Mayr *et al.*, 2012; Khazaal *et al.*, 2014). This leads to changes in the findings, for example Cranford *et al.* (2008) invited a random sample of 2502 undergraduate students to take part in an online survey on alcohol use. Of the non-responders, 221 were followed up via telephone survey to complete an abridged version of the online survey. Cranford *et al.* (2008) identified that the non-responders drank significantly less frequently than responders, even when demographic differences between the responders and non-responders were controlled for. As such, it is likely that the sample in this study represents those more interested in earlier CP identification, however the impact of the sample on the results is unknown.

Response bias describes the factors that influence participants to respond inaccurately or falsely to question (Furnham, 1986). Demand characteristics and social desirability bias are two forms of response bias. Demand characteristics describe how a participant could be alerted to the goals of the study and change their responses to meet the goals. Similarly, social desirability bias describes when a participant changes their responses to appear more in line with social norms or with expectations (Van de Mortel, 2008). Participants were made aware that the survey was trying to identify the earliest signs of CP and therefore may have included concerns they themselves did not have or only presented concerns that fitted with the known signs of CP to fit with expectations.

Recall bias is characterised by the accuracy in which participant recall information (Infante-Rivard and Jacques, 2000). It can be caused by under- and over- reporting by participants resulting in distorted data. In this study, the caregivers were asked to retrospectively share their earliest concerns, which were not checked against NHS records. Because of this, it is likely that caregivers may have omitted concerns that they had developed over time, they may have also mixed their own concerns with those shared with them by HCPs, as seen in the 'concerning medical history' theme. Therefore, due to potential self-section bias, response bias, and recall bias, the results may not be an accurate representation of the concerns developed and reported by caregivers or the timeline in which these events occurred.

Secondly, the opinions and experiences of GPs have not been included in the survey. GPs have a gatekeeping role, as explained in Chapter 1, and therefore they have to make decisions based on the information presented to them and on their knowledge of relevant conditions. Understanding their opinions, experiences, and knowledge of CP would have highlighted the key signs they use to identify infants with CP and how often caregivers of typically developing children report similar concerns. Furthermore, they may have been able to elaborate on the strategies they use to prevent over-referral of infants who turn out to have typical development, improving cost, time, and resource efficiency. Notably, in 2018, Action CP reported low levels of GP and Health Visitor training around identifying CP. In particular they identified only 24 (of 147) local authorities that provided training, and 75 (out of 186) NHS trusts that failed to answer if they provided training and an additional 16 trusts stated that they did not provide any training on identification of CP. This suggests that some

PHCPs may not have the knowledge to be able to identify early signs of CP and that further UK wide training is needed. It should be noted that online training packages that could be implemented do exist, such as the Training in Early Detection for Early Intervention (TEDEI) course which provides training in how to detect early atypical motor behaviour in infants aged 0-6 months old (Officer *et al.*, 2021). Further research is needed around the specific concerns raised in primary care by caregivers, how PHCPs handle these concerns, and, importantly, how often the concerns raised in this survey are also raised by caregivers of typically developing children.

2.5.10 Conclusion

Overall, the results demonstrate that although caregivers identify the same concerns that clinicians who specialise in CP report to be key, delays still occur. One potential reason for the delays is the non-specific nature of the concerns, leading a GP to be unsure if the condition is self-limiting, requiring a watch and wait approach. Other reasons are that GPs may be relying on their contextual knowledge of the patient to guide them in how seriously they are to take the parents' concerns at this point, GPs wanting to observe specific symptoms first hand, as well as a lack of training on the symptoms of CP and the importance of early referral. Further research is needed to identify if non-specific feature of the presentation and/or utilisation thresholds are resulting in delays to CP referral. The next chapter will assess caregivers' responses around the primary care referral experience. The aim of this analysis is to identify what delays are occurring and to highlight potential opportunities for intervention.

Chapter 3. 'You are navigating the ocean alone in a reed boat with no map or oars.' Parental experiences of accessing primary care referral for their infants with Cerebral Palsy.

3.1 Introduction

This chapter examines the causes of delays within primary care referral for infants with Cerebral Palsy (CP) and how the delays influence the pathway through primary care taken by the caregiver and infant. Using the Andersen Model of Total Patient Delay (TPD) (Walter *et al*., 2012), survey data describing parental experiences of the primary care referral process was mapped (Chapter 2), describing various types of delay. Centrally, the most frequent type of delay, diagnosis delays, occur in primary and secondary care, where caregivers' concerns are not recognised, an alternate diagnosis is given, or a 'watch and wait' approach is taken. The chapter then discusses the three core determinants of the referral process; acknowledgment of parental concerns, HCP's awareness of CP, and problems with the referral itself. Notably, caregivers reported having to repeatedly attend primary care services prior to receiving a referral. The patterns of delays reflects reports for other paediatric and adult conditions, suggesting that underlying factors are influencing the referral process, such as awareness of symptoms, and the patient's self-perceived eligibility for medical care. These underlying factors align with the analytic concept of 'Candidacy', a seven-stage dynamic process in which patients and HCPs negotiate the patient's eligibility for medical care (Dixon-Woods, *et al*., 2006).

Understanding the causes of delay occurring in primary care can help to reduce the delays occurring within primary care referral to therapy and diagnosis. The survey showed that infants whose CP symptoms are first identified in primary care are more likely to have a milder CP severity, and experience, on average, a 6 month delay compared to their counterparts identified in secondary care (Chapter 2). Furthermore, the earlier in the infant's life the caregivers raised their concerns the more likely they would experience delays to referral. This occurred despite few significant differences in the concerns caregivers raised between those who received a delayed referral or an immediate referral. As such, more research is needed to understand why these delays are occurring.

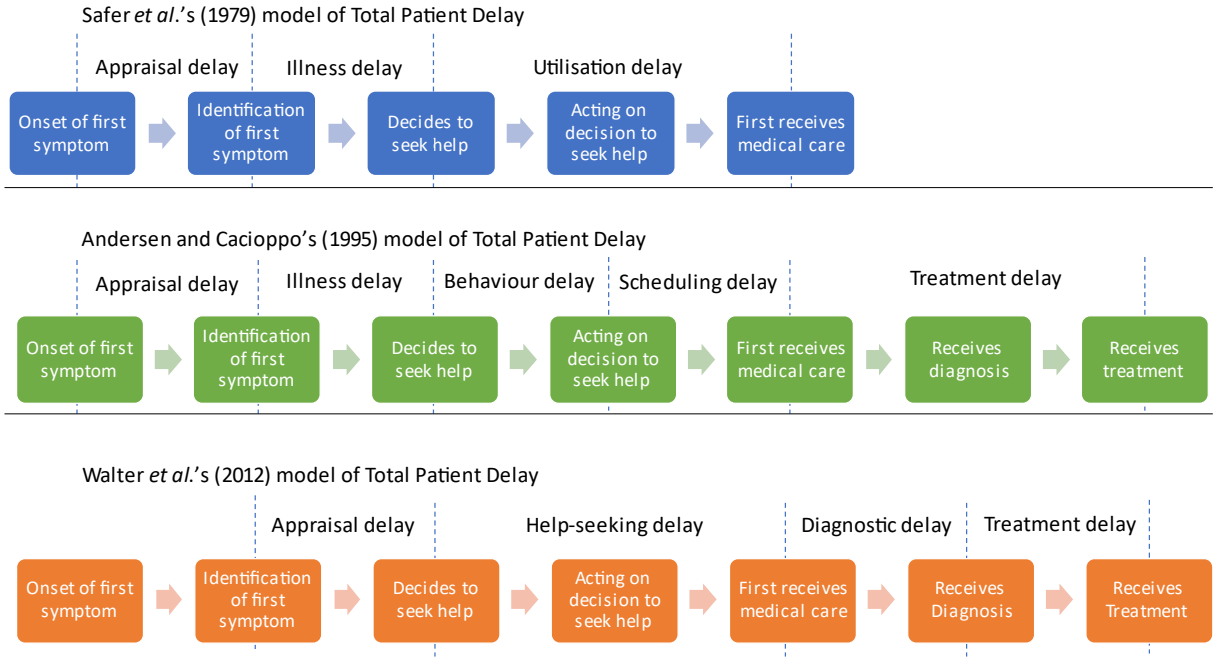


Figure 3 The three models for evaluating Total patient delay

Exploration of the caregivers' experience of primary care may identify the factors resulting in delays. Models of TPD look to identify the reason for delays and the length of the delays occurring between the first bodily change and the beginning of treatment through the use of qualitative and quantitative analysis. The most widely used model of TPD was first published by Safer *et al.* (1979) to identify delays in cancer diagnosis and treatment. Safer's original model comprised of three stages of delay, as shown in Figure 3; appraisal, the time taken for the patient to identify a symptom as a sign of illness; illness, the time the patient takes from deciding their ill to deciding to speak to a medical professional; and utilisation, the time taken between the patient deciding to seek care and their first health-care appointment. Andersen and Cacioppo (1995) further developed the Safer *et al.* (1979) model to comprise of five stages of delay, in which Safer *et al.*'s (1979) utilisation delay was split into; behaviour delay, describing the time between deciding to seek medical care and acting on this decision; and scheduling delay, the time between acting on the decision and receiving medical care, as shown in Figure 3. Andersen and Cacioppo (1995) also added a 'treatment delay' to describe the time taken from first receiving medical care to receiving treatment. The final development of the model, by Walter *et al.* (2012), redefined Andersen's model into four stages of delay, as shown in Figure 3: Appraisal delay, consisting of delays occurring between the patient detecting a bodily change and deciding to seek out medical care. Help-

seeking delays, consisting of delays occurring between deciding to seek-medical attention and attending the first consultation with a health care professional. Diagnostic delays, consisting of delays between the first consultation with a health care professional and the individual receiving a diagnosis. The final delay type is Pre-treatment delay, which consists of delays between receiving a diagnosis and the start of treatment. Although all three models were originally developed for identifying delays in adult cancer referral. The Andersen model has since been used successfully to assess delays in a range of paediatric conditions such as Cancer (Dixon-Woods *et al.*, 2001), Diabetes (Usher-Smith, Thompson and Walter, 2013; Rohilla *et al.*, 2021) and Multiple Sclerosis (Hinton and Kirk, 2015).

This chapter will explore the caregiver experience of the primary care referral system when their infant is not identified as at risk for CP at around the time of birth, but in later months within the community.

3.2 Methods

As described in Chapter 2, the data was collected using an online survey of parents and caregivers of children diagnosed with CP (n=255) between 5/6/2019 to 15/11/2019. The survey was made up of 9 sections; welcome letter, information sheet, consent form, child's demographics, earliest concerns, report of earliest concerns, experiences of the referral and diagnosis process, caregiver's demographics, and debrief, respectively. In particular earliest concerns, and report of earliest concerns, included free text questions asking participants about what their earliest concerns were, what they were doing when they first became concerned, and what happened if they did not receive an immediate referral after raising their concerns to a Health Care Professional (HCP). The experiences of the referral and diagnosis process also used free text questions to ask participants what they felt was good, what could have been improved about the service they experienced, and what they would change to improve the service they experienced. It was from these questions the qualitative analysis was carried out.

The qualitative data was mapped against the Andersen Model of Total Patient Delay (Andersen and Cacioppo, 1995; Walter *et al.*, 2012). The data was coded using the Andersen Model definitions from Walter *et al.* (2012) in a realist, deductive, semantic approach (Braun

and Clarke, 2006). Due to a deductive approach being taken, both researchers (JB and CS[3]) read through the survey responses and the delay category definitions reported by Walter *et al.* (2012). They then agreed on the interpretations of Andersen Model category definitions before beginning coding. After coding a quarter of the survey responses JB and CS compared their coding to check that both were continuing to interpret the definitions in the same way. Inter-rater agreement was assessed using Cohen's Kappa.

A second thematic analysis of the same data was also carried out to identify the determinants of the referral process as described by caregivers of children with CP. This was carried out by reading and re-reading the survey responses to identify potential themes. Potential themes were presented to AB and TR for review before the final themes were written up. The data was coded using a realist, inductive, semantic approach (Braun and Clarke, 2006) by JB only.

Pathways through the referral system reported by caregivers were plotted using Lucidchart, an online collaboration software tool (https://lucid.co/product/lucidchart, 2020).

3.3 Results - Andersen model of Total Patient Delay

Across the data, caregivers reported different forms of delays in the CP referral and diagnosis process. *Appraisal delays* occur between the development of the first symptom and the decision to seek medical help. We saw a little of this in the data, with some caregivers being unaware that their infants' symptoms were not typical. *Help-seeking* delays follow appraisal delays and encompass the time between the caregiver deciding to seek medical help and their first appointment with a HCP. Help-seeking delays occurred regularly, with delays focusing around the caregiver waiting for their next scheduled appointment or due to the caregiver needing time to build their confidence in their concerns. When caregivers did meet with their HCP they could experience delays to diagnosis, *diagnosis delays*. These were the most frequent delays in the data, revolving around factors occurring in primary and secondary care. The final delay, *treatment delay*, is the time between receiving the diagnosis and starting treatment. No reasons for treatment delays were

[3] CS - Charlotte Sieboth. At the time of the analysis Charlotte was a undergraduate psychology student at Newcastle University on a placement year working with Dr Lindsay Pennington.

identified in this data set. The following section of the chapter will explore the different reasons for the delays by following the timeline in which delays occur.

3.3.1 Initial Concerns: Appraisal Delays

Even though they identified their infant's initial symptoms, caregivers reported not seeking out medical attention straight away due to being 'unaware' that the symptom was not typical or because their infant seemed 'fine'. Others thought they were 'imagining' the symptoms their infant was presenting, as outlined in the following quote:

> At about 3-4 weeks we noticed a hand preference with movement. Being medical parents we downplayed this (!). By 8 weeks we were convinced. My Gran made a comment that he was "going to be a leftie" which made us realise that we were not imagining things. (F075)

In this case, a potential observation around 'hand preference' is initially downplayed by parents trying to compensate for their professional expertise and then confirmed over time through another observation matching their own. One caregiver also reported a missed opportunity. A missed opportunity for referral occurred when both the caregiver and HCPs failed to recognise the infant's symptoms. This infant did not receive referral until a teacher recognised the infant's symptoms. The infant's mother explained that he was her first child so she did not know that he was falling behind on development and the Health Visitor also did not notice anything unusual in his development. Another participant (M167) reported her earliest concerns to be that her son was very stiff and needed help at age 3 years to play on climbing frames. By 5 years of age he was unable to jump and 'constantly dribbled'. They (M167) reported that the first concerns raised to a HCP was that her son was unable to hold a pencil, unable to write, unable to balance, and was unable to judge depth at age 7 years. This infant was 6+ years old when they received a diagnosis of mild (GMFCS II) Quadriplegia.

3.3.2 Asking for Advice: Health-Seeking Delays

Although some caregivers developed concerns, they decided to wait until their next scheduled appointment with a HCP to raise them. This only occurred in infants who were receiving clinical follow up. Others felt they needed to build their confidence in their concerns before approaching a HCP. They described collecting more evidence before going to their GP. They built their confidence either through talking to others, looking their concerns up on the internet, or by spending time further observing their infant's symptoms.

They described doing these things due to self-doubt making them think 'it would be nothing and [that they would be] wasting an appointment.' (M033). For one caregiver the need to build confidence was due to the fear of being labelled as paranoid:

> I remember pacing around the house holding the phone, dreading calling the GP for another appointment, for fear of being labelled the paranoid first time mother (despite being a midwife), but also knowing that something wasn't right and I HAD to call. (M039)

For some of these caregivers the delay came after previously having their concerns dismissed by friends, family, or HCPs, leading them to doubt their own observations.

3.3.3 Reasons for not referring: Diagnosis Delays

After having booked their appointment to see a GP, caregivers described delays occurring within primary care and secondary care that delayed their diagnosis. Upon meeting with a GP caregivers were met with one of three reasons for delay within primary care. The first occurred due to caregivers' concerns being 'brushed off' by their GP or Health Visitor, or by the HCPs not sharing the caregivers' concerns. The second was due to the HCP offering an alternative reason for their infants' symptoms, such as hypermobility or late development. The third was due to HCPs choosing to 'watch and wait' for 2-3 months to see how the infant developed.

Often caregivers reported that they repeatedly approached the same or different HCPs while seeking their infants' diagnosis. These caregivers highlighted that they began to 'loop' through the primary care system. Plotting of these pathways demonstrates loops occurring within the system (see Figure 4). Some of these caregivers described going through these loops up to five times before a referral was given.

Once the infant and caregiver had been referred to secondary care they could also be faced with further delays. Unlike the previous delays, delays in secondary care were seldom reported by more than one or two caregivers. At first some had their GP referral rejected by their local hospital (n=1), or their appointments being postponed for 'almost 6 months' (M110; n=1). When some caregivers spoke to the secondary care HCP treating their infant, they felt their concerns were ignored (n=2) or were initially refused requests to be referred to a specialist or for an MRI (n=5). Additionally, for some the MRI report was falsely reassuring (n=6), only for a second MRI carried out 'privately at 3 yrs old [to show] brain

damage consistent with greater than 20mins lack of oxygen at birth' (M010). Some infants suffered from comorbidities, such as epilepsy, that required treatment before investigation into the CP diagnosis could begin (n=1). Others were given a misdiagnosis (Erbs palsy, n=1), or were bounced 'around the system' between different hospitals and different specialists (n=2). One parent felt that this was due to no cohesive communication between HCPs or between NHS trusts. For infants who were under clinical follow up after being identified as high risk at birth, some were reported to be discharged before a diagnosis was made despite still demonstrating symptoms (n=3).

3.3.3 Waiting for therapy to begin: pre-treatment delay

No delays to treatment were clearly described by the participants. Some caregivers described experiencing referral delays; however, it was not clear if these delays were for diagnosis or treatment.

3.4 Determinants of referral experience

Three determinants of referral were identified that resulted in caregivers experiencing either an immediate or delayed referral. They consist of acknowledgement of concerns, CP awareness, and problems with the referral itself.

3.4.1 Acknowledgement of concerns.

Caregivers who received immediate or delayed referral reported on whether the GP had acknowledged their concerns. Those who received immediate referral reported either feeling grateful their concerns were listened to, taken seriously, and were asked encouraging questions, or that despite the GP not sharing their concerns, the GP still referred them on. In contrast the delayed group reported that their concerns were met with either initial reassurance from primary HCP that there was 'no problem', that the infant needed to be potty trained before a referral could be made, or a 'watch and wait' approach. This was previously identified as diagnosis delays by the Andersen model and resulted in caregivers looping through the referral pathway at the GP interactions level, Figure 4.

Caregivers whose concerns were met with reassurance felt that their concerns were brushed off as they reported being told that their infant 'just needs time' to catch up, that they were 'seeing things', or that an early hand preference or not using one hand as well as the other was just 'early development'. They felt that they were given 'false' reassurance due to the

HCP not having the knowledge to be able to recognise CP, resulting in these caregivers also reporting that they felt they had to fight for a referral. The HCPs' actions in these cases also took a toll on some of the caregiver's mental health, making caregivers believe they were paranoid, overprotective, neurotic, or bad parents. These beliefs were identified as reasons for delay in help-seeking delays. Some reported they were given incorrect information, such as the 'physio would not see her until [the infant was] out of nappies' (M218). Caregivers who reported a 'watch and wait' approach reported being told that their infant may just need more time, but to report back to the HCP after a couple of weeks to months to see if anything had changed. Regardless of what these caregivers were told they reported feeling that they either had to wait for referral or fight for referral by continually seeing the same HCPs or visiting different HCPs. Many of these caregivers described wanting their concerns to be acknowledged and encouraged and suggested that HCPs listen more to caregiver's concerns and 'acknowledge that a parent knows their child best' (M002). Overall, these occurred at the GP and Health Visitor interaction levels of the referral process, resulting in the caregivers looping through the process (see Figure 4).

3.4.2 Awareness of CP

Those who received immediate referral praised their HCPs for early recognition, while those who received delayed referral noted a lack of HCP CP awareness. Caregivers explained how reporting of specific CP symptoms to their HCPs, such as convulsions or fisting did not result in referral. Similarly, the infant's medical history, or lack of history, was reported to be used as a reason not to immediately refer, or for caregiver concerns to not be 'taken seriously'.

Caregivers who noted a lack of CP awareness suggested that primary HCPs, Health Visitors in particular, should be given further training to make them more aware of the signs of CP and how to test for CP. Some also felt that all HCPs would also benefit from having further training on the early warning signs of CP, on CP in general, and on the 'less obvious categories of CP' (M056). One caregiver also highlighted that HCPs need to be aware of the different types of services, such as the different types of therapy services, so that infants are referred to the services best for treating their needs. The lack of CP awareness resulted in primary care diagnostic delays. Overall, caregivers who reported experiencing this also described looping through the primary care pathways, shown in Figure 4, up to 5 times before a referral was made.

3.4.3 Problems with the referral itself

Even when referral was made, caregivers did not always receive quick and appropriate referrals from primary care. Some reported quick referrals but others reported delays, however it was not always clear if caregivers were describing referral for diagnosis or for therapy. Some caregivers felt that the referral and diagnosis process was drawn out and should be improved. They felt that the 'watch and wait' approach is not the right approach, and instead that GPs should either carry out basic tests to determine if a referral for diagnosis or therapy is needed, be prepared to refer these infants on immediately for diagnosis or therapy or inform caregivers that they can self-refer to physiotherapy.

Some infants were referred to departments that were not able to provide treatment for the infants' symptom(s) before the CP diagnosis was given. This resulted in the infant being discharged or receiving an internal referral to paediatrics or physiotherapy. One caregiver also described how their initial 'referral was rejected by Hospital 3' (M053) resulting in their GP referring them to another department. Some suggested that these infants should be referred to specialist paediatricians, such as paediatric neurologists, rather than general paediatricians in addition to being referred to physiotherapy for early intervention at the same time. Another suggestion was to allow caregivers the ability to self-refer to paediatricians or physiotherapists regardless of the GP's agreement.

Finally, some caregivers felt that scanning infants as soon as possible after a concern is raised would also help speed up the process. They pointed out that their child's diagnosis relied on a 'positive' MRI and for some, if they had not fought, their infant would have been 3 years old by the time an MRI was made available to them. Overall, the referral itself resulted in caregivers going between primary and secondary care until a referral to the correct department was given or caused delay within secondary care to diagnosis.

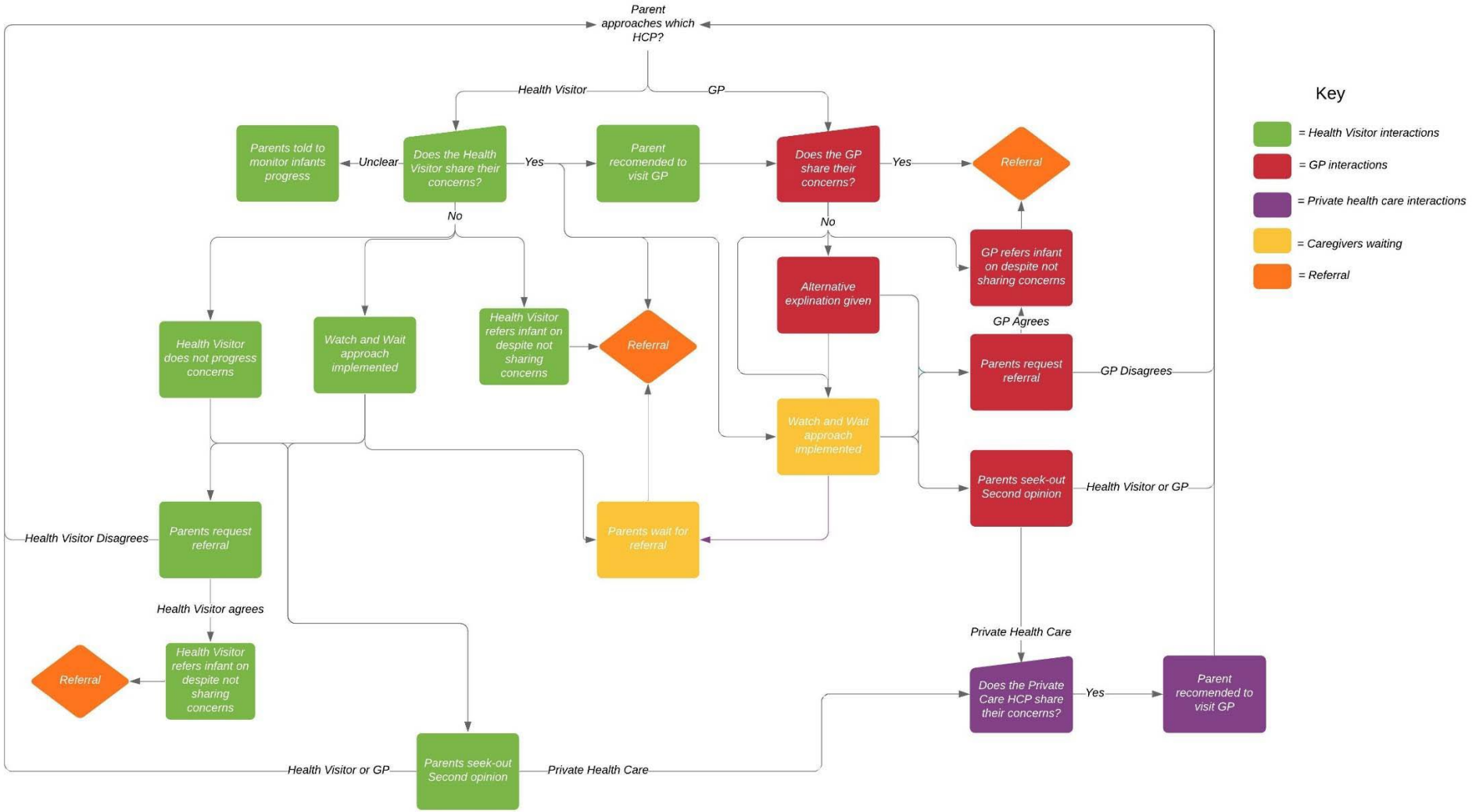


Figure 4 The pathways through primary care experienced by parents and caregivers when raising their concerns about their infants motor development

3.5 Discussion

3.5.1 Findings - Delays described by the Andersen model

The results of this study demonstrate multiple delays occurring between the initial first symptom developing and diagnosis, and three determinants of immediate or delayed referral. Appraisal delays consisted of caregivers being unaware of a symptoms meaning until pointed out by another. Help seeking delays consisted of caregivers researching and building evidence to take to their GP or by caregivers deciding to wait until their next appointment to raise their concern. Diagnosis delays within primary care consisted of alternative diagnoses, watch and wait approaches, initially giving caregivers reassurance or telling caregivers they had no reason to worry. This supports the theory presented in Chapter 2 that the non-specific nature of the concerns may be resulting in delays. In contrast, diagnosis delays in secondary care consisted of administrative issues, HCPs delaying internal referrals, misdiagnoses, and comorbidities. Although no pre-treatment delays were reported, the other findings align with those reported in the literature around other paediatric and adult conditions such as Cancer (Dixon-Woods *et al.*, 2001; Molassiotis *et al.*, 2010; Walton *et al.*, 2013; Clarke *et al.*, 2014; Parsonage *et al.*, 2017), Diabetes (Usher-Smith, Thompson and Walter, 2013), and Multiple Sclerosis (Hinton and Kirk, 2015).

There are two potential reasons why no pre-treatment delays were identified. The first is that the data was not collected with the intention to undergo Andersen model analysis. As such the caregivers were never prompted to talk about pre-treatment delays, which may have resulted in the data being unclear in terms of where the delays were occurring. The second is that the Andersen model criteria for pre-treatment delays may not fit the data produced from CP care. When infants are referred to secondary care for suspected CP, the National institute for Health and Care Excellence (NICE) guidelines suggest that the infant be referred to a multidisciplinary team (NICE, 2017a). As such, these infants may have begun to receive treatment, such as physiotherapy, before a diagnosis of CP was given, and therefore pre-treatment delays may be accounted for within diagnosis delay. However, without further research, it is unclear as to why no pre-treatment delays were identified.

3.5.2 Findings - Determinants of the referral experience

The determinants of the referral process - acknowledgement of concerns, CP awareness and problems with the referral itself - influenced if an infant received an immediate or delayed

referral. Infants who received a delayed referral had multiple primary care appointments before a referral was given, resulting in them entering a looping path. Across the determinants, caregivers suggested that increased proactive testing, training for HCPs, and guidance in which concerns should prompt referral is needed.

3.5.3 Primary care training and guidance around early CP

Training and guidance are two components of the CP referral process that have been previously identified as needing improvement. In 2016 and 2018, Action CP submitted Freedom of Information requests (FOIs) to UK Local Authorities (LAs), Clinical Commissioning Groups (CCGs), and NHS trusts about their CP service provision. Within the FOIs questions included HCP training, care pathways, and service frameworks. In 2018, CP specific training for primary HCPs was most frequently reported as generic disability training. Similarly, training frequency was reported as either biannually, sporadically or only upon request, with one NHS trust reporting the last CP training session to occur in 2015. Out of the 56 responding NHS trusts, only 19 NHS trusts reported having or developing care pathways in line with NICE (2017) guidelines. The other 37 NHS trusts reported no specific formal pathway for CP. These figures demonstrate the training provided to HCPs around CP and the provision of formal CP care pathways to be poor, despite NICE (2017) guidelines being published.

The lack of consistency in training and referral pathways, along with the lack of testing may be due to limitations with the NICE (2017) guidelines. Although the guidelines provide key information they do not provide guidance for use in primary care. For example, when looking for signs of CP, HCPs are advised to consider using Prechtl's General movements assessment (GMs) (Einspieler *et al.*, 2004). However the GMs requires undergoing a training course lasting 4 days, consistent practice of the GMs is needed for accuracy, and can only be used within the first 4-5 months of life. However, as shown earlier, caregivers typically do not begin to report concerns until after 3 months of age, often meaning the GMs cannot be used once a concern is reported. Similarly, the GMs has been shown to have poor psychometric properties when used in the general population and in low-risk infants (Bouwstra *et al.*, 2010; Bennema *et al.*, 2016), meaning the GMs should not be used for general screening. No other screening tools or measures are advised in the NICE (2017a) guidelines, likely explaining part of the lack of thorough testing in primary care. Similarly, the

guidelines identify the 'possible early motor features' of CP as Atypical movements, Atypical tone, Delayed motor development, Feeding difficulties, and Early asymmetrical hand preference. However, this list of symptoms does not demonstrate the range of symptoms infants with CP may exhibit, as shown in Chapter 2. Furthermore, when these features are identified, the guidelines only provide advice to refer to secondary care if the infant is identified as at increased risk due to risk factors at around the time of birth. However, as discussed in Chapter 1, 40-50% of infants do not have identifiable risk factors. As a result, infants without identifiable risk factors and/or those with less common symptoms may not receive an early referral. Therefore, the NICE (2017) guidelines do not go far enough to support identification of CP within primary care.

3.5.4 Previously made suggestions on how to improve CP identification in the community

Suggestions on how to improve UK primary care referral for CP have already been made. In 2014, a Parliamentary Enquiry (Action CP, 2014) looked at CP provision within the UK and provided recommendations on how to improve early identification of CP. The Enquiry recommended 1) greater emphasis on parental concerns, 2) commitment to rapid referral and elimination of watch and wait approaches, 3) more widespread use of GMs, and 4) improving awareness of CP among GPs and Health Visitors. However, apart from the use of GMs, these recommendations were not included in the NICE (2017) guidelines. Shortly after, Richardson (2018), CEO of Action CP, underwent a fellowship to observe the CP services currently provided by the CP Alliance in Australia. Richardson's (2018) report provided two suggestions of approaches which could be implemented in the UK.

- The first approach suggested was a CP register. The Australian CP register provides a list of infants identified at risk of CP in hospital and within the community, and enters these infants into an adjoining screening program, CP Check-Up, described below. The CP register includes a community advisory team who provide support to primary HCPs with identification of infants within the community.

- The second approach was the implementation of screening programs. Richardson (2018) described 4 overlapping screening programs. 1) Neonates, for 0-3 month high risk infants. 2) 3 month assessment, consisting of the GMs, the Hammersmith Infant Neurological Assessment and the Bayley Scales of Infant and Toddler Development assessments. 3) Early Detection and Diagnosis clinics, a follow up service for those

attending neonates as well as accepting referrals from parents, GPs, community therapists and paediatricians. 4) CP check-up, a comprehensive and holistic surveillance program for infants at risk, or diagnosed with, CP. Infants in this program receive assessments every six months between birth and 6 years, and yearly appointments after 6 years.

Together, the Australian CP register and CP screening programs resulted in around 50% of infants attending the CP Alliance clinics being diagnosed within the first year of life, 75% by their second year, and 90% by their third. This is a slight improvement on the UK diagnosing 34% in their first year, 69% by their second year and 87% by their third, as reported in Chapter 2.

In relation to all types of referrals in primary care, Greenwood-Lee *et al.* (2018) identified the problems and solutions to primary care referral across all conditions through a narrative review. They identified that referral guidelines and education programmes generally serve as the foundation for interventions, as on their own they may be ineffective. Greenwood-Lee *et al.* (2018) suggest that guideline and educational interventions should be built on by incorporating communication with secondary care specialists, such as: referral reply letters from SHCPs; relationship building and collaboration on care practices between PHCPs and SHCPs; peer review and/or supported patient assessment implemented through primary triage clinics within secondary care; and peer review groups between PHCPs with consultant engagement. Alternatively, Greenwood-Lee *et al.* (2018) also suggested the implementation of standardised referral forms, checklists, scoring systems, and assessment tools specifically designed to be used within primary care to help improve referral quality and decrease delayed referrals and unnecessary referrals. These suggestions are supported by Blank *et al.* (2014)'s systematic review of problems and solutions in primary care referral. Although Greenwood-Lee *et al.* (2018) and Blank *et al.* (2014) do not directly support Richardson's (2018) proposal, they do agree that a broader, richer, referral infrastructure is needed, which increases the level of skills within primary care.

However, CP registers and UK screening programs have previously been available in the UK. In the 2014 Parliamentary Enquiry (Action CP, 2014), the UK charity SCOPE provided evidence of an advisory assessment service (AAS) they had previously provided in London that was accessible to families across the UK. The AAS gave parents access to a 2-3 day

assessment carried out by a multi-disciplinary team of professionals. The team would provide the parents with a detailed report of the infant's specific needs and provide signposting to appropriate follow-on services. The service was offered as evidence as a way to improve CP identification, however required funding to be able to restart. Since the enquiry, this service has not been restarted. Similarly, the provision of a CP register was also recommended in the 2014 Parliamentary Enquiry and again in the Action CP (2018) report. However, at the time of writing, a UK wide CP register has not been created despite registers already existing in Northern Ireland and Scotland. Thus, despite suggestions being put forward on how to improve the UK CP screening program, none of these suggestions have been put into action.

3.5.5 Screening tools, an alternative approach?

One way to improve primary care referrals that targets training, guidance and testing is through screening tools. Screening tools are defined as checklists or questionnaires that can be used by HCPs to identify infants with developmental delays. Multiple systematic reviews have demonstrated which screening tools have good psychometrics for detecting various conditions (Villeneuve *et al.*, 2013; Wallace *et al.*, 2015; Thabrew *et al.*, 2017; Marlow, Servili and Tomlinson, 2019; Sim *et al.*, 2019). Tools such as GMs, and the Hammersmith Infant Neurological Examination (HINE) (Haataja *et al.*, 1999) were recommended by Novak *et al.* (2017a) for early identification of infants with CP due to their excellent accuracy. Although it would be possible to perform the HINE within a GP appointment, like the GMs the HINE requires training to use, which may not be accessible to primary HCPs. However, other screening tools, such as the Ages and Stages-3 questionnaire (Squires *et al.*, 2009), have been designed with the parent answering the questions, removing the need for HCP training to use it. Each of these tools could be used to support primary HCPs decision-making on if an infant requires referral to secondary care. An intervention based on the introduction of a screening tool into primary care could target all three determinants.

It should be noted that the analysis took an inductive approach, meaning that the themes were developed from only the data provided by caregivers. As such the systems and structures that HCPs are required to work within are not described within the data. However, understanding of these systems and structures may provide alternative explanations for the diagnosis delays described.

Currently NHS referral systems are currently overwhelmed and referral of all infants with signs of CP would not be manageable. In February 2023 alone, there were 378,746 more referrals made across England than there were available appointment slots (NHS Digital e-RS team, 2023). Because of the excessive demand, Clinical Commissioning Groups (CCGs) (now Integrated Care Systems) tried to reduce the number of referrals made from primary to secondary care (PULSE, 2015; Baird *et al.*, 2016; PULSE, 2019). Some of the changes CCGs tried to make included increasing condition management duties to GPs and cash incentives for reducing referral rates (PULSE, 2015; Baird *et al.*, 2016; PULSE, 2019). Changes in referral systems have also resulted in GPs feeling less able to access support from secondary HCPs, and consultants being less likely to refer to another consultant, resulting in patients being 'bounced back' to GPs, only for GPs to have to refer patients on a second time (Baird *et al.*, 2016).

On top of this, the signs of CP overlap with other conditions and with typical development. For example, in Chapter 2 caregivers of infants with CP identified problematic feeding as being one of the earliest concerns they developed. However, meta-analysis has demonstrated around 43% of typically developing 0–5-month-old infants to also have problematic feeding (Pados *et al.*, 2021). Therefore, GPs are required to determine if the condition is self-limiting, can be managed within primary care, or requires specialist treatment. Depending on the caregiver's report, GPs may lean towards the infant having a self-limiting condition and reassuring the caregiver. In turn, once the family receive the CP diagnosis for a secondary HCP, they may feel that the GPs gave them an alternative diagnosis and false reassurance. Similarly watch and wait approaches are likely being used to help determine if the concerns are of self-limiting conditions or if the child requires a referral, helping to avoid any unnecessary referrals. However, without including the opinions of GPs, the reasons for these approaches can only be speculated on.

3.5.6 Candidacy may explain the underlying factors influencing the referral process

The similarity between the determinants of delay found in this study, and those reported in other conditions suggests there are underlying factors influencing the referral process from primary care regardless of the condition presented. One concept that describes these underlying factors is candidacy. In 2005, Dixon-Woods *et al*., conducted a critical interpretive

synthesis review of healthcare access in vulnerable groups. Candidacy is used to describe how individuals assess their eligibility for medical attention and how they legitimise their interaction and engagement with services. For example, the first stage of candidacy 'Identification of candidacy' describes how individuals identify if they need medical attention, or that they are a candidate for medical attention. Some individuals can be quick to identify their candidacy and seek out medical attention straight away. Others with the same signs may downplay their signs and only identify their candidacy for medical attention when they can no longer manage their symptoms on their own. Candidacy is a dynamic process, being consistently redefined by the patient and their HCPs with seven overlapping stages, described in Table 8.

Overall, six aspects of candidacy can be identified within the data presented. The first, identification of candidacy is seen within the appraisal and help-seeking delays. Caregivers reported being unaware that their infant's symptoms were not typical, or that they needed to develop their evidence base due to fears of having their concerns dismissed or being labelled by HCPs. We see this in other contexts too, such as paediatric arthritis, cancer, and diabetes, where parents of children and young people attribute their child's symptoms to everyday things, such as accidents and self-limiting conditions (Dixon-Woods *et al.*, 2001; Usher-Smith, Thompson and Walter, 2013; Clarke *et al.*, 2014; Kirkpatrick *et al.*, 2018; Pedersen *et al.*, 2020) and they look for advice from family and friends, or from the internet before seeking medical care (Usher-Smith, Thompson and Walter, 2013; Clarke *et al.*, 2014; Pedersen *et al.*, 2020). Furthermore parents report postponing seeking health care due to fears of wasting the GP's time and looking like a fool for requesting multiple appointments with their GP (Usher-Smith, Thompson and Walter, 2013); Pedersen *et al.* (2020).

The second, navigation of services, was demonstrated through caregivers and their infants being referred to departments who could not provide treatment, as described in *problems with the referral itself* determinant. In these situations it was the primary HCPs who failed to navigate the services correctly on behalf of the caregiver and infant. Furthermore, as caregivers became more knowledgeable about the requirements for diagnosis they began to request referrals for brain scans. We see similar findings in other contexts too. For example Kirkpatrick *et al.* (2018) identified an individual who felt their referral had 'spiralled on to different places' despite their mother identifying and suggesting her child had arthritis

Candidacy stage	*Description*	*Evidence*
One - identification of candidacy	The process in which an individual comes to recognise their symptoms as needing medical intervention.	Dixon-Woods *et al.* (2006) found that individuals from more deprived circumstances were more likely to manage their own health, and to see their own candidacy as a 'series of crises'. More disadvantaged communities were also more likely to downplay the importance of their symptoms due to normalisation of symptoms and the fear of being blamed by HCPs.
Two – navigation of services	An individual's knowledge of the services provided and understanding on how to make contact with and how to access services.	Dixon-Woods *et al.* (2005) highlighted that deprived communities are not always aware of the services available to them. However, even when they are aware of the services they are not always able to access them, due to issues such as transport and working hours.
Three - permeability of services	The ease in which an individual can access services. Permeability covers several potential barriers, such as the level of gatekeeping, the complexity of the referral process, and the cultural alignment of the services with the persons needs and values	Dixon-Woods *et al.* (2005) classified porous services as requiring fewer candidacy qualifications to access, for example Accident and Emergency, whereas low permeability services, such as referral, demand candidacy qualifications. Low permeable services often have high levels of non-attendance by disadvantaged individuals. This can be due to factors such as appointment systems requiring fixed addresses, or individuals feeling culturally misaligned from the values of the health services.

Candidacy stage	*Description*	*Evidence*
Four - appearance at services	The individual's ability to assert their candidacy for medical care. To make a claim, individuals need to be able to formulate and articulate their issues and to be presented credibly.	Dixon-Woods *et al.* (2005) identified these to be issues for individuals from lower incomes, as their middle-class counterparts may be more adept at explaining and demanding services.
Five - adjudication by HCPs	How an individual is judged by their HCPs, which subsequently influences their progression through the services and their access to care. Ultimately adjudication results in an individual being classified as being deserving or not deserving of care.	Dixon-Woods *et al.* (2006) highlighted that HCPs take into account their perception of the patient when deciding if their patent would do well from undergoing an intervention, leaving more deprived patients at a disadvantage.
Six - offers and resistance to services	How an individual may refuse offers at multiple stages of their journey, including resisting appointment offers, referral offers and treatment	Dixon-Woods *et al.* (2005) demonstrated that despite GPs identifying their patients candidacy and offering to refer the patient to services that could provide support, patients can and may choose to not be referred or given medication.

Candidacy stage	*Description*	*Evidence*
Seven - operating conditions and local production of candidacy	The factors at social and macro levels that influence candidacy	Factors identified by Dixon-Woods *et al.* (2005) included the availability of local resources for addressing candidacy, and relational aspects which develop between the healthcare provider and patient over multiple visits.

Table 8 The seven stages of Candidacy by Dixon-Woods et al. (2005 & 2006)

multiple times to HCPs. Dixon-Woods *et al.* (2001, p. 673) identified parents using 'private medicine, alternative medicine, accident and emergency departments, or visits to specialists about other problems' to subvert the system to get their children medical attention sooner. Similarly parents have been identified to talk to different GPs or go straight to hospital when the initial interactions with standard care did not meet their needs (Pedersen *et al.*, 2020).

The third, permeability of services, is shown through caregivers reporting being turned down for referral within diagnostic delay as well as the determinants of acknowledgement of concerns and CP awareness. As those rejected for immediate referral described HCPs not sharing or not legitimising their concerns, a high threshold for referral is shown. Again delayed referrals are seen in other paediatric conditions, with parents reporting watch and wait approaches being used in Diabetes (Usher-Smith, Thompson and Walter, 2013), or HCPs providing alternative explanations for their child's symptoms of arthritis (Kirkpatrick *et al.*, 2018) and leukaemia (Clarke *et al.*, 2014), such as the condition being self-limiting. Some parents also found themselves in disputes with their GPs over their infant's need for referral and/or experienced long delays to see specialists once a referral was made (Dixon-Woods *et al.*, 2001). As explained above, when parents identified the permeability of primary care services to be too low they attended other services such as private care (Dixon-Woods *et al.*, 2001). Although, this study did not find evidence of further factors, such as the patient having no fixed address that causes issues in permeability, this is likely due to the survey being online, which will have potentially prevented individuals from more deprived backgrounds taking part. Similarly in the paediatric literature factors such as fixed addresses were not described.

The fourth, appearance at services, is demonstrated within the help-seeking delays and within the concerns reported in Chapter 2. Some caregivers reported having to build an evidence base to support their concerns before they could report them. Similarly, despite most of the caregivers reporting their concerns either in context to how they discovered them or by using medical language, some caregivers reported not being able to describe what caused them concern, as shown in Chapter 2. This demonstrates that the ability to articulate concerns when appearing in services is affected in some caregivers. Again parents have reported seeking advice from family and friends, or from the internet before seeking

out medical care for diabetes (Usher-Smith, Thompson and Walter, 2013), and cancer (Clarke *et al.*, 2014; Pedersen *et al.*, 2020).

The fifth, adjudication by HCPs, is shown in the reasons given to caregivers as why they were not referred. This includes reasons such as telling the caregiver that the infant just needed time to catch up and therefore did not warrant treatment. Again we see these similar reasons given to parents when they seek help for their child's condition such as their child having a self-limiting condition that would pass with time (Dixon-Woods *et al.*, 2001; Clarke *et al.*, 2014; Kirkpatrick *et al.*, 2018; Pedersen *et al.*, 2020) These reasons also align with the theory that the non-specific nature of the caregiver's concerns may be resulting in delays due to HCPs determining if the infant's presentation is within normal limits or ruling out other potential conditions.

The sixth candidacy stage to be demonstrated in this study's data is operating conditions and local production of candidacy. Although this study did not assess for resource availability, delays due to secondary care appointments being postponed and caregivers choosing to undergo private MRIs to reduce waiting times, demonstrates a lack of available resources. However, in the literature parents report experiencing difficulties in getting appointments with GPs. For example, Usher-Smith, Thompson and Walter (2013) reported about one parent who waited 20 days for an appointment, which the parent felt was within the 'normal timescale' for an appointment.

One candidacy stage, 'offers and resistance', was not identified. This stage may not have been demonstrated due to the population taking part in the survey. Recruitment for the survey occurred online and was optional. This likely resulted in caregivers who resisted offers to services to also resist taking part in the survey or in the interviews carried out in the literature. Alternatively, it could be due to the infants in this study not being able to resist treatment. During Dixon-Woods *et al.* (2001) interviews of parents whose children were diagnosed with cancer, one parent reported having to convince their 8 year old child that they needed to seek medical care for the pain they were in.

3.5.7 Conclusion

Overall, the results demonstrate multiple delays occurring between initial onset of the infant's symptoms and the caregivers receiving their infant's diagnosis. Furthermore, the

immediateness of the referral was influenced by the HCP acknowledging the caregiver's concerns, the HCPs' CP awareness, and by any problems that occurred with the referral itself. However, these delays are not unique to CP and may be explained by the concept of candidacy. Although candidacy identifies the problem caregivers face, it does not identify the causal factors that could be manipulated through intervention. Therefore, further research is needed to determine the underlying factors causing delays in primary care referral.

The next stage of this thesis will look to determine if there is a currently available screening tool which could be implemented within primary care to reduce delays. Ultimately I will show that currently available screening tools often rely on developmental milestones, and no single screening tool covers all of the parental concerns identified in Chapter 2. Overall I will conclude that tools designed for completion by parents and caregivers should be further developed to ensure they identify all relevant concerns parents and caregivers could raise.

Chapter 4. How are items identified for inclusion in infant motor screening tools? A scoping review.

This chapter will examine how currently available motor screening tools were developed and if and how parental opinions were included within their development. Multiple systematic reviews have identified Prechtl's General Movements Assessment (GMs) as the gold standard for early Cerebral Palsy (CP) screening. However limitations with GMs, as noted in Chapter 1, prevent it from being used as a tool within primary care settings. Other tools have also been suggested for use within primary care, but their limitations, as explained in Chapter 1, prevent them from being an efficient resource for identifying infants with CP within the community early. Examining how the tools were developed and whether parents were included in the development may help identify a tool that meets the needs of identifying infants who may have CP within primary care.

This chapter will show that there are 42 tools currently available for screening infant motor development. However, only 5 include parents in their development and only 2 asked parents about what content to included. Additionally only 1 included parents of children with disabilities, the PEDI-CAT (Dumas *et al.*, 2010). Network analysis of the literature used by the screening tools to develop their items will demonstrate 36 of the included tools to have developed their items from the same literature sources.

4.1 Introduction

One way to improve detection of CP is through screening. Screening tools allow for a standardised and methodological examination of risk factors and/or symptoms to determine if further, more in-depth assessment is needed. Although screening tools do not identify if an individual has a specific condition, they can identify individuals who are at risk. Assessments of early motor development are frequently used to help guide clinicians in their decisions on the development of an infant. Many screening tools currently exist, each with their own purpose.

To identify if a screening tool is good, one must look at the sensitivity, specificity, and predictive values of the tool. Sensitivity describes the proportion of individuals with the condition that are correctly identified by the screening tool as having that condition (Trevethan, 2017). Specificity describes the proportion of individuals without the condition

that are correctly identified by the screening tool as not having the condition. Similarly, the positive predictive value describes the probability that those identified as positive for the condition on the screening tool do indeed have that condition. Conversely the negative predictive value describes the probability that those identified as negative for the condition on the screening tool do not have that condition. The higher each of these four values the more accurate the screening tool is as the number of false positives and false negatives is reduced. Typically the tool that has the highest sensitivity, specificity, and predictive values for identifying a condition is referred to as the 'gold standard'.

Multiple systematic reviews have identified GMs (Einspieler *et al.*, 2004) as the gold standard for identifying infants at risk of CP. Spittle, Doyle and Boyd (2008) assessed the clinometric properties of tools designed to assess preterm infants during the first year of life. Based on having the highest reliability scores[4], they recommended the Test of Infant Motor performance (TIMP) (Campbell, 2012), the Alberta Infant Motor Scale (AIMS) (Piper and Darrah, 1994b) and GMs (Einspieler *et al.*, 2004). Bosanquet *et al.* (2013) identified the GMs, alongside brain imaging techniques (such as MRI), as being the tool with the best evidence and predictive accuracy for CP. When assessing the best measures for early identification of CP, Novak *et al.* (2017b) identified the GMs, the Hammersmith Infant Neurological Exam (HINE) (Haataja *et al.*, 1999), and brain imaging techniques as the best predictors for CP when infants are under five months of age. Finally Kwong *et al.* (2018) also identified GMs as the best tool for assessing spontaneous infant movement.

However, GMs is not practical for most Primary Health Care Professionals (PHCPs). As discussed in Chapters 1 and 3, GMs is a qualitative assessment of spontaneous early movement. To be able to use GMs, practitioners must undergo a 4-day training course and frequently practice to ensure accuracy. However, training opportunities around CP for PHCPs are currently sparse (Action CP, 2018). Unless training schedules are changed, it is unlikely PHCPs will be able to undergo the training required to use GMs. A smart phone App (Baby Moves) has been developed to allow parents to send videos of their infants to trained GM examiners, which could overcome some of the issue of needing to train large numbers of PHCPs in GMs. However, Kwong *et al.* (2019) reported issues with the quality of the videos submitted by parents. Furthermore, the GMs demonstrates poor psychometrics for use in

[4] Reliability describes how consistent a screening tool is at measuring the same construct in the same setting.

the general population (Bouwstra *et al.*, 2010; Bennema *et al.*, 2016). Therefore, even if the Baby Moves smartphone app becomes a viable option for primary care, some form of gatekeeping would be required to ensure the sensitivity, specificity, and predictive values of the GMs through the Baby Moves smartphone app remain high. The lack of training, the limitations in the smart phone app, and the poor validity of the GMs in the general population demonstrate multiple issues that need to be overcome before GMs could be used within primary care.

Motor screening tools for use in primary care have been recommended, however they also have limitations. As discussed in Chapter 1, Kjølbye *et al.* (2018) recommended only the Ages and Stages Questionnaire 3 (ASQ3) and the Early Motor Questionnaire (EMQ) as motor screening tools to use in primary care. However, both of these tools require multiple uses for an infant to be identified as delayed and have open-ended questions that may only capture infants with clear asymmetries.

Recently it has also become important to consider participatory design in the development of such tools. Participatory design is defined as 'a process of investigating, understanding, reflecting upon, establishing, developing, and supporting mutual learning between multiple participants in collective 'reflection in action'' (Robertson and Jesper, 2013). In essence, participatory design allows those who will use the technology being developed to have a say in how it is developed. This often occurs through an iterative process of giving participants a prototype(s) to interact with and feedback on, before adjusting the prototype(s) based on their feedback and giving the participants the updated prototype(s) once again. As such, participatory design allows the final tool to be more tailored to the needs of the users and often leads to higher levels of acceptance (Tang *et al.*, 2018). The involvement of parents and PHCPs in the development of the screening tool may demonstrate a tool that could be introduced into primary care practices with less disruption.

Content validity indicates how representative the items on a tool are of the targeted construct. There are many methodologies for assessing content validity, such as; Delphi surveys and quantitative judgements by experts. Across methods assessing for content validity, experts are asked to report on how representative the target construct is. Although specialist HCPs are the experts in their field, parents are increasingly being recognised as the experts in their children. Identifying tools that incorporated both specialist HCP opinions as

well as parental concerns, either through the literature they sourced or through the use of participatory design, may also highlight tools whose content aligns more with the types of concerns raised in primary care. As such it would be expected that these tools would be more sensitive to identifying infants in the community with undiagnosed CP.

This scoping review will aim to understand how tools aimed at motor screening in infants aged term to 6 months corrected age were developed. In particular, it will determine whether participatory design methodology was used and what literature was used to develop the included items.

Aims

- Establish whether parents were included in the development of early motor screening tests.
- Determine how items for early motor screening tests were developed.

4.2 Methods

This is a scoping review of current motor screening tools for CP. Firstly how the items were developed, such as from the literature, interviews with experts, and the inclusion of caregivers or parents, was reviewed. Next the descriptive statistics of the literature used to develop the motor items across all of the included tools were summarised. Finally a network citation map was created of the literature used to develop the motor items across all of the included tools. The network citation map allowed for identification of interconnected and standalone components, as well as identification of the core publications.

This review initially started as a systematic review. The original protocol was drafted using Preferred Reporting Items for Systematic Reviews and Meta-Analysis Protocols (PRISMA-P). The protocol was registered on PROSPERO on the 21/1/2019 (registration number: CRD42019119255).

The original plan for this systematic review was to present a broad review of early motor screening tools currently available in the literature. Previous literature had used very narrow inclusion criteria, such as only including publications published in a specific geographical area. This likely would have excluded currently available screening tools, and therefore a broader scope was used. However, at the end of the full text review 242 papers had been included. Rather than restrict inclusion criteria further, the aim of the review changed to

analyse how each of the included tools was developed, with a focus on where the evidence for the items came from.

4.2.1 Identifying the research question and relevant studies

The research questions that guided this new approach to the review were 'What tools (assessments and questionnaires) are there for detecting atypical motor development in infants aged 38 weeks gestation to 6 months corrected age?', 'How were these tools developed?' and 'What was the degree of stakeholder involvement?'. Five electronic databases were searched (MEDLINE, EMBASE, PsycINFO, PubMed and Web of Science) on the 7/12/2018. An updated search was carried out on 15/11/2021. Search terms were adjusted for each database. A detailed search strategy is presented in Appendix D.

4.2.2 Eligibility criteria

Studies were included if;

1) They aimed to measure motor development.
2) At least 50% of the study sample were between ages 37 weeks gestation and 6 months corrected age. When the data was split into categories that were individually analysed (such as 3m, 9m, 12m) these studies were included.
3) The aim of the paper was to describe development of the tools; and/or to describe psychometric properties of the tool; and/or to describe use of the tool with specific populations and settings.

Studies were excluded if they;

1) Only included the tool in the study as an outcome measure, rather than as a screening tool.
2) Only developed a tool for a specific population that was not CP, (i.e. a tool that can only be used in infants with HIV).
3) Used the tool to assess motor development.

Two independent reviewers (JB and GE[5]) assessed the titles and abstracts using Rayyan QCRI (Ouzzani *et al.*, 2016). Both reviewers reviewed all of the titles and abstracts. Inter-rater

[5] GE – Grace Edmonds. Grace was a undergraduate placement year student working with Dr Anna Basu in 2018/19

reliability demonstrated 'moderate' agreement (Cohen's Kappa = 0.64)(McHugh, 2012). When conflicts occurred, the reviewers downloaded the full paper and discussed.

A full text review was then carried out by the three independent reviewers (JB, GE, and CJ[6]). JB reviewed all titles and abstracts while GE and CJ reviewed 10% each. As Rayyan does not have a randomisation option, the titles and papers were chosen by both reviewers assessing every tenth paper when the papers were in alphabetical order. One reviewer began at paper 1 while the other reviewer began at paper 5. Inter-rater reliability was demonstrated to be 'moderate' between JB and CJ, and 'strong' between JB and GE (Cohens Kappa = 0.75 and 0.89, respectively).

The reference lists of the included papers were then screened for papers that may have been missed in the initial search.

4.2.3 Data Analysis

Screening tools used by the included papers were identified. For each screening tool the text describing its development and the texts used to develop the content of the tool were identified.

Data about each tool's development was entered into two tables on Microsoft Excel. The first table described where the data for the items came from, splitting the data into: primary research; the literature; clinical experience; caregivers and parents; experts; and not clear. The second table detailed the methods used in developing the items. This table split the data into methods including: parents; expert opinions; quantitative methodology; and no details given.

Network analysis is the visualisation of a network to look at the relationships between different components of the network. Components are defined as smaller elements that make up part of the network map. CorText Manager was used to map the full citation network map. Mapping the full network enabled the identification of each network component. It also visualises where different tools have cited overlapping literature either directly (2+ tools citing the same publication) or indirectly (tools citing 2+ different

[6] CJ – Ceit Jesmont. Ceit was a medical doctor on a research placement with Dr Anna Basu in 2019.

publications which in turn cite the same publication). CorText Manager is a free to use, online bibliometric tool available at https://www.cortext.net/.

CiteNetExplorer (van Eck and Waltman, 2014) was used for publication analysis. CiteNetExplorer is an open source citation network exploration tool available from https://www.citnetexplorer.nl/, which allows for analysis and visualisation of citation networks.

The core publications analysis was carried out in CiteNetExplorer to identify the core publications that influenced the content of the items within the included screening tools. The core publications analysis was carried out on a citation network built from the publications referenced by the screening tools in relation to the development of the tools items. In core publication analysis, core publications are defined as publications cited by a minimum number of other publications within the network. This study classified core publications as those that had been cited at least twice within the network. To identify clustering within the core network, the 'drill down' function was first used to remove publications from outside of the core network. Then 'Clustering analysis' was used to assign the core publications to clusters based on the levels of association between the publications (resolution [7]= 1; minimum cluster size = 5). Drilling down the network before carrying out the cluster analysis was necessary as attempts at analysing the whole network resulted in clusters forming around each of the tools, rather than demonstrating associations between the publications.

4.3 Results

The search identified 14,860 texts; removal of duplicates reduced this to 7792 texts (Figure 5). Overall, 294 texts were included, identifying 42 screening tools, shown in Table 9. Five tools were excluded; due to lack of access (n=3; Mullen Scale of Early Learning, Taiwan Birth Cohort study tool and Woodside), or texts not being available in English (n=2; Taipei II and Voijta).

[7] Resolution determines how much detail is offered at the lowest level of the classification system. The higher the resolution the more groups will be identified with fewer citations per group. A resolution of 1 is default.

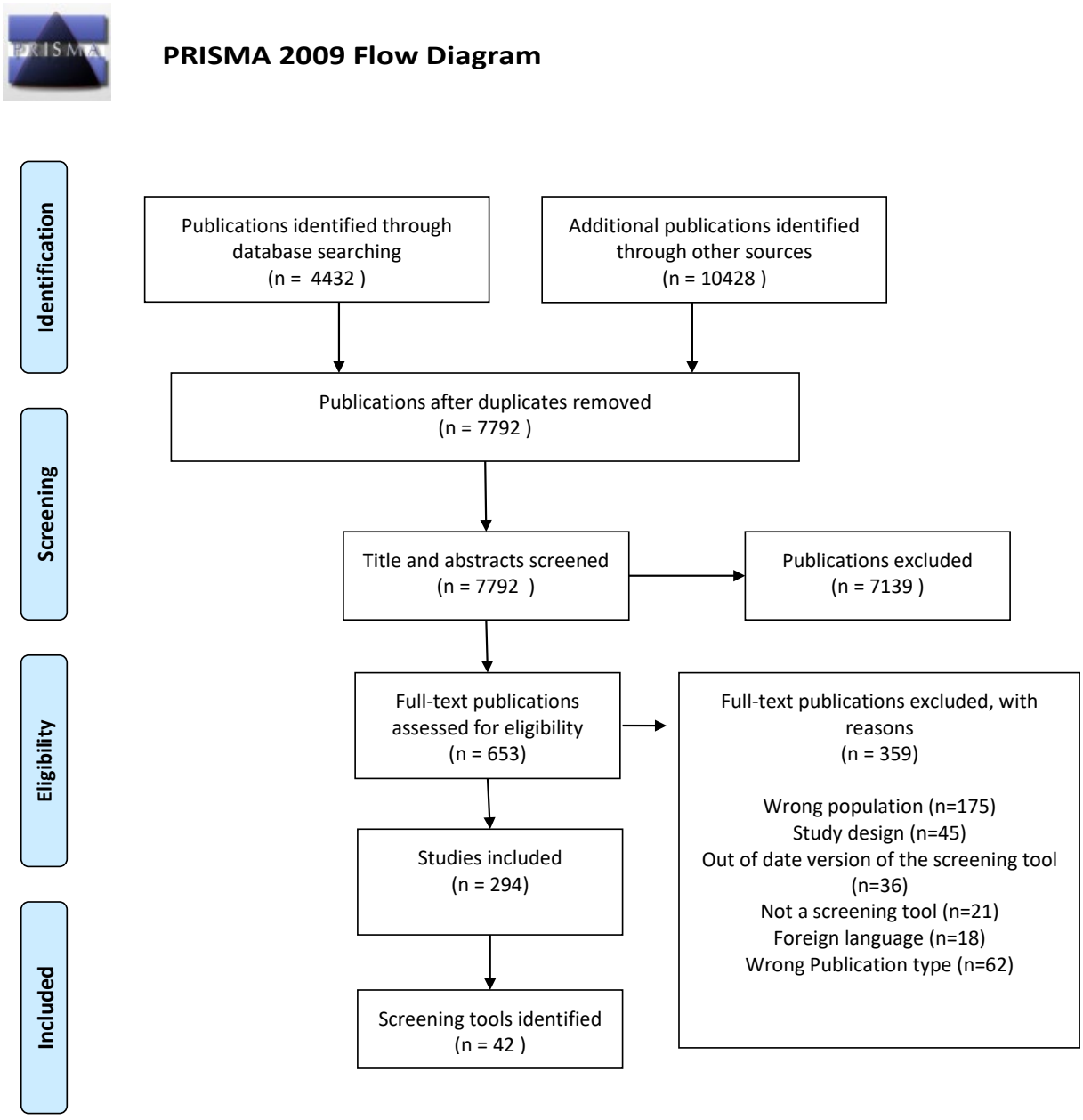


Figure 5 Flow diagram of the study selection process, modified from Moher, et al. (2009)

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
1	Ages and Stages Questionnaire	ASQ3	Squires *et al.* (2009)	Developmental screening in children aged 1 month to 5 ½ years.	Parents and caregivers	Yes	Yes	Yes		
2	Alberta Infant Motor Assessment	AIMS	Piper and Darrah (1994a)	Observational assessment scale of gross motor maturation between birth and independent walking.	HCP	Yes		Yes		
3	Amiel-Tison Neurological examination		Amiel-Tison and Grenier (1983)	Neurological examination for assessing neuromotor development between	HCP	Yes			Yes	

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
				birth to 12 months of age.						
4	Bayley Infant Neurodevelopmental Screener	BINS	Aylward (1995)	Developmental screening tool for ages 3-24 months.	HCP					
5	Bayley Scales of Infant and Toddler Development, Third Edition.	Bayley III	Bayley (2006)	To assess motor, cognitive, language, social emotional, and adaptive behaviour development in babies and young children.	HCP	Yes		Yes		
6	Cambodian Developmental	cDMAT	Ngoun *et al.* (2012)	Culturally sensitive and environmentally appropriate milestone	HCP	Yes		Yes		

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
	Milestone Assessment Tool			assessment tool for Cambodian children living rurally up to age 6 years.						
7	Caregiver Reported Early Development Instruments	CREDI	Short form (McCoy, Waldman and Fink, 2018) Long form (Waldman *et al.*, 2021)	Population level measure of early childhood development focusing on milestones and behaviours from birth to age 3 years.	Parents and caregivers	Yes	Yes	Yes		

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
9	Early motor pattern profile	EMPP	Morgan and Aldag (1996)	CP screening tool consisting of common neurological findings, such as hand fisting.	HCP	Yes				
10	Early Movement Indicator	EMI	Greenwood *et al.* (2002)	Screening and monitoring of motor skill progression that may alter in response to intervention.	HCP	Yes				Yes
11	Grasp and Reach Assessment of Brisbane	GRAB	Perez *et al.* (2016)	Screen for asymmetries in unilateral and bilateral upper limb reaching and grasping behaviours in infants	HCP	Yes		Yes		

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
				with asymmetrical brain injury.						
12	Hammersmith Infant Neurological examination	HINE	Haataja *et al.* (1999)	Examination of neurological function in infants aged 3 to 24 months. The HINE can be also used to screen for CP.	HCP	Yes		Yes	Yes	
13	Hammersmith Neonatal Neurological Examination	HNNE	Dubowitz, Dubowitz and Mercuri (1999)	Neurological examination of neonates.	HCP	Yes		Yes	Yes	

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
14	Hand Assessment for Infants	HAI	Krumlinde-Sundholm *et al.* (2017)	Assessment of bi-lateral hand use and quantification of asymmetries in hand use for infants age 3 to 12 months.	HCP	Yes				Yes
15	Harris Infant Neuromotor Test	HINT	Harris, Megens and Daniels (2010)	Screening tool for neuromotor, cognitive, and behavioural concerns in infants aged 3 – 12 months	HCP (Parents and caregivers are asked to complete 5 items)	Yes		Yes	Yes	

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
16	Infant and Young Child Development	IYCD	Lancaster *et al.* (2018)	Multiculturally sensitive tool to measure: fine and gross motor; receptive and expressive language; and socioemotional development in children age 0-3 years.	Parents and caregivers	Yes		Yes		
17	Infant Motor Profile	IMP	Heineman (2010)	Assessment of spontaneous infant motor behaviour to assess development between 3 and 18 months of age.	HCPs	Yes				Yes

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
18	Infant Neurological International Battery	INFANIB	Ellison, Horn and Browning (1985)	Developmental screening tool for assessing neuromotor development in infants aged 1-18 months.	HCPs	Yes				
19	Infant neuromotor assessment		Magasiner (1993)	Neurological screening assessment of infants aged 4 ½ months.	HCPs	Yes				
20	Malawi Developmental Assessment Tool	MDAT	Gladstone *et al.* (2008) Gladstone *et al.* (2010a)	Culturally appropriate developmental assessment tool of motor, language, and social development in	Parents and caregivers	Yes	Yes	Yes		

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
			Gladstone *et al.* (2010b)	African children aged 0-6 years.						
21	Milani-Comparetti		Milani-Comparetti and Gidoni (1967)	Neurodevelopmental examination of 0-2 year old children.	HCPs	Yes		Yes	Yes	
22	Movement Assessment for Infants	MAI	Chandler, Andrews and Swanson (1980),	Assessment for neurological dysfunction in infants aged from birth to 1 year.	HCPs	Yes				

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
23	Movement Quality Measure		Janssen *et al.* (2012)	To allow comparable qualitative assessment of motor skills in children across paediatric physical therapists and longitudinal assessments.	HCPs	Yes		Yes		Yes
24	Neonatal Intensive Care Unit Network Neurobehavioral Scale	NNNS	Lester and Tronick (2004)	Comprehensive assessment of neurological integrity and behavioural function in typical developing infants and	HCPs	Yes				

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
				infants at risk or exposed to drugs.						
25	Neonatal Neurodevelopmental Examination	NNDE	Allen and Capute (1989)	Assessment of posture, tone, reflexes, symmetry, oromotor function, cranial nerve function, auditory and visual responses, and behaviour in neonates.	HCPs	Yes				
26	Neuro-sensory Motor Developmental Assessment	NSMDA	Burns, Ensbey and Norrie (1989)	Assessment and classification of neuromotor development in	HCPs	Yes				

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
				children aged 1 month to 6 years.						
27	PediaTrac		Lajiness-O'Neill *et al.* (2018)	Web based questionnaire for tracking infant development over time between ages 0- 12 months, in sensorimotor, deeding, sleep, language, cognition, socio-emotional and attachments domains.	Parents and caregivers	Yes	Yes	Yes		

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
28	Pediatric Evaluation of Disability Inventory - Computerised Adaptive Test (PEDI-CAT) (Developed from the PEDI)	PEDI-CAT	Dumas *et al.* (2010)	Computer adaptive test that assesses daily activities, mobility, social/cognitive, and responsibility in children aged birth – 20 years.	Parents and caregivers	Yes	Yes	Yes		
29	PEDS: Developmental Milestones	PEDS:DMs	Brothers, Glascoe and Robertshaw (2008)	Assessment of parental concerns and the child's developmental progress in motor, language, self-help, and self-emotional domains	Parents and caregivers	Yes				

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
				between birth and 7-11 years.						
30	Posture and Fine Motor Assessment of Infants		Case-Smith and Bigsby (2001)	Assessment to identify if an infants motor skills are developmentally delayed between 2 and 12 months.	HCPs	Yes				
31	Prechtl's General Movements Assessment. GM optimality score for preterm and writhing GMs, and	GMs	Einspieler *et al.* (2004) Ferrari, Ciono and	Gestalt assessment of infant movements up to 20 weeks corrected age.	HCPs					Yes

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
	Assessment of Motor Repertoire – 3 to 5 months		Prechtl (1990)							
32	Rapid Neurodevelopmental Assessment		Khan *et al*., (2010)	Assessment to determine functional status across fine motor, gross motor, vision, hearing, speech, cognition, behaviour and seiezures in children aged birth – 16 years.	HCPs	Yes		Yes	Yes	

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
33	Specific Test of Early Infant Motor Performance	STEP	Gower *et al.* (2019)	Developmental screening test for early motor deficits in infants age 34 weeks gestational age and 4 months corrected age.	HCPs	Yes				
34	Standardized Infant NeuroDevelopmental Assessment Neurological scale	SINDA	Hadders-Algra *et al.* (2019)	Screening tool to detect infants aged 6 weeks – 12 months at high risk of neurodevelopmental disorders.	HCPs					

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
35	Structured Observation of Motor Performance	SOMP-I	Persson and Strömberg (1993)	Screening for infants with atypical motor development aged 0-12 months.	HCPs	Yes				
36	Test of Infant Motor Performance	TIMP	Campbell (2012)	Assessment of postural and selective motor control of functional performance in infants aged 34 weeks gestational age and 4 months corrected age.	HCPs	Yes		Yes		
37	Test of Infant Motor Performance - Screening Inventory	TIMPSI	Campbell (2012)	Screening assessment of postural and selective motor control	HCPs	Yes				

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
				of functional performance in infants aged 34 weeks gestational age and 4 months corrected age.						
38	Test of Motor and Neurological function		DeGangi, Berk and Valvano (1983)	Assessment of automatic and equilibrium reactions, muscle tone, primitive reflexes, and qualitative movement in infants aged 0-12 months	HCPs	Yes			Yes	

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
39	The Denver developmental screening test II	Denver II	Frankenburg *et al.* (1992)	Screening for developmental problems in infants and children up to age 6 years.	HCPs	Yes				
40	The Neoneuro		Sheridan-Pereira, Ellison and Helgeson (1991)	Neurological examination of full term neonates.	HCPs	Yes				
41	The Neurological Examination of the Full-term Newborn Infant	NNE	Prechtl (1977)	Neurological examination of newborn infants.	HCPs					

	Tool Name	*Acronym*	*Reference*	*Intended use*	*Designed to be used by*	*Item development methods*				
						Literature	*Parent opinions*	*Expert opinions*	*Researcher's own clinical experience*	*Primary research*
42	Touwen		Touwen (1976)	Neurological and developmental examination of infants across the first year of life.	HCPs	Yes				

Table 9 Screening tools included in the scoping review

Although all the tools included assessed infant motor development, the included tools were developed for different purposes, see Table 9. For example, only seven of the included tools were developed for use by caregivers (ASQ-3, CREDI, IYCD, MDAT, PEDI-CAT, PediaTrac, and PEDS:DMs). Twelve were developed as neurological assessments (Amiel-Tison, EMPP, HINE, HNNE, Infant neuromotor assessment, Millani-Comparetti, MAI, NNNS, GMs, Neoneuro, NNE, and Touwen). Nineteen were developed to assess development (AIMS, BINS, Bayley III, CDMAT, CREDI, EMI, IYCD, IMP, MDAT, Milani-Comparetti, Movement quality measure, NSMDA, PediaTrac, PEDI-CAT, PEDS:DMs, Rapid Neurodevelopmental assessment, STEP, TIMP, and TIMPSI) and sixteen were developed to screen for emerging developmental conditions such as developmental delay and CP (ASQ-3, BINS, Bayley III, EMPP, HINE, HINT, INFANIB, NNDE, PEDS:DMs, Posture and fine motor assessment of infants, STEP, SINDA, SOMP, Test of motor and neurological function, and Denver II). Two were developed to assess only hand function (GRAB and HAI). Seven were explicitly developed to be culturally or population sensitive (ASQ3, AIMS, CDMAT, CREDI, IYCD, MDAT, and Rapid neurodevelopmental assessment). Seven were explicitly developed to standardise assessment (Amiel-Tison, EMPP, HNNE, Movement quality measure, NNNS, SOMP-I, and TIMPSI). Three were developed to be quick assessments (BINS, CDMAT, and Denver II) while one was developed to be a comprehensive assessment (Bayley III).

As described in previous reviews, the ASQ-3, AIMS, Amiel-Tison, Bayley III, Denver II, GMs, HINE, HINT, INFANIB, MAI, NeoNeuro, NNE, NNNS, NSMDA, Posture and fine Motor Assessment, Rapid, SOMPI, TIMP, Touwen, were described as having acceptable reliability and validity by the authors (Majnemer, 1998; Spittle, Doyle and Boyd, 2008; Heineman, 2010; Tronick and Lester, 2013; Kjølbye *et al.*, 2018; Dorothy *et al.*, 2019). Validity and Reliability as categorised by the authors is presented in Table 10 for the other measures.

Table 10 Reliability and Validity of the included screening tools.

Tool	Validity	Reliability	Reference
BINS	Good internal consistency	Good test-retest reliability and inter-rater reliability	(Aylward, 1995)
cDMAT	Still requires testing	excellent inter-rater reliability	(Ngoun *et al.*, 2020)
CREDI short form	adequate criterion validity	good retest reliability	(McCoy, Waldman and Fink, 2018)
CREDI long form	good construct validity, criterion related validity	good test-retest reliability, and internal consistency	(Waldman *et al.*, 2021)
EMPP	Predictive (sensitivity = 90.1%, specificity 87.3%)	Inter-rater reliability (90.34%)	(Morgan and Aldag, 1996)
EMI	excellent criterion validity	good inter-rater reliability and split half reliability	(O'Grady and Dusing, 2015)
GRAB	moderate to strong construct validity	strong internal consistency	(Perez *et al.*, 2016)
HNNE	moderate predictive validity	excellent inter-rater reliability	(Eeles *et al.*, 2016; Howard *et al.*, 2023)
HAI	good predictive validity	excellent inter-rater and test-retest reliability	(Ryll *et al.*, 2021; Ullenhag *et al.*, 2021)
IYCD	Still requires testing	good-excellent inter-rater and test-retest reliability	(Gladstone *et al.*, 2021)
IMP	Fair to moderate concurrent validity	Moderate-Good inter-rater reliability	(Heineman *et al.*, 2013)
The infant neuromotor assessment	Predictive validity (positive predictive value = 85.3%, negative predictive value = 98.6%)	Good inter-rater reliability	(Magasiner *et al.*, 1997)
MDAT	Good content and face validity	Adequate to excellent inter-rater reliability	(Gladstone *et al.*, 2010b)
Milani-Comparetti	Low predictive validity (sensitivity of 33%)	Acceptable to excellent Inter-rater reliability Good to excellent test-retest reliability	(VanderLinden, 1985; Stuberg *et al.*, 1989)
Movement Quality Measure	Content validity	Moderate inter-rater reliability. Good-	(Janssen *et al.*, 2012; Dekkers *et al.*, 2018)

		excellent item agreement.	
NNDE*	Adequate to excellent Sensitivity and specificity	-	(Allen and Capute, 1989)
PediaTrac	Good construct validity	Good internal consistency reliability	(Lajiness-O'Neill *et al.*, 2018)
PEDI-CAT	Significant discriminant validity	test-retest reliability (>.957)	(Haley *et al.*, 2012)
PEDS:DMS	sensitivity and specificity >70%	High test-retest and inter-rater reliability	(Brothers, Glascoe and Robertshaw, 2008)
STEP	Predicative validity with excellent sensitivity and specificity	Good to excellent Intra- and inter-rater reliability	(Gower *et al.*, 2019)
SINDA	Satisfactory predictive validity	Excellent intra- and inter- rater reliability.	(Hadders-Algra *et al.*, 2019)
TIMPSI	Concurrent validity	Excellent intra- and inter-rater reliability and test-retest reliability	(Campbell *et al.*, 2008; Krosschell *et al.*, 2013)
Test of Motor and Neurological function	Moderate decision validity	High inter-rater reliability (0.93-0.97)	(DeGangi, Berk and Valvano, 1983)

4.3.1 Item development

Items from the included tools were developed from the literature (n=36, see Table 9) from clinical experience (n=6, see Table 9), or from primary research (n=5, see Table 9). 2 tools had not reported where they had developed their items from at the time of writing (NNE and SINDA) (Prechtl, 1977; Hadders-Algra *et al.*, 2019).

Primary research was used to develop the items on the EMI, GMs, HAI, IMP, and the Movement Quality Measure (Greenwood *et al.*, 2002; Einspieler *et al.*, 2004; Heineman, 2010; Janssen *et al.*, 2012; Krumlinde-Sundholm *et al.*, 2017). The GMs, HAI, and IMP developed their items from video observations of infant development. The Movement Quality Measure developed items by interviewing experts and focus groups using Nominal Group Technique. In contrast, the EMI began their development process by carrying out a national survey of parents and professionals to validate a general movement outcome measure, that had previously been identified by Priest *et al.* (2001). A literature search was

then used to further develop the general movement outcome measure and the items used within the tool.

Expert opinions were involved in the development of items for 18 tools, see Table 9. Descriptions of the methods used are presented in Appendix E.

Parental opinions were included in the ASQ3, CREDI, MDAT, PEDI-CAT, and PediaTrac (Squires *et al.*, 2009; Dumas *et al.*, 2010; Gladstone *et al.*, 2010b; Lajiness-O'Neill *et al.*, 2018; McCoy, Waldman and Fink, 2018; Waldman *et al.*, 2021). The ASQ has undergone three revisions since it was first developed. During the first revision, feedback from parents, alongside feedback from project staff, nurses, and paediatricians, was used to reword items to clarify their meanings (Squires *et al.*, 2009). No information was provided regarding whether parents were included in the second and third revisions of the ASQ.

The PEDI-CAT recruited parents of children with disabilities (n=6) whose children were under 21 years of age to take part in focus groups. The focus groups aimed to provide feedback on the items and response scales of the PEDI (Dumas *et al.*, 2010). The parents were asked 'if there were other important functional skills in each of the PEDI's three content domains that should be addressed' (Dumas *et al*., 2010), if the items were clear and understandable, and about the response scales. Only 1 parent who took part in the focus groups had a child with CP (hemiplegia). After further focus groups with clinicians, 11 parents (6 of whom had a child with a disability) took part in cognitive interviewing. The cognitive interviews examined the content, format, and comprehension of the item responses. No parents of children with CP were included in the cognitive interviews.

Both the CREDI and the PediaTrac used cognitive interviews with caregivers to identify how the caregivers interpreted the items and which items were difficult to understand, inappropriate, or incomplete (Lajiness-O'Neill *et al.*, 2018; McCoy, Waldman and Fink, 2018). 'Approximately 60 caregiver-child pairs' (McCoy, Waldman and Fink, 2018) took part in the cognitive interviews for the CREDI, while 11 caregivers took part in the cognitive interviews for the PediaTrac. Neither study reported the demographics of the children being responded about.

The MDAT used 'village' focus groups to identify the developmental items to be included in their tool (Gladstone *et al.*, 2010a). 10 focus groups were carried out involving mothers,

fathers and grandparents across four areas of Malawi. No data was given on the children or grandchildren of the participants. To avoid preconceived western ideas of child development, Gladstone *et al.* (2010a) asked participants about their experiences of child development and what they believed a child with developmental problems could not do. The concepts and ideas raised in the interviews were then used to generate new items and to modify westernised items identified in the literature.

There were 11 tools for which the process of selecting items was not described (Milani-Comparetti and Gidoni, 1967; Prechtl, 1977; Chandler, Andrews and Swanson, 1980; Amiel-Tison and Grenier, 1983; Allen and Capute, 1989; Persson and Strömberg, 1993; Haataja *et al.*, 1999; Lester and Tronick, 2004; Heineman, 2010; Khan *et al.*, 2010; Hadders-Algra *et al.*, 2019), and 2 tools for which we were unable to access the relavent texts (Mullen, 1995; Case-Smith and Bigsby, 2001).

4.3.2 Item mapping

Initial network mapping in Cortex Manager demonstrated 32 of the 42 tools to form a single interconnected component, meaning that the 32 tools were connected to each other through their citation networks. 6 tools (Denver II, GRAB, HAI, PEDI-CAT, PEDS:DMs, and Test of Motor and Neurological Functions) were identified to have formed their own individual components, meaning that each of these tools did not share citations with the other tools included in the analysis. Due to being unable to access the original texts, the SINDA, the Posture and Fine Motor Assessment of Infants, and the BINS were shown as individual nodes not connected to any components.

Analysis of the initial network map demonstrated clear missing citations between different editions of publications. For example the Bayley Scales of Infant Development, the Bayley II, and Bayley III are succeeding versions of the California Scales of Development (Bayley, 1969). However, the Bayley III does not directly cite the California Scales of Development, and therefore a citation between the two was not included in the original network map. As a result the citations between clear succeeding versions and editions of included publications were added to produce a second network map, Figure 6.

The addition of citations between succeeding versions resulted in the largest single interconnected component going from 32 tools included to 36 tools included, Figure 6. Of

the Denver II, GRAB, HAI, PEDI-CAT, PEDS:DMs, and Test of Motor and Neurological Functions, only the Grab, HAI, and PEDS:DMs remained as individual components.

4.3.4 Analysis of the core component

Analysis of the citation network within CiteNetExplorer confirmed that that the largest inter-connected component consisted of 200 publications with 225 citations.

Core publications analysis demonstrated 47 core publications that had been used to develop the items included in the screening tools. Between the 47 core publications there were 79 citation links, Figure 7A. The 47 core publications were made up of 23 books, 21 journal articles, 1 editorial, 1 technical report and 1 Masters thesis. Descriptions of the publications are given in Table 10. 29 of the core publications described one of included screening tools. The other 18 publications included 11 publications that described the development or validation of a tool or examination technique not included in this review, 4 publications that provide information on infant development, and 3 publications that describe original research.

Cluster analysis of the 47 core publications identified three groups, shown in purple, green, and blue in Figure 7. As an image containing all the publication names could not be made, images were made of each cluster. The blue cluster consists of publications that focus infant development from birth and include preterm assessment. These publications typically look at development over the first months of life with a couple exceptions that also address the first couple years of life, such as Touwen (1976) and Egan (Egan, 1990) who addressed development up to 2 years and 4 ½ years respectively, Figure 7B. The green cluster contains publications that assess infant development over the first years of life, starting from birth, or shortly after birth, up to 6 years of age, Figure 7C. The purple cluster demonstrates publications that describes development from infancy, through childhood and into adulthood, Figure 7C.

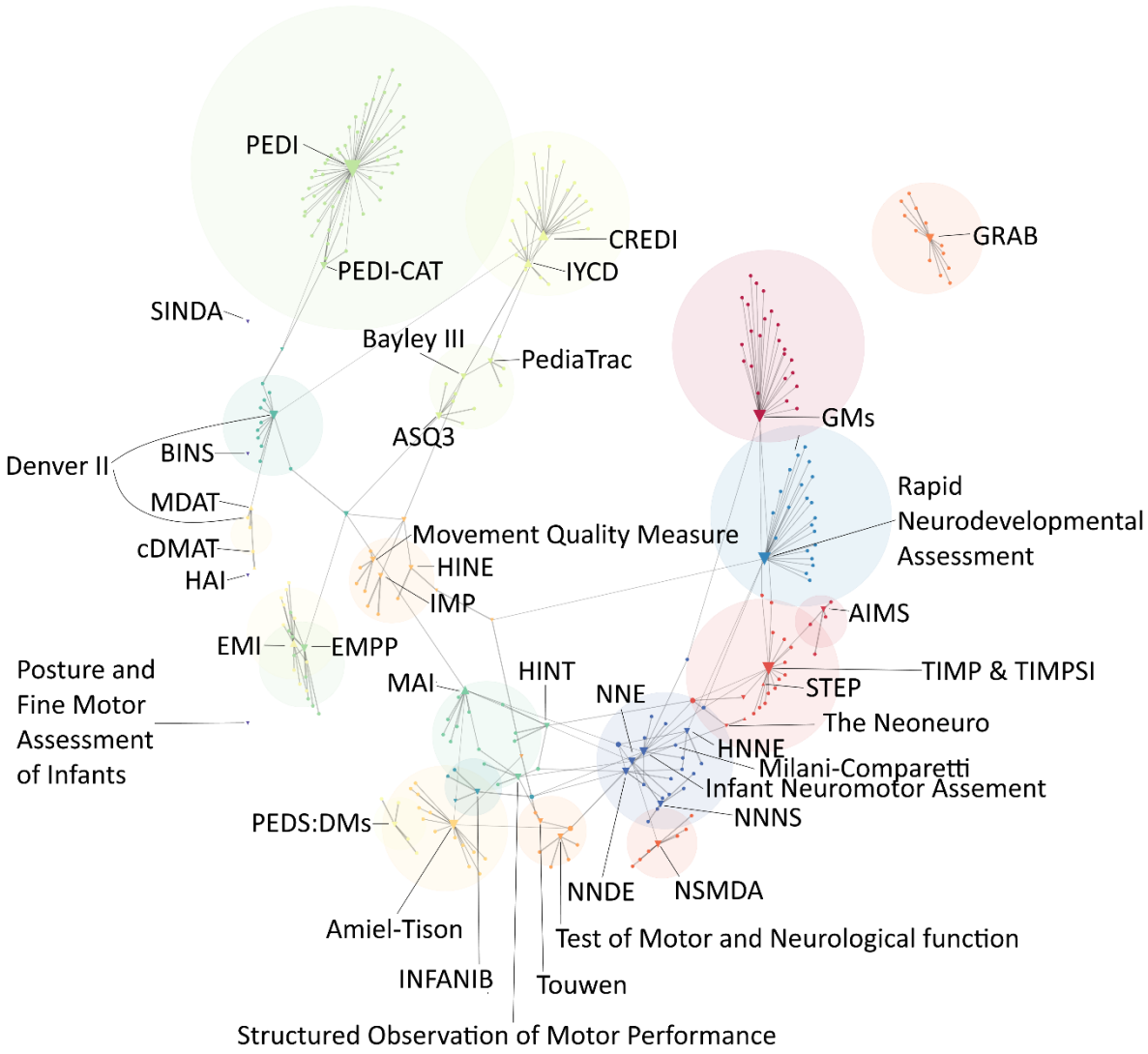
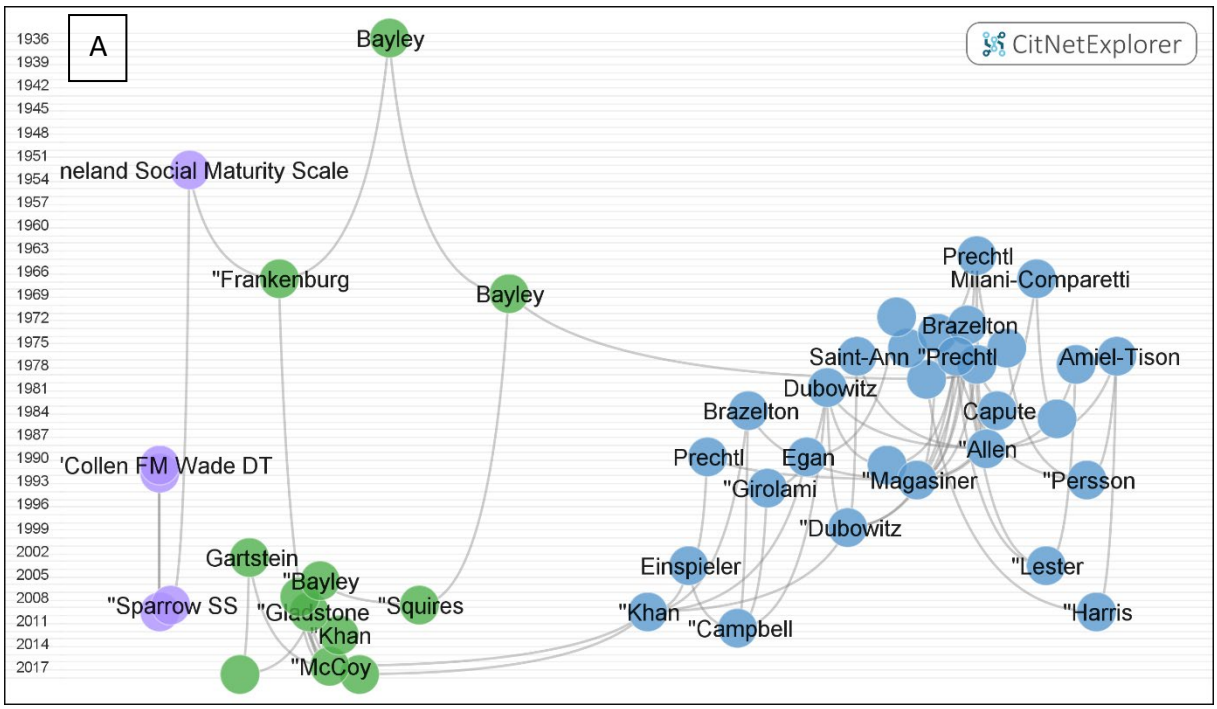


Figure 6 Network map of the literature used to develop the items on currently available motor development screening tools. Transparent circles represent clustering of publications. Coloured triangles and circles represent publications within the network. Grey lines between publications represent citations. Cluster analysis resulted in clusters forming around each tool. This is likely due to the citation network being built from the references of each tool. Network mapping demonstrated 36 of the 42 tools to form one component, as shown by the grey lines (citations) between the coloured clusters. In total 6 tools formed individual components with no citations between themselves and the other components; BINS, GRAB, HAI, PEDS:DMS, Posture and fine motor assessment of infants, and SINDA,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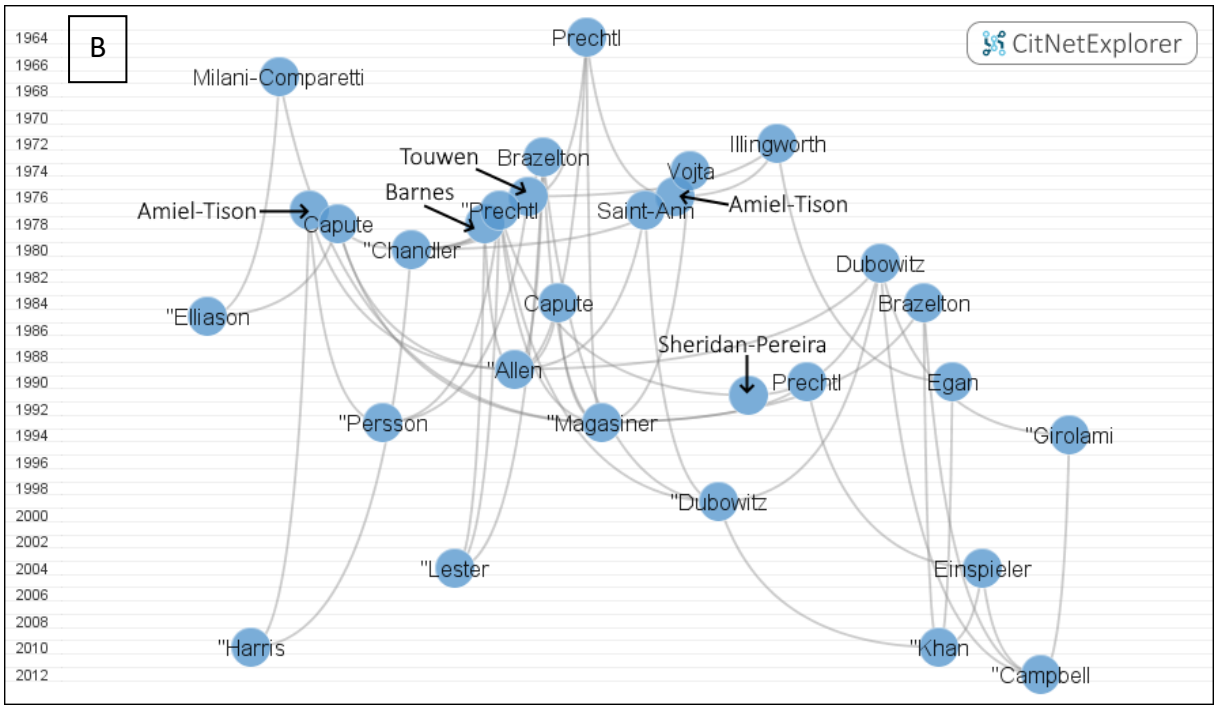
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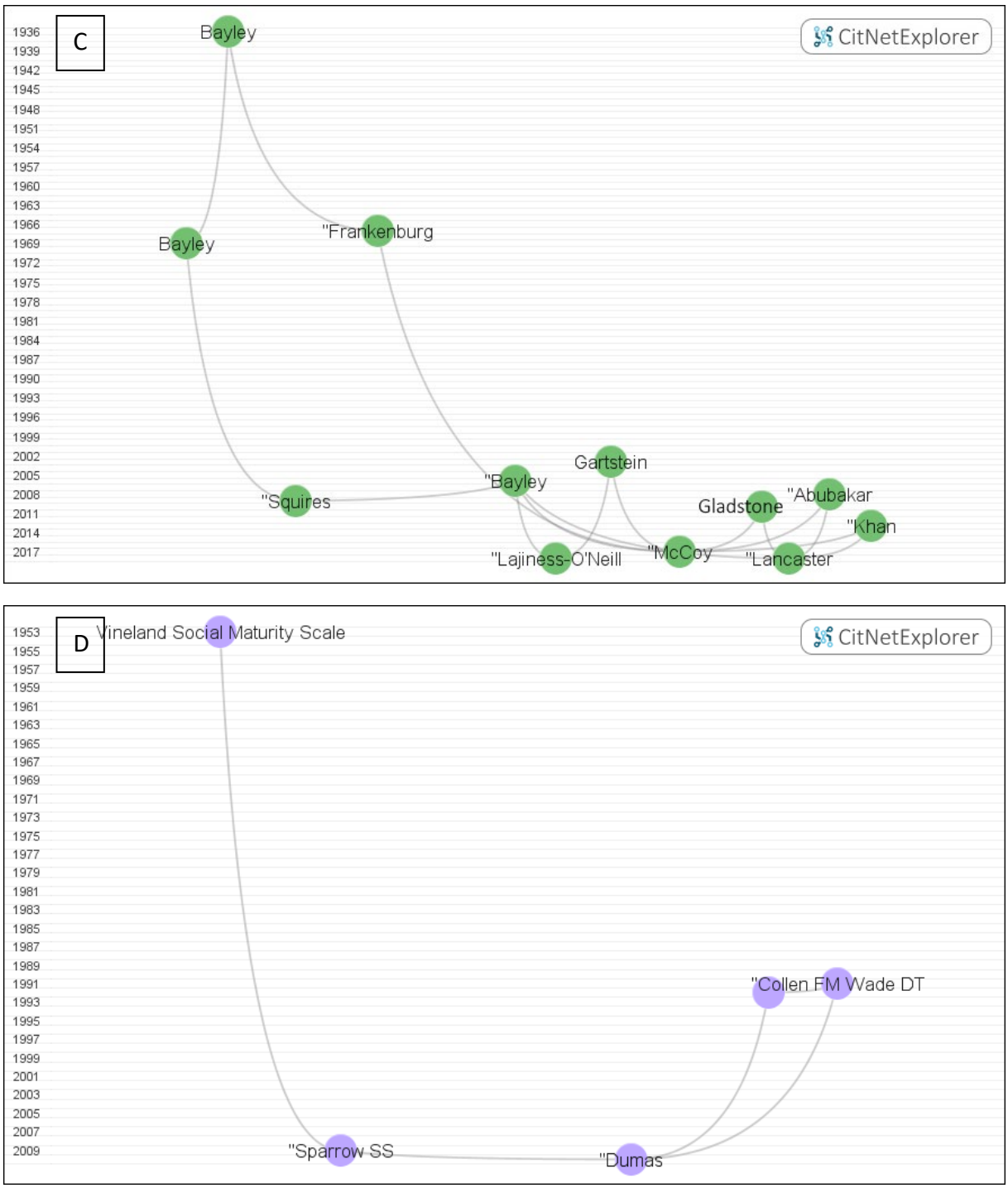


Figure 7 Cluster analysis of the core publications influencing the content of currently available screening tools. Each circle represents a publication within the core network. Grey lines between circles represent citations. Colours represent clusters. Although the y-axis shows the year of publication, the x-axis has no value and only serves to show distance between the publications. A) The full network. In total 47 publications were identified as core publications. Cluster analysis identified three groups, shown in purple, green, and blue. As an image containing all the publication names could not be made, images were made of each cluster.All four images were created in CiteNetExplorer.

Publication	*Publication type*	*Description*
Describes included screening tool		
Allen and Capute (1989)*	Journal article	Describes the NNDE when performed at full term or at discharge from a neonatal intensive care unit in a population of high risk infants. Neurological outcomes were followed up 1-5 years later. The NNDE was found to be significantly predictive of CP and neurological disfunction.
Amiel-Tison (1976)*	Journal article	Description of a neurological examination used by Amiel-Tison.
Bayley (1936)*	Book	Manual for the Californian Infant Developmental Scales, a previous version of the Bayley III.
Bayley (1969)*	Book	Manual for the Bayley Scales of Infant Development, a previous version of the Bayley III.
Bayley (2006)	Book	Manual for the Bayley III
Campbell (2012)*	Book	Manual for the TIMP and TIMPSI.
Chandler *et al*. (1980)*	Book	Manual for the MAI
Dubowitz *et al*. (1999)*	Book	Describes the first version of the HNNE.
Dubowitz and Dubowitz (1981)*	Book	Describes the second version of the HNNE.
Dumas *et al*. (2010)*	Journal article	Describes the process of increasing the item inventory of the PEDI for use in the PEDI-CAT.
Einspieler *et al*. (2004)*	Book	Manual for the GMs

Publication	*Publication type*	*Description*
Ellison *et al*. (1985)*	Journal article	Describes the development of the INFANIB.
Frankenburg and Dodds (1967)*	Journal article	Describes the development and standardisation of the Denver developmental screening test.
Gladstone *et al*. (2010)	Journal article	Describes the development and psychometric analysis of the MDAT in rural Africa.
Haley *et al*. (1992)*	Book	Manual for the PEDI.
Harris *et al*. (2010)*	Book	Manual for the HINT
Khan *et al*. (2010)*	Journal article	Reliability and validity analysis of the RNDA against the adapted Bayley II.
Khan *et al*. (2013)	Journal article.	Psychometric analysis of the RNDA in 77 children aged >2 to 5 year old, against the Test of Adapted Behaviour and the intelligence quotient tests within the Bayley Scales of Infant Development II, Stanford Binet Intelligence Scale, Wechsler Preschool and Primary Scales of Intelligence.
Lajiness-O'Neill *et al* (2018)	Journal article	Development and psychometric analysis of the PediaTrac.
Lancaster *et al* (2018)	Journal article	Development of the Infant and Young Child Development (IYCD) tool
Lester and Tronick (2004)*	Journal article	Describes the history of infant assessment before describing the NNNS' features, the scientific basis for the examination, and the developmental model in which it is based on.

Publication	*Publication type*	*Description*
Magasiner (1993)*	Masters thesis	Identified postures that predicted adverse neurodevelopmental outcomes by examining postural development in very low birthweight and typical birthweight infants. The resulting predictive postures were developed into the INA.
Milani-Comparetti and Gidoni (1967) *	Journal article	Provides descriptions of the items included in the Milani-Comparetti tool.
Persson and Strömberg (1993)*	Journal article	Presents the protocol for the Structured Observation of Motor Performance along with the interobserver agreement and interobserver consistency.
Prechtl (1977)*	Book	Manual for the NNE (2nd edition)
Prechtl (1990)	Editorial	Provides an argument for qualitative examination of general movements in infants, also known as Prechtl's GMs.
Sheridan-Pereira *et al*. (1991)*	Journal article	Development of the NeoNeuro.
Squires *et al* (2009)	Technical report	Technical report for the ASQ3 including the development of the ASQ3 and the psychometrics.
Touwen (1976)*	Book	This study examined the developmental course of items used in neurological and developmental screening in low risk infants. The items that were found to predictive were used in the Touwen assessment.
Describes a tool not included in the review.		

Publication	*Publication type*	*Description*
Abubakar *et al.* (2008)	Journal article	Describes the psychometric testing of the Kilifi Developmental Inventory in 423 children aged 6-35 months in Kenya.
Brazelton (1973)	Book	Manual for the Neonatal Behavioural Assessment Scale.
Brazelton (1984)	Book	Manual for the Neonatal Behavioural Assessment Scale second edition. The NBAS consist of 53 items assessing habituation, social interactive responses and capabilities, motor system, state organisation and regulation, autonomic system, and reflexes. The NBAS can be used on infants aged 35 weeks gestation to 2 months corrected age. The Primitive Reflex Profile consists of 9 primitive reflexes scored on a 5 point scale ranging from 0 (absent) to 4 (Obligatory or controlling the patient).
Capute (1978)	Book	Reviews the literature on reflexes in typically developing new-borns before presenting their grading system for quantifying the presence of seven primitive reflexes. The Primitive Reflex Profile has been standardised from birth to 2 years
Capute *et al*. (1984)	Journal article	Describes the standardisation of nine primitive reflexes in typically developing infants. Follow up occurred at 1 year of age using the Bayley Scales.
Collen *et al*. (1991)	Journal article	Describes the development of the Rivermead Motor Assessment Gross Function scale for measuring mobility after head injury or stroke. The Rivermead is designed for use in adults.

Publication	*Publication type*	*Description*
Dall (1953)	Book	Manual for the Vineland Social Maturity Scale. The Vineland Social Maturity scales measured social maturity and social competence through the domains of; self help general, self-help dressing, self-help eating, communication, self-direction, socialisation, locomotion, and occupation. The Vineland Social Maturity Scales are valid from birth (Sparrow, 2011).
Gartstein and Rothbart (2003)	Journal article	Describes the psychometric analysis of the revised Infant Behaviour Questionnaire, a parent report measure of infant temperament.
Girolami and Campbell (1994)	Journal article	Evaluation of a neurodevelopmental treatment protocol designed for improving motor control in preterm infants identified as high risk for developmental disability. Outcomes were assessed using the Neonatal Behavioural Assessment Scale and a supplemental motor test that assesses postural control.
McCoy *et al* (2017)	Journal article	Development and psychometric analysis of the Early Childhood Development scale against the Bayley III.
Sparrow *et al*. (2009)	Book	Manual for the Vineland Adaptive Behaviour Scales. The Vineland Adaptive Behaviour Scales measures adaptive behaviours through three domains; communication, daily living skills, and socialisation (Sparrow, Cicchetti and Saulnier, 2021). The latest version (Vineland-3) also measures motor skills and maladaptive behaviours and can be used from birth.
Describes original research		

Publication	*Publication type*	*Description*
Amiel-Tison (1977)	Journal article	This study looked for the presence of abnormal hypertonia of the neck extensor muscles in newborns. In particular the study reports the frequency in which neck extensor hypertonia occurs in infants with signs of cerebral insult.
Prechtl and Beintema (1964)	Book	This study identified the developmental course and the consistency of neurological signs in the neonatal period. This study also identified the extent and for how long each of the neurological signs were influenced by pre-, peri- and post- natal factors.
Saint-Anne Dargassies (1977)	Book	This book presents a study of neurological development in pre-term and term infants. The data was collected using a standardised examination. Within the pre-term data, neurological characteristics by gestational age are presented. Further analysis of the data by foetal age is also presented.
Provides a guide on development and screening		
Barnes *et al*. (1982)	Book	This book provides descriptions of individual reflexes split across four categories; primitive reflexes, prehensile reactions, righting reactions, and equilibrium reactions. Additionally the book presents theories around reflexes and how they relate to motor activity.
Egan (1990)	Book	A practical guide for PHCPs on detection of developmental problems in children. The guide covers history taking, clinical tests of hearing, examination of visual behaviour and acuity, observation of developing motor skills, and language/performance profiles.

Publication	*Publication type*	*Description*
Illingworth (1972)	Book	Describes typical infant development, developmental assessment that can be carried out without specialist equipment, and the difficulties and pitfalls with developmental assessments.
Vojta (1974)	Book	Recommendations of exercises in prone for infants at risk of motor problems as identified by a series of postural reflexes, and deviations in tonus.

Table 11 Descriptions of the 38 core publications. The core publications were identified though core analysis of a citation network. The citation network consisted of publications referenced by the included screening tools in regards to the development of their items. Core analysis was carried out to identify which publications had influenced the content of screening tool items and was carried out using CiteNetExplorer.

4.4 Discussion

In this chapter I showed that early motor development screening tool items were developed from the literature, clinical experience, or from primary research. The content of these items was taken from publications on other tools, primary research, or from books providing guidance on infant development. Few screening tools reported the inclusion of parents or developing the tool from original research. When parents were included, their opinions were most often used to review the content of the items already included. Only the PEDI-CAT included parents of children with disability in the development of the items, however this was only undertaken with one parent of a child with CP.

4.4.1 Tools should measure the entire theoretical construct

Screening tools are often developed by clinical teams for good practical reasons. When developing a tool, the content of the items should cover the entire theoretical construct the tool is aiming to measure. This allows the tool to have content validity. Not only are clinical teams often the ones to use the tool, they also have expertise in the condition the tool is screening for to develop items the cover the theoretical construct. However, the lack of parental inclusion in the development of items may have affected the content validity due to the theoretical construct not being fully covered by the included items. Chapter 2 of this

thesis identified that caregivers report the same signs that have been identified in the literature as well as additional signs of CP that have not been reported.

Discussion about the inclusion of patients and caregivers in the design, conduct, and analysis of research, rather than being the subjects of research, was first incorporated into the UK's Research Governance Framework in 2005 (Department of Health). However, this review only identified 5 of the included tools to have included parents and caregivers within the development of the tool (the ASQ3, CREDI, MDAT, PEDI-CAT, and PediaTrac (Squires *et al.*, 2009; Dumas *et al.*, 2010; Gladstone *et al.*, 2010b; Lajiness-O'Neill *et al.*, 2018; McCoy, Waldman and Fink, 2018; Waldman *et al.*, 2021)), with 18 of the 42 tools being published after 2005 (ASQ-3, Bayley III, cDMAT, CREDI, GRAB, HAI, HINT, IYCD, IMP, MDAT, Movement quality measure, PediaTrac, PEDI-CAT, PEDS:DMs, Rapid Neurodevelopmental Assessment, STEP, TIMP, and TIMPSI (Bayley, 2006; Brothers, Glascoe and Robertshaw, 2008; Squires *et al.*, 2009; Dumas *et al.*, 2010; Gladstone *et al.*, 2010a; Gladstone *et al.*, 2010b; Harris, Megens and Daniels, 2010; Khan *et al.*, 2010; Campbell, 2012; Janssen *et al.*, 2012; Ngoun *et al.*, 2012; Perez *et al.*, 2016; Krumlinde-Sundholm *et al.*, 2017; Lajiness-O'Neill *et al.*, 2018; McCoy, Waldman and Fink, 2018; Gower *et al.*, 2019; Hadders-Algra *et al.*, 2019; Waldman *et al.*, 2021)). The UK's inclusion of patients and caregivers symbolises a broader move within the global healthcare sector to be more inclusive. Given this background, it is interesting that so few have involved parents and/or caregivers. Furthermore only 2 tools (MDAT and PEDI-CAT) asked parents to contribute to the development of the theoretical constructs. It is likely that the content of the majority of these tools does not cover the entire theoretical construct of infant development, let alone CP as noticed by caregivers. Although the theoretical content of parental concerns could have been identified by PHCPs, PHCPs were also unlikely to be included. If so, these tools may not be optimal for assessing CP in primary care as they may not identify the range of early concerns parents observe and then raise. The next chapter will look at the concerns presented by caregivers and the items reported in the included screening tools, to determine if the entire theoretical construct is covered.

4.4.2 Further network mapping is needed

Further research is also needed into the full network map around the screening tools. This review identified only 5 tools that were developed from original research (EMI, HAI, IMP,

Movement quality measure, and GMs (Greenwood *et al.*, 2002; Einspieler *et al.*, 2004; Heineman, 2010; Janssen *et al.*, 2012; Krumlinde-Sundholm *et al.*, 2017)), while 37 used previous literature to develop their items (ASQ3, AIMS, Amiel-Tison, Bayley III, cDMAT, CREDI, EMPP, EMI, GRAB, HINE, HNNE, HAI, HINT, IYCD, INFANIB, Infant neuromotor assessment, MDAT, Milani-Comparetti, MAI, Movement quality measure, NNNS, NNDE, NSMDA, PediaTrac, PEDI-CAT, PEDS:DMs, Posture and Fine motor assessment of infants, Rapid neurodevelopmental assessment, STEP, Structured observation of motor performance, TIMP, TIMPSI, Test of motor and neurological function, Denver II, The Neoneuro, and Touwen. (Milani-Comparetti and Gidoni, 1967; Touwen, 1976; Chandler, Andrews and Swanson, 1980; Amiel-Tison and Grenier, 1983; DeGangi, Berk and Valvano, 1983; Ellison, Horn and Browning, 1985; Allen and Capute, 1989; Burns, Ensbey and Norrie, 1989; Sheridan-Pereira, Ellison and Helgeson, 1991; Frankenburg *et al.*, 1992; Magasiner, 1993; Persson and Strömberg, 1993; Piper and Darrah, 1994b; Morgan and Aldag, 1996; Dubowitz, Dubowitz and Mercuri, 1999; Haataja *et al.*, 1999; Case-Smith and Bigsby, 2001; Greenwood *et al.*, 2002; Lester and Tronick, 2004; Bayley, 2006; Brothers, Glascoe and Robertshaw, 2008; Gladstone *et al.*, 2008; Squires *et al.*, 2009; Dumas *et al.*, 2010; Gladstone *et al.*, 2010a; Gladstone *et al.*, 2010b; Harris, Megens and Daniels, 2010; Khan *et al.*, 2010; Campbell, 2012; Janssen *et al.*, 2012; Ngoun *et al.*, 2012; Perez *et al.*, 2016; Krumlinde-Sundholm *et al.*, 2017; Lajiness-O'Neill *et al.*, 2018; Lancaster *et al.*, 2018; McCoy, Waldman and Fink, 2018; Gower *et al.*, 2019; Waldman *et al.*, 2021)). However, this review is limited by only including the publications cited by the screening tools (layer 1) and not further investigating the citations made by the publications that were cited by the screening tools (layer 2). This has prevented identification of the original research used to develop the included tools. Despite this, multiple citations of the same publications occurred, with 36 of the 42 included screening tools developing their content from overlapping literature. Furthermore, cluster analysis split the core publications by the age ranges that the publications were aimed at. As age was the only difference between the three clusters it further suggests that the content included in these core publications also overlap with the differences occurring as the age increases. This raises some key questions: Why have so many tools been created that rely on the same original data? Why are new tools often citing previously published tools as a source for their item development? Are the tools measuring different but overlapping concepts? Or are they measuring the same concepts for different

underlying reasons? As the tools in this study were required to measure motor development in the first six months of life the concepts that guided their development must overlap to some degree, as is demonstrated by what the tools aim to measure. Further network mapping would identify the original research on which these tools have indirectly based their content and would help to determine if new tools were developed as new research was published.

However, it should be noted that full mapping of the citation network may prove to be difficult due to the changing standards of methodological reporting which served as a limitation in this study. Over time the requirements to publish methodological decisions have improved. The older screening tools included in this review, such as the NNE (Prechtl, 1977), provided little to no methodological input. For example, despite Prechtl publishing a large number of original research papers prior to publishing the NNE, by not providing this information within the NNE, this information was not included in this review . More recently developed tools, such as the ASQ-3 (Squires *et al.*, 2009) and PEDI-CAT (Dumas *et al.*, 2010), have given clear descriptions of their methodologies. Although this review is limited in its ability to describe the item development of all tools, it does show a progression to good reporting practices over time.

4.4.3 Conclusion

Overall this study found that items were most frequently developed from the literature. Due to different tools citing the same publications, the need for all of these tools is unclear and further research is needed. Similarly, the tools often cited previously published tools as their source for item development, and few included parents or caregivers in the development of the tool. As Chapter 2 demonstrated, parents identify the same and additional concerns than presented in the literature, further research should be carried out to identify if the concerns parents raise overlap with the items included on screening tools.

In this next chapter I will continue to analyse the tools identified in this review. I will show that the items used within these tools most often revolve around grasping, and reflexes and reactions; with head lift in prone being the most commonly included item. However, I will also show that the items they include do not address the range of concerns parents raised in Chapter 2, and that only the tools aimed at completion by parents use non-medical language that is accessible to all.

Chapter 5. How compatible are screening tools with caregivers' concerns? A comparison of caregivers concerns to screening tool items.

This chapter will compare the concerns raised by caregivers of children with Cerebral Palsy (CP) to the items included in screening tools developed to identify infants at risk of motor problems. In particular it will compare the content and the language used between the concerns and items. Previously this thesis demonstrated that caregivers of children with CP identify the same and additional concerns to those published in the literature (Chapter 2). Additionally, Chapters 3 and 4 identified only 2 screening tools to have included caregivers in the development of the item content, with most tools solely relying on literature and expert opinions. Inclusion of parents in the development of these screening tools resulted in the content domains being culturally sensitive due to the domains being developed from the concerns parents raised (Gladstone *et al.*, 2010a; Gladstone *et al.*, 2010b) and expanded the domains previously identified by the research team (Dumas *et al.*, 2010). To determine if the content of currently available screening tools covers the whole of early parental and caregiver concerns, I compared the content of the screening tools identified in Chapter 4 to the parental concerns identified in Chapter 2. In this chapter I will show that the most frequently included items were aimed at developmental milestones. I will also show that lexical differences occurred between the caregiver concerns and the tools aimed at completion by clinicians. Finally I will outline that none of the included tools covered the breadth of the caregiver concerns. I will then argue that these findings demonstrate flaws in the currently available screening tools and that either these tools need updating or a new tool needs to be developed.

5.1 Introduction

A part of good communication is a shared understanding through the use of shared language. Shared language allows for both individuals to communicate freely with little to no misunderstandings. In clinical practice, Health Care Professionals (HCPs) attempt to develop a shared language with their patients, allowing HCPs to translate between the patient's concerns and the medical models of disease (Thomas and McDonagh, 2013; Astbury, Shepherd and Cheyne, 2017). However, multiple barriers exist that can prevent the development of a shared language in primary care, such as time constraints and how

patients present their symptoms (Parker *et al.*, 2020). When the development of a shared language does not occur it can lead to delays in referral and diagnosis. Chapter 3 demonstrated diagnosis delays occurring within primary care due to caregiver concerns being perceived as 'brushed off 'or not being shared by clinicians. Furthermore, some caregivers reported having felt the need to collect evidence of their concerns by searching them online. The delays reported may be exacerbated by the lexical differences between the materials accessible to the clinicians and the way caregivers present their concerns. Chapter 2 identified lexical differences between the concerns parents raise and the reported signs of CP in the literature. As screening tools are one of the materials accessible to primary HCPs, comparing the lexicon to the caregiver's lexicon may identify if a barrier to shared language development is occurring.

Aims;

To compare the content and wording of parental concerns to the motor items within screening tools

5.2 Methods

5.2.1 Design

This is a qualitative comparison study between the items included on screening tools for atypical motor development in infants aged 6 months and under, as identified in Chapter 4, and a dataset of the earliest concerns reported by caregivers, as reported in Chapter 2.

5.2.2 Materials

Motor development items were taken from the screening tools identified in Chapter 4. When screening tools categorised their items, only the items identified by the screening tools as assessing motor development were included. When screening tools did not categorise their items all items on the tool were included.

The caregiver concerns consist of qualitative descriptions of the earliest concerns identified by caregivers in a free-text response survey outlined in Chapter 2. The survey identified three types of concerns; *day to day observations*, *developmental milestones*, and *troubling medical history*. Day to day observations describes concerns related to observations caregivers made in their every day interactions with their infant, such as parental instinct and difficulties dressing their infant due to tone in the infant's limbs. Developmental

milestones describes concerns regarding delayed development compared to pre-established motor milestones, such as sitting, or the infant attempting a milestone in an atypical way, such as crawling while one arm is trapped under their body. Troubling medical history contains caregiver concerns related to the infant's medical history, such as being born preterm and therapeutic hypothermic cooling, often drawing on more medicalised language. It was assumed that these concerns were influenced by interactions with medical professionals and medical information. Troubling medical history and developmental milestones were not included in this analysis due to the potential influence of medical professionals and infant development literature aimed at parents on the way parents describe their earliest concerns. The assumption was made that troubling medical history would not be included in the tools items, due to the infant's medical history being collected before an assessment takes place. A further assumption that developmental milestones would be included in the tool's items due to developmental milestones already being used as indicators of typical/atypical development.

5.2.3 Data analysis

The items from the tools were thematically analysed to group items into larger categories, such as grouping items carried out in prone together and items assessing reflexes together.

Going through each tool individually, the content of each motor item was compared to the content of each of the day to day concerns. When an item and a concern identified the same sign, the item was entered into a table (Microsoft® Excel®) against the concern it matches (vertical axis) and the tool it came from (horizontal axis). For example the concern of *parental instinct* is defined as a 'gut feeling' caregivers may have about their infant not developing typically but the caregiver may not be able to explain what is causing them concern. In contrast the concern of asymmetrical movement is defined as the infant moving one limb (such as the arm) more or less frequently than the other. If we take the item 'Does your infant move both arms and legs equally (symmetry)?' from the PediaTrac (Lajiness-O'Neill *et al.*, 2018), it would not identify parental instinct, but it would identify asymmetrical movement. Therefore this item would be recorded against the PediaTrac tool and asymmetrical movements concern.

Once all of the motor items had been assessed and recorded against the concerns, the table was split into tools aimed at HCPs and tools aimed at parents and caregivers for completion.

The tools aimed at parents and caregivers identified in Chapter 4 were the ASQ-3, CREDI, IYCD, MDAT, PEDI-CAT, PediaTrac, and PEDS:DMs (Brothers, Glascoe and Robertshaw, 2008; Squires *et al.*, 2009; Dumas *et al.*, 2010; Gladstone *et al.*, 2010b; Lajiness-O'Neill *et al.*, 2018; Lancaster *et al.*, 2018; McCoy, Waldman and Fink, 2018; Waldman *et al.*, 2021). Once split, the lexicon used in the items and the concerns were compared to identify which tools used a lexicon that reflected the language used by HCPs and which reflected the language used by caregivers. For example, tone is described medically using the terms hypertonia or hypotonia while caregivers use terms such as 'stiffness 'or 'floppiness'.

5.3 Results

Overall 336 items were identified across the 42 tools. The items were organised into 11 categories; prone, supine, sitting, standing, locomotion, hand use, eyes, movement quality and posture, reflexes and reactions, tone, and other, see Table 11. The items that make up these groups can be found in Appendix F.

5.3.1 Item frequency

The most common category of items reported in the tools were hand use (n=27) and reflexes and reactions (n=26). In contrast tone (n=19) was the least reported category between the tools. The categories reported by each tool is shown in Appendix G.

The five most frequently reported items were: Head lift in prone (n=22), Lifts and maintains head posture in sitting (n=21), Rolling from prone to supine (n=21), Reaching (n=20), Head lag when pulled to sit (n=20). All of these items measure developmental milestones that typically develop within the first 8 months of life.

5.3.2 Comparison of existing screening tool items to parental concerns

All parental concerns were covered within the items of the screening tools, see Table 12. However, tone was not assessed by any of the screening tools aimed at parents. In contrast, parental instinct was not assessed by any of the tools aimed at clinicians, as would be expected.

Comparison of the items demonstrated the tools aimed at completion by parents to use similar lexicon to the parental concerns. For example the item 'Is your infant putting hands, feet, or objects in his/her mouth?' in the PediaTrac uses the same language as a survey

participant did in describing her movement concern, ‘He would never grab his feet and put them in his mouth as the other children his age were doing.’ (M113)

In contrast, the tools aimed at completion by health care professionals (HCPs) often used more medical language such as ‘Volitional movement’ (MAI) or ‘Resistance to passive movement in upper extremities’ (Test of motor and neurological function).

Category	*Description*	*Example*
Prone	Items assessing the infant's development when the infant is in the prone position	'Controls head in prone' (Bayley III) (Bayley, 2006)
Supine	Items assessing the infant's development when the infant is in the supine position	'Active use of hips' (MAI) (Chandler, Andrews and Swanson, 1980),
Sitting	Items assessing the infant's development when the infant is in the sitting position	'Variability in sitting up behaviour' (Infant Motor Profile) (Heineman, 2010).
Standing	Items assessing the infant's development when the infant is in the Standing position	'Supported standing' (Amiel-Tison) (Amiel-Tison and Grenier, 1983).
Locomotion	Items assessing the infant's development when the infant is transporting themselves from one location to another, such as crawling, walking and running.	'Is your infant capable of moving from one place to another?' (PediaTrac) (Lajiness-O'Neill *et al.*, 2018)
Hand use	Item assessing the infant's ability to reach, grasp and manipulate objects	'Grasps from an easy position' (HAI) (Krumlinde-Sundholm *et al.*, 2017)
Eyes	Items assessing the infant's eyes, such as fixing and following, and nystagmus	'Looks for fallen spoon' (BINS) (Aylward, 1995)

Category	*Description*	*Example*
Movement quality and posture	Items assessing the infant's movement and posture that are not specific to a particular position, such as movement quality and arm posture	'Trunk rotations' (GMs) ((Einspieler *et al.*, 2004)
Reflexes and reactions	Items assessing the infant's reflexes and responses to reflex tests	Asymmetrical Tonic Neck Reflex
Tone	Items assessing the infant's tone.	'Arm traction' (HINE)(Dubowitz, Dubowitz and Mercuri, 1999)
Other	Describes items that did not fit into the above categories	Alertness and imitation

Table 12 The categories of items included in motor development screening tools for infants age term to 6 months

Parent concern	*Parental language used*	*Examples of items that address the concern from parent based tools*	*Examples of items that address the concern from clinical based tools*
Asymmetrical movements	'look at something to his left but reach over with his right.' (M028) 'Dominant side'	Does your infant move both arms and legs equally (symmetry)? - PediaTrac Does your infant move both arms and legs equally (symmetry)? - PediaTrac	Symmetrical movements – Denver II Asymmetry in movements – Movement Quality Measure.
Eye gaze	'He took a long time to follow the object during his 4 month Health Visitor check.' (M016) 'one eye turning in' M151	Does your infant follow objects with his/her vision? – PediaTrac When you face your baby, does he or she look at you, even if only for a little while? – PEDS:DMs	Eye movements – HNNE Eye muscle control – Harris Visual pursuit movements to object – Touwen
Feeding difficulties and physical development	'could not feed properly was the first sign at two months' – M126 'Struggled to with feeding within the first few weeks after birth, ended up with an NG [Nasogastric] tube at 12 weeks old due to failure to thrive.' – M253	Eating and mealtime progresses from swallowing pureed/blended/strained foods to opening sealed bags and boxes. – PEDI-CAT 'Does your baby open his mouth when he sees the bottle, breast or pacifier' – PEDS:DM	Sucking behaviour – Amiel-Tison Gag - NNDE Sucking reflex – Rapid

Parent concern	*Parental language used*	*Examples of items that address the concern from parent based tools*	*Examples of items that address the concern from clinical based tools*
Movement	'age 5 - 6 months, she didn't use two hands to take a larger object like a ball for example' (M067) 'He would never grab his feet and put them in his mouth as the other children his age were doing.' (M113) 'Legs didn't stop moving (constantly stretching them) and seemed in pain when touching his legs this was from about 10 months old.' (M093)	Brings hands together over chest, touching fingers. – ASQ3 Is your infant putting hands, feet, or objects in his/her mouth? – PediaTrac If your baby is lying on her back can she pass a toy from one hand to the other? – PEDS:DMS	Abnormal movements – Amiel-Tison Bimanual holding of an object – HAI Fluency of motor behaviour – Infant motor profile Spontaneous motility of the legs – Touwen.
Parental Instinct	'I new [*sic*] something was wrong when my daughter was born, but know [*sic*] physical signs' (M071)	As you begin to fill out this survey, how are you feeling about your infant? – PediaTrac Does anything about your baby worry you? - ASQ3	
Posture	'Awkward leg positioning' (G242) 'His grandmother noticed he was arching backwards' (M016) 'At 4.5mo we noticed he was keeping his right hand fisted and not using it' (M207)	Hand posture – ASQ3 Are your baby's hands open most of the time, not in a fist? – PEDS:DMS	Permanent closure of the hands - Amiel-Tison Arms at rest - HINE Abnormal postures - NNNS

Parent concern	*Parental language used*	*Examples of items that address the concern from parent based tools*	*Examples of items that address the concern from clinical based tools*
Reactions and reflexes	'very delayed reflex reactions (if any reaction at all).' (M066) 'From newborn- had severe startle reflex' (M096) '2 days old always turned to the same side (atnr)' (M011)	Palmar grasp – ASQ3	Startle – HNNE Deep tendon reflexes – EMP Protective extension – MAI
Temperament	'he cried and cried and cried, would not go into the pram and car seat without crying non stop' (M007) 'very unsettled and needy' (M104) 'very passive nature (he was too good a baby)' (M163)	'Is your infant able to console and comfort his/her self?' - PediaTrac	Irritability – HNNE Consolability – Neoneuro Cuddliness – NNNS
Tone	'We observed that she had mixed tone as soon as she was born … Very floppy and high tone' (F014) 'I had changed my friends daughters nappy and noticed how flexible her legs were. Originally I thought our child wasn't reaching milestones due to prematurity.' (M017) '6-7 months when I tried pulling her into a sitting position from laying down she wouldn't bend in the middle at all.' (M129)		Resistance to passive movement in upper extremities – Test of motor and neurological function Muscle tone consistency – MAI Estimate the appropriateness of neck and trunk tone for gestational age – NNDE

Parent concern	*Parental language used*	*Examples of items that address the concern from parent based tools*	*Examples of items that address the concern from clinical based tools*
Sleep	'Never slept from birth' (M093) 'Not sleeping, screaming all the time'	Does your infant sleep between feedings, do they have short periods of wakefulness? – PediaTrac	Unusual pattern of wakefulness – Amiel-Tison

Table 13 Reported parental concerns and the screening tool items that assess for them.

5.4 Discussion

The results demonstrated that no single screening tool addressed the entirety of concerns reported by caregivers. The caregiver concern of *Tone* was not assessed by the screening tools developed for use by parents, while *Parental instinct* was not assessed by the tools developed for use by clinicians. Although the screening tools developed for use by parents used similar lexicon to the parental concerns, this was not the case for tools developed for use by clinicians. The most frequently used item categories were *Reflexes and reactions*, *Reaching and grasping, and Sitting* despite head lift in prone being the most frequently used single item. Thus, the screening tool items focused on developmental milestones. No single tool had items that identified all the parental concerns.

5.4.1 Lexical differences

As expected lexical differences occurred between the parental concerns and the clinical based tools, but not between the parental concerns and the parent based tools. Fundamentally the clinical and parent based tools were developed for different populations. Clinically based tools were not developed to be presented to parents, as such no clinically based tools included parents in the development (see Chapter 4). Instead, specialist HCP opinions and clinical experience were typically used in the development of 15 of the 38 identified tools that reported the development process. However, these tools did not include PHCPs in their development. Although some PHCPs receive exposure to infant motor assessment during their training, such as trainee GPs being recommended to undergo a Paediatrics placement (The Royal College of General Practitioners and the Royal College of Paediatrics and Child Health, 2016), not all PHCPs receive the same level of training. For example, nurses from non-paediatric backgrounds can train to become Health Visitors. However, infant assessment is not a key criteria of the Standards of proficiency for specialist community public health nurses (The Nursing and Midwifery Council, 2004). As such some Health Visitors may not understand the language included in these tools. In contrast, during the development of the parent based tools, parents and caregivers were asked to comment on the language used in the 6 of the 7 tools development to ensure the items were understandable to lay people (ASQ3, CREDI, IYCD, MDAT, PediaTrac, and PEDI-CAT (Squires *et al.*, 2009; Dumas *et al.*, 2010; Gladstone *et al.*, 2010b; Lajiness-O'Neill *et al.*, 2018; Lancaster *et al.*, 2018; McCoy, Waldman and Fink, 2018; Waldman *et al.*, 2021)).

5.4.2 The problem with Developmental Milestones

All five of the most frequently used items across the parental and clinical tools measured developmental milestones, however the range of time for developmental milestones to occur may not make them best suited for early assessment. Developmental milestones are typically used to identify when an infant may be falling behind on a skill they should be developing. Developmental milestones were also identified by the caregivers who took part in the survey. However, developmental milestones have wide variations in the times that they occur between infants. For example, Touwen (1976) studied the development of low-risk typically developing Dutch infants. Despite being from the same population, Touwen identified that the ability to roll from prone to supine typically emerged between the ages of 3 months to 8 months, while crawling emerged between 4.5 months and 10 months. Due to these wide variations, milestones are not effective for identifying delays early within a population.

Furthermore, the development of milestones is influenced by the infant's environment. For example, Karasik *et al.* (2015) evaluated the ability of 5 month old typically developing infants to sit independently across different countries. The largest difference occurred between the Italian and Cameroonian infants, with only 17% of Italian infants and 92% of Cameroonian infants sitting independently at 5 months. During the hour long assessment Karasik *et al.* (2015) identified that infants in Cameroon spent most of their time sitting on the ground or on adult sized furniture, whereas the Italian infants spent little to no time on the ground or on furniture, but rather spent their time in their mothers arms. Another limitation of using milestones is the need to correctly adjust for prematurity based on the infants corrected gestational age. Without corrections premature infants will appear to be delayed in their milestones. Therefore, due to the wide variation in the onset of milestones, both within and between populations, any over reliance on specific milestones in early assessment is not optimal for identifying infants needing referral early.

5.4.3 Measuring the theoretical construct: Caregiver concerns

Importantly none of the included tools had item content that spanned the whole content of the caregivers' concerns. This could be due to not including parents and caregivers in the development of the tools. In Chapter 4, this thesis identified that 5 tools (ASQ3, CREDI, MDAT, PEDI-CAT, and PediaTrac) that included parents and caregivers in their development.

Only the MDAT and the PEDI-CAT reported including parents and caregivers in content development, however only the PEDI-CAT included a parent of a child with CP. As such, topics such as *tone* may have been overlooked as items that parents/carers are able to identify within the tools aimed at caregivers. This also raises the issue of content validity. Content validity relies on the entire theoretical construct being covered (Lynn, 1986). Items such as tone not being included may suggest that these tools do not have content validity for assessing parental concerns for CP.

In the UK the ASQ-3 is used with all families to monitor infant development at 9-12 months and at 2 years of age (NHS, 2020b). The ASQ-3 results are supported by in person developmental reviews carried out by Health Visitors at monthly intervals between birth and 6 months; bimonthly intervals between 6 and 12 months; and quarterly intervals after 12 months. In this setting it is clear that missing items such as tone may not have much impact due to the frequency in which the infant's development is reviewed. However, the COVID-19 pandemic had a large impact on Health visiting services. Interim findings into the impact of COVID-19 on primary care pregnancy and young families, identified Health Visitors to have experienced the highest rates of redeployment (Barlow *et al.*, 2020). Those not redeployed expressed concerns about their increased caseloads and their reduced their ability to identify infants who required additional support. Additionally, two thirds of the respondents reported having less than 10% of their contact with clients in the home or in clinic. Given the lack of in person contact it is likely that issues surrounding tone could have been missed, emphasizing the importance of including all relevant items. Creators of these parent-focused tools should look to incorporate new items to ensure content validity.

However, not including *parental instinct* within the clinician based tools does not raise issues with the content validity. Unlike the parent based tools, concerns about the infant's development will most likely have already been raised resulting in the clinician based tool being used. If so, having a parental instinct item would not add anything to the results. Instead these tools likely rely on the communication between parents, caregivers, and clinicians alongside the items within the tool to identify which items the infant struggles to complete.

5.4.4 Limitations

This review was limited by the retrospective nature of the caregiver concerns. This review cannot be certain that the concerns reported are accurate and have not been influenced by the caregiver speaking to HCPs. To overcome this, future prospective studies should look to periodically interview caregivers of infants at risk of developing CP about the motor concerns they have until a diagnosis is given.

5.4.5 Conclusion

In summary, this review demonstrated that the clinically-based screening tools assess all the expected caregiver concerns. However, parent-based tools do not assess tone. This finding could be due to the caregiver's ability to identify a potential concern with tone being over-looked. Future research should look to include all key stakeholders in the development of items within the tools to ensure all relevant content is included and that the tool is accessible to all users.

In this next chapter I will describe the development of two information sheets developed for helping new parents to identify the difference between typical development and atypical development consistent with CP. I will describe the four issues that arose during the focus groups; types of tools parents want, how and when they want to access information; How to improve parent understanding of atypical development; how much information parents want and need; and Managing disagreements between participants. I will also explain that two information sheets were created, rather than a screening tool, due to the difficulties participants described in trying to find information defining and explaining the difference between typical and atypical infant development.

Chapter 6. Developing new tools to identify infants at risk of Cerebral Palsy in the community. A participatory design study.

This chapter describes the development of two tools for identifying infants at risk of Cerebral Palsy (CP) based on the earliest concerns caregivers raise to primary health care professionals (PHCPs). Parents of children with CP identify the same and additional CP signs compared with what is reported in the literature (Chapter 2), however, few screening tools included parents in the design and development of the tool (Chapter 4) and none describe the full range of signs reported by caregivers (Chapter 5). Iterative interviews with key stakeholders were carried out with the aim to develop a new tool for CP screening within the community.

A screening tool was initially suggested due to my awareness of parents of children with CP commenting that they felt their concerns were not being heard by Health Care Professionals (HCPs) when raising them. Additionally, a screening tool was felt to be a cost efficient way to identify parental concerns that met the need for screening as described by Richardson (2018) and the All-Party Parliamentary Group on Cerebral Palsy (2021). This chapter will show that despite my assumptions, the participants did not want a screening tool consisting of questions about their infant's development. Instead, the parents suggested developing an information sheet that would help to shape their ideas and to help initiate and guide their conversations with HCPs. They felt it was the lack of informational resources which has caused delays in referral from primary care. The parents wanted accessible information available to them about a range of issues, both specific and non-specific to CP as well as information about typical development to help them discriminate when their infant is doing something atypical.

6.1 Introduction

Parents are usually the first to notice early symptoms of developmental difficulties in their infants and have been termed as 'lay epidemiologists' by Arksey (1994) due to their profound knowledge of their own child. Chapter 2 described the results of a caregiver survey of free text questions around the earliest concerns the caregivers had of their infant's development when their infant had undiagnosed CP. The caregivers reported additional

signs of CP not reported in the current literature. The content of a tool should encapsulate the entire theoretical construct the tool aims to measure. One way to ensure this is to include parents in the development of the tools content.

However, the inclusion of parents in any part of the development of screening tools is not common. Chapter 4 consisted of a scoping review into the development methods of currently available screening tools. The review identified 3 parent-based screening tools for infant motor development to have included parents in the development of the tool. However, only one of these tools, the PEDI-CAT (Dumas *et al.*, 2010), asked a parent of a child with CP to advise on the content of the tool during development. The parent was reported to have a child with Hemiplegic CP, which although is the most common form of CP does not represent the variation in severity and presentation that occurs within CP. Ideally, parents/caregivers of children with other types of CP could have been included to represent children with disabilities across the spectrum. Although new items could be added to the PEDI-CAT to ensure it covers the full content, an alternative is to develop a tool specifically for screening for CP early within primary care based on caregiver's concerns.

As discussed in Chapters 1 and 3, caregivers who are the first to identify their infant's first difficulties often report feeling that their concerns go unheard by primary Health Care Professionals (HCPs). When this occurs caregivers often begin a complicated journey through the primary care pathway, with accounts of caregivers attending up to five GP appointments before receiving a referral for their infant. A new tool should aim to reduce, if not stop, caregivers looping through the primary care pathway. However, parents of infants with CP are not the only stakeholders for this tool. This tool may also be presented to parents of typically developing infants and will be used by PHCPs. By including all key stakeholder opinions in the development of the tool, the tool can be shaped to fit alongside current practices in a way that supports all users. In turn this increases the chance of the tool being understood, adopted and normalised. Therefore, this chapter aims to develop a new tool for CP, based on the input from caregivers and HCPs.

6.2 Methods

This project involves a qualitative research study with 15 online semi-structured group and individual interviews with key stakeholders to obtain their observations of early signs of CP,

their opinions, and feedback on the development and design of a screening tool. Ethical permission was granted by the Wales Research Ethics Committee 7 (19/WA/0328).

6.2.1 Initial content.

It was decided to focus on the earliest concerns caregivers report as the tool itself will be aimed at helping identify those early concerns. Chapter 2 identified the earliest concerns caregivers develop when their infant has undiagnosed CP from a free-text online survey. The concerns identified in Chapter 2 were used as the initial content for the tool. The descriptions of the concerns were pared down to make the list more manageable. This involved removing direct quotes and making each description more succinct. During this process, concerns regarding the infant's troubling medical history were removed due to the medicalised language and the focus around treatments given at around the time of birth that would not be typical in infants discharged from hospital shortly after birth.

6.2.2 Participatory design process

Participatory design, or cooperative design, is a process in which the end users are actively involved in the design of the research or the product (Robertson and Jesper, 2013). Iterative approaches are often used to allow re-evaluation and feedback on any modifications of changes made over multiple cycles. The aim of this is to improve the quality and value of the research or the product to the end users. In particular workshops and focus groups allow for demonstrations and subsequent discussions around the materials that have been presented. A participatory design approach with key stakeholders was chosen to increase the chances of the tool being understood, adopted and normalised into primary care practices with new parents. Initially 5 focus groups were planned, 2 with parents of children with CP, 1 with secondary health care professionals, 1 with primary health care professionals, and 1 with parents of typically developing children. However, due to COVID-19 the focus groups were moved online. Due to potential issues with bandwidth and participants needing to be able to see one another the number of participants per group was capped at 5. In response, the number of groups increased, and one-to-one interviews occurred when participants were unable to attend a group session. However, the COVID-19 pandemic also affected recruitment, due to the additional pressures parents and HCPs faced, such as childcare and understaffing. This resulted in at most 3 participants per group, therefore from this point on

the focus groups will be referred to as interviews. Revisions to the tool were made iteratively based on the feedback from each interview.

6.2.3 Setting

The interviews were held online using Microsoft teams.

6.2.4 Participants inclusion criteria

The inclusion criteria consisted of:

1) Parents and/or caregivers of
    a. Children with a diagnosis of Cerebral Palsy.

       OR

    b. Typically developing children.

Or

2) HCPs who either:
    a. Work in a primary health care setting with infants.

       OR

    b. Work in a secondary health care setting with infants AND have specialist knowledge of CP (Such as a paediatrician, Paediatric Occupational therapist and Paediatric Physiotherapist).

For HCP groups, purposive sampling was used to ensure participants were from different professional groups, such as Health Visitors and GPs.

Originally it was planned to purposively sample parents and caregivers of children with different types of CP to ensure input from a range of experiences. However, due to a lack of interest this was not possible.

All participants gave fully informed consent to participate, demonstrated an ability and willingness to attend the interview, and were fluent in the English language. Fluency in English language was required to allow discuss between participants. Future research should look to include minority ethnicities access and/or include individuals with different first languages.

6.2.5 Participants exclusion criteria

Parents or caregivers who did not look after the child in question between birth and six months' corrected age (to ensure that the data collected focuses on the early signs of CP).

Parents and/or caregivers whose child is not typically developing and has not been given a diagnosis of CP or a CP subtype by a health professional (to ensure the data collected focuses on the infants who go on to be diagnosed with CP).

Health care professionals who work in a secondary health care setting who do not have specialist knowledge of CP.

6.2.6 Approach to participants

Potentially eligible parents were made aware of the study through their local parent carer forums using e-flyers and through online social media posting of survey e-flyers on Facebook and Twitter by the research team (appendix H).

Health Care Professionals were approached by their governing bodies (Royal College of Occupational Therapists; The Chartered Society of Physiotherapy), by their NHS Trust (The Newcastle Upon Tyne Hospitals NHS Foundation Trust) and Primary care facilities using e-flyers. Online social media posting of survey e-flyers on Facebook and Twitter by the research team were used to boost recruitment.

Participants were encouraged to share the e-flyer with people they knew who might be interested in the survey (snowball sampling). As parent carer forums, governing bodies, NHS trusts, and Primary care facilities shared the e-flyer through private communications I do not know how many people were made aware of the study.

6.2.7 Screening and consent

At the start of the interviews a member of the research team went through the information sheet with the participants and checked the participants were eligible to take part. If eligible and willing to take part, participants were asked to give verbal consent at the start of the focus group. Verbal consent was audio recorded using an encrypted Dictaphone and through the Microsoft© teams recording function. Participants were reminded that they could withdraw from the study at any time without giving a reason and that they did not need to share their video.

After giving consent participants were asked to complete a brief questionnaire indicating their sociodemographic information.

6.2.8 Procedure for Interviews

Participants were sent an information pack at least 48 hours before their interview for them to read and consider if they wanted to take part. Information packs included an information sheet, a consent form, and a sociodemographic questionnaire, these are shown in Appendix H. Ground rules were established and verbal consent was obtained from each of the participants at the start of each interview. The topic guide focused around the items included, such as the language used and if other items should be added, and the design of the tool, such as how it should be presented (i.e. questionnaire), and how the tool would fit into everyday life. Participants were asked if they would remove or add items, or if they would change aspects of the tool to better fit how they would use the tool.

The interviews were audio recorded and transcribed verbatim before being anonymised. Participant names were changed to pseudonyms.

6.2.9 Analysis

The anonymised verbatim transcripts were analysed using thematic framework analysis. The analysis was carried out by one researcher (JB) using a semantic, critical realist, inductive approach: This means that the data was analysed using the surface meanings and that no underlying ideas, assumptions, or conceptions were considered in the analysis; that I assumed that all participants experienced the same world but in different ways; and that themes were developed from the data (Braun and Clarke, 2006).

The emerging themes were presented at 'data clinics' where the research team shared their interpretations of the data. After the data clinics, amendments were made to the tool from the feedback of the previous focus group.

6.3 Results

A total of 25 people expressed interest in the study. One person stopped responding to emails before an interview was arranged, one became ill and was unable to take part, and one person did not meet the eligibility criteria (SHCP not working with infants with CP). In total 22 people (n=11 parents; n=18 women) took part. Only one person (Alice) took part in more than one interview. Information about the parents and their children, and HCPs is

shown in Table 13 and Table 14, respectively. 1 parent and 1 HCP did not complete sociodemographic questionnaires.

Participants were aged between 31-61 years (n=19, mean age 48 years, 1 participant gave an age range of 50-60 years). The majority were White European, with a university degree (Diploma = 1; GCSE or equivalent = 1), and were married, in a civil partnership, or were cohabitating with long term partner (Widowed = 1, Prefer not to say = 1). The HCPs had a mean of 28 years of experience (n = 9, range = 28 years); 1 participant reported '20+' years of experience). Ages of the children ranged from 20 months to 26 years. All but one of the children with CP have Hemiplegia. One parent of a child with CP, Riley (I12), also reported having a nephew who also had a diagnosis of CP.

Participant's first impressions of the tool were that it is 'definitely something that that is needed' (Bethany, IG1) and has 'genuine value' (Thomas, I15). The parents felt that there had not been enough information around typical infant development from a single reliable source when their children were born and were aware how they personally spent 'hours and hours googling everything, reading case studies and everything' (Alice, I1). This was because they had felt that they needed 'some evidence' to go with to the GP to back up their concerns. They felt like the tools gave them answers to their concerns and provided them with reassurance that their infants with CP were doing okay. This was because they could see that their infant had still completed some of the items that were typical which took them away from thinking of the worst case scenario to thinking that actually some of what their infant is doing 'is quite normal' and therefore it may not be as bad as they first thought. Even within the interviews, parents of children with CP began to remember things they noticed about their infant but had not realised it was to do with CP. For example Petra described how she had thought her son's tone was just his personality;

> Phineas was, was stiffer than the other babies um [Jess: yeah] and he did, when I had him in a sling, which I did because he cried a lot, um, I was always worried he was gonna fall out 'cause he was pushing himself backwards, um but I didn't really I just thought that was him. This is awful. I just thought that was him being difficult because he was such a crying baby and but but but in retrospect when I read the section I thought gosh

that just reminds me of this particular baby in the sling compared to the others (Petra, I10).

Petra was not the only parent to put their infant's CP features down to their infant's personality due to their lack of infant development awareness. Overall, the parents hoped that the tool would allow for an infant and their family to be helped sooner.

Participant	*Interview (I) number*	*Relationship to child*	*Childs name*	*Cerebral Palsy type or typically developing*	*Child's age*	*Age of typically developing siblings*
Alice	I1	Mother	Alex (m)	Hemiplegia	20 months	
Bethany	I1	Mother	Briar (f)	Hemiplegia	5 years	
Claire	I2	Mother	Cara (f)	Hemiplegia	16 years	18 and 14 years
Daisy	I2	Mother	NA (m)	Quadriplegia	10 years	5 years
Keira	I8	Legal guardian	Kali (f)	Typically developing	3 years	
Nicole	I9	Mother	Nina (f), Nathaniel (m), and Nico (m)	Typically developing	15 months, 5 years, and 7 years	
Noah	I9	Father	Nina (f), Nathaniel (m), and Nico (m)	Typically developing	15 months, 5 years, and 7 years	
Petra	I10	Mother	Phineas (m)	Hemiplegia	26 years	26 and 26 years (triplets)
Phineas	I10	Father	Phineas (m)	Hemiplegia	26 years	26 and 26 years (triplets)
Riley	I12	Mother	Alex (m)	Hemiplegia	2.5 years	7 months
Violet	I14	Mother	Rose (f)	Hemiplegia	16 months	
		Vivan	Vivian (f)	Hemiplegia	10 years	10 years (Twin)

Table 14 Information about the parent. Note: f - denotes female, m - denotes male

Participant	*interview (I) number*	*Profession*
Elaine	I3	Conductive Education teacher
Francis	I3	Community physiotherapist
Grace	I3	Conductive Education teacher
Heather	I4	Paediatric occupational therapist
Iris	I4	Paediatric occupational therapist
Jackie	I5	Paediatric speech and language therapist
Kaia	I6	Health Visitor
Lily	I7	Health Visitor
Madelyn	I7	Health Visitor
Sam	I13	GP
Thomas	I15	GP

Table 15 Information about the Health Care Professionals

The PHCPs also felt that the tool could easily be implemented into practice, such as for giving parents 'some information about what you might expect as your child's developing and what to look out for when to seek help.' (Sam, I13) or as something for PCHPs to discuss with parents.

From the original list of concerns developed in the survey, participants modified the language, presentation, and the item content in to two information sheets for parents (a short information sheet and a long information sheet) and a separate technical information sheet for PHCPs. Participants did not want 'another' screening tool regardless of if it listed the signs of CP or asked questions around these signs. Instead, the parents wanted information sheets that described the signs of CP alongside typical development, and included signposting and safety advice. From now on, I will discuss the developed tools as the short information sheet, the long information sheet, and the technical information sheet. A summary of the changes made after each interview is shown in Table 15.

The short information sheet, shown in Appendix I, describes concerns parents felt they would initially look for. This included concerns around: having a parental instinct that something was wrong; their infant's movements, such as the quality of the infant's movement being jittery; muscle tone and posture. A short description of developmental milestones was also included. Parents liked the short information sheet and thought it was useful, simple, and brief. They felt that the short version should be easily accessible to new parents, such as through inclusion in the Red Book, and provide sign posting to the full list of concerns presented on the long form. PHCPs preferred the short information sheet to the long one as it was to the point and contained 'signs that parents might not think about' (Sam, I13). However, there were differences of opinion regarding whether the short information sheet was short enough, despite HCPs being able to imagine themselves using the short information sheet with families. This was because some HCPs felt parents would not read more than one A4 page of information.

The long information sheet, shown in Appendix J, provides a more comprehensive range of the concerns raised in Chapter 2 and additional items added by parents and HCPs. Each CP sign is discussed in terms of what would be expected typically, and where relevant, signposting and additional information was included when signs overlapped with other conditions seen in typical developing infants (such as tongue tie). Parents liked and wanted to have access to all of the information. However, they felt that this should be something that new parents should have easy access to, but should not be the first thing they read, due to it potentially being overwhelming and stressful for a new parent to read. The PHCPs were split, with some feeling that the long information sheet was too removed from CP. Others felt it had sufficient depth to give to parents who had concerns and would give new parents a 'better footing' to start raising their concerns from.

The need for a technical information sheet for PHCPs was raised by parents and by one Health Visitor (Kaia). The topics for inclusion included; scientific information explaining what CP is, the causes of CP, the risk factors indicative of CP, a list of key early signs to look out for, and a list of potential referral pathways. However, the information suggested for inclusion overlapped with the current information given by the NHS (NHS, 2020a) and by the National Institute for Health and Care Excellence (NICE) (NICE, 2017a). Some of the PHCPs mentioned that they would be

happy to use the short and long information sheets or would rather have the key early signs to be implemented into the referral forms. Due to the overlaps with currently available advice and the lack of consistency in opinions across only 5 individuals, the technical information sheet will not be further discussed within the results of this study.

Interview with	*Changes made*
1- Two parents of children with CP	**To both information sheets:** Items were grouped under headings to aid navigation. Addition of an introduction to explain the reason for the information sheet. Inclusion of additional suggested items (babbling and head size). 'Strategies' or tasks parents could try, such as fixing and following were added. Link to birth to five book added due to it being a infant development resource previously given to parents in the UK and being referenced to within the Red Book.
2- Two parents of children with CP	**To both information sheets:** Two versions of the information sheet were created (a short information sheet and a longer information sheet) as parents felt having all of the information at once would be too stressful. Addition of further descriptions of atypical development and what milestones to expect in the coming years with a child with CP. Changing language around atypical development to 'alternative' development. Strategies and tasks removed as were deemed to stressful.
3- Three SHCP	**To both information sheets:** Approximate age guidance added to give parents a time frame of when to expect items to occur. **Long information sheet only:** Early communication section added. Elaboration added to explain what responses infants should have to certain items.
4- Two SHCP	**To both information sheets:** Increased the clarity of the items, simplified the language used, and changed the wording to prevent upsetting new parents. **Long information sheet only:** Items added (excessive weight gain due to limited movement, head control, moulding to the parents, reaction to touch). Items with negative connotations were rewritten. Examples changed to be more inclusive, for example within the temperament section the example of baby massage being a situation that might upset infants was changed to bathing. This was because bathing is more likely to be carried out by everyone whereas baby massage may not be accessible to all families.

Interview with	*Changes made*
5- One SHCP	**Long information sheet only:** Items added (variable feeding and difficulties weaning).
6- One PHCP	**To both information sheets:** Further simplification of the language used. More information added about when you would expect infants to complete the items. Items were changed from statements into questions to function as prompts. **Long information sheet only:** Inclusion of ICON safety message to the temperament section to bring the information in line with current guidance. Refinement of feeding for prolonged periods of time and the startle reflex to rule out typical variation.
7- Two PHCP	**To both information sheets:** Gentle introductions added explaining what the information sheets are for, reassuring parents what is typical, and explaining where to go if they do have concerns. Addition of images in the form of widgets*. Simplifying language so that it speaks to the parent in a friendly manner. Addition of typical development descriptions. Addition of the need for persistence of atypical items. Ages at which motor milestones typically develop added. Further explanation about due date added **Long information sheet only:** Order of the items on changed so that the most CP representative items are at the top. Signposting to further advice added. Wording changed to normalise crying and prolonged feeding. Addition of infant getting tired during feeding. Change weight and growth to clothing sizes. Remove labels such as 'too settled'. Remove birth to five book web-link as it is no longer used in England.
8- One parent of a typical developing child	**To both information sheets:** Captions added to the figures. **Short information sheet only:** Added headings into the introduction to help aid navigation.

Interview with	*Changes made*
9- Two parents of typical developing children	**To both information sheets:** Explain how to use this information within the introduction. Remind parents that these may be issues but are probably not issues for their infant into the introduction. Age related items highlighted in yellow. Clarification around what is atypical in terms of unilateral preferences. **Long information sheet only:** Added further information around breast feeding and latching difficulties. Added in about sudden unexpected changes in clothing sizes and the use of the Red Book centiles. Added in suggestions on what to do when an infant cries from ICON.
10 – Two parents of one child with CP	**To both information sheets:** Wording edited to ensure atypical items are clearly atypical and to ensure consistency across items. **Long information sheet only:** Further explained tummy time with image. Image of the Asymmetrical Tonic Neck Reflex (ATNR) added with an explanation.
11- One parent of a child with CP	**Long information sheet only:** ATNR wording made clearer.
12- One parent of a child with CP	**To both information sheets:** Explaining what words like coo and babble mean. **Long information sheet only:** Safety advice around temperament and sleep placed into red boxes to highlight it to parents. Link to advice on how to help a choking infant added as choking is an item within the feeding section. Explanation of what growth centiles are and how to use the ones in the Red Book. Within the Parental instinct section added that family and friends may be trying to help but if you are concerned to speak to a GP/HV. ATNR description further clarified. Responses header changed to better describe the content.

Interview with	*Changes made*
13 Part 1 – one PHCP	**To both information sheets:** Preamble explaining what the information sheets are for and to ask parents to get their infant checked out. Images were colour coordinated to the traffic light system used within National Institute for Health and Care Excellence (NICE) guidance, such as the guidance for fever in under 5s (NICE2019) **Short information sheet only:** Fixing and following added to match 6-8wk development check and to create consistency between the short and long information sheets. **Long information sheet only:** The need for persistence or suddenly occurring postures added. What is typical crying and what is not typical crying made clearer
14- One parent of a child with CP	**Long information sheet only:** Removed using a dummy from suggestions to soothe an infant as it has links to delayed speech development. Web-link to sleep training added due to the participants professional experience with new parents of typically developing infants having difficulties getting into routines resulting in sleep problems similar to the included items.
13 part 2 – One PHCP	**Long information sheet only:** Items added about emergency scenarios (turning blue, struggling to breathe, and head size increases suddenly) as participant felt it was something parents may look for. More clarity round eye gaze given. Added when infants start to learn to self-soothe within the sleep section to normalise sleep. Reworded feeding to explain the focus on CP signs rather than just feeding in general, such as removing specific information around breast feeding and providing a link to the NHS site on breast feeding instead.
15- One PHCP	**To both information sheets:** Explained what is meant by a 'word' in language development. Ages changed to represent when the last date we could expect infants to reach age related items.

Table 16 A summary of the changes made to the information sheets throughout the interviews.

** Widget refers to a simple but informative illustration.*

During the development of these information sheets four issues emerged from the data; types of tools parents want, how and when they want to access information; how parental understanding of typical and atypical development could be improved through the use of simpler language and the inclusion of images; how much information parents want compared to the amount of information PHCPs are willing to give them; and how disagreements between participants were managed across interviews.

6.3.1 Types of tools parents want, how and when they want to access them

Types of tool parents want

The parents felt strongly about the tools being information sheets about the signs of CP that they could access when they wanted rather than being a questionnaire or screening tool used by a PHCP. This was because they had experienced difficulties in determining if their infant was developing atypically. For example, Alice described spending 'hours and hours Googling everything, reading case studies' while going back to the GP 'so many times saying look I've read this, I know they tell you not to, but I've read this and it sounds exactly like what Alex's got.' (Alice, I1). In contrast, Bethany described that when her son was born, she was given a physical copy of the Birth to Five book, a book on how to care for an infant up to 5 years of age with signposting to relevant organisations. She described the book as being 'brilliant' as whenever she developed a concern she could turn to the relevant pages. Notably, the Birth to Five book is no longer used by the NHS. The parents agreed that they would have appreciated having a hard copy resource that they could have looked at when they previously had developed concerns and that they could have taken to the GP with them to 'back up' their concerns. Parents suggested having different methods of getting this information out such as having a concise version and a more detailed version online. This was because they felt all the information was important for new parents to have. However, they also felt being presented with all of the information in one information sheet would be terrifying and stressful for a new parent. The sentiment of needing a short concise version, and a longer detailed version was shared by the HCPs, who felt that giving parents all of the information at once would overwhelm them.

6.3.2 How parents want to access information?

The parents and HCPs suggested a wealth of ways the short information sheet could be given to new parents. This included baby groups, charities, the Healthy Start

programme, leaflets, parent classes, parenting tips, parenting websites, posters in public spaces, and wallet cards. However, the most common suggestion was to include the short version in the Red Book as 'everybody will get a Red Book' (Thomas, I15). They described how the Red Book is already a big part of a child's development as the Red Book 'talks about milestones'(Daisy, I2) as well being 'somewhere to keep everything' (Petra, I10) that parents are given. Some parents felt that if it was to 'make any sort of impact' (Violet, I14) it would have to be something that was in the Red Book that the Health Visitor went through with the parents as parents 'probably wouldn't read it' (Nicole, I9) on their own. Similarly, parents and HCPs felt that new parents would 'lose it' if the information sheet was in any other hard copy form.

The next most frequent suggestion was including it as a leaflet within the midwife or Health Visitor packs. Parents described how the packs contained things like their 'maternity notes (…) and then like leaflets about stuff. And all the info about this stuff. Like learn about. Sepsis, meningitis' (Nicole, I9) and that including the short version in that pack would increase the chances of them reading it. In particular, the idea of having a leaflet within the midwife or Health Visitor pack was preferred by those who felt their Red Book was not that big 'of a thing' (Riley, I14) for those that had not really engaged or used it.

However, the idea of the short version being a leaflet was not liked by all of the participants. In particular, Daisy highlighted that you 'just think you get so many bits of paper, just generally in life and as a new parent you get so many bits of paper.' (I2). Through having it as another piece of paper would result in it become lost or 'thrown away'. Instead, Riley suggested turning it into an A5 booklet;

> It would stand apart from a one sheet document. 'cause like I say, people go I'll give you this, I'll give you that and have a read through that. If I saw something a bit more substantial and it was in like a little book I'd be like I'll have a read of that. (Riley, I14)

In contrast, the long information sheet was only ever suggested to be some form of online resource. Either presented on a charity's website, parenting websites, or as part of currently available resources on child development.

6.3.3 When do parents want it?

Overall, there was no consensus between the parents about when they would want to be given the short form. Suggestions on when to give the information was split between during pregnancy and shortly after birth. Nicole thought 'It would have felt like a lot' (Nicole, I9) to have been given the information sheets as a new parent, and it 'would be very stressful' (Nicole, I9). However, she noted that the timing in which the parents were given the tools could negate some of the negative effects, such as giving the short information sheet out during pregnancy would give parents chance to digest and understand it. Most felt that they 'wouldn't have read it or remembered it' (Alice, I1) and instead they would look at a short information sheet if and when they needed it. However, parents on both sides felt that it 'wouldn't matter' when the information was given as different people interact with the leaflets they are given throughout pregnancy in different ways.

In summary, parents in this study wanted to be given as much information as possible due to having previously experienced difficulties in accessing the information themselves. They acknowledged that the amount of information was too much to be given all at once and multiple versions of difference conciseness were needed. Parents wanted a concise hard-copy tool that was included with other information about infant development, such as the Red Book or within maternity packs. They also wanted all of the information to be accessible online. However, parents were unsure on when would be best for a new parent to be introduced to the concise hard-copy tool.

6.3.4 Improving understanding

Although the initial list of concerns was developed using the language used by parents in the survey, changes to the wording of the information sheets were suggested multiple times to improve clarity and increase simplicity. For example, the occupational therapists felt the descriptions given by parents needed to be changed to reduce variation in interpretation and to be made clear enough that 'somebody who doesn't know (…) or has any experience' (Iris, I4) of infant development could understand them.

To further improve parental understanding of the items, the Health Visitors suggested including descriptions of what typical development is. They suggested changing the items to

explain what babies 'usually do' before giving the atypical item. For example, Madelyn suggested changing the item on delayed smiling to;

> babies usually start to smile back at you from eight weeks after their due date or before. [Jess: mm] When you smile at your baby. Uhm, if your baby does not smile back at you when you smile at them, then, you need to talk to somebody about that (Madelyn, I7)

The parents liked the inclusion of information on typical development as they felt that 'the more you know about what is normal, the better' (Peter, I10) and they recognised that they knew 'more or less nothing' (Peter, I10) about infant development when they first brought their infants home.

In addition to wording changes the Health Visitors suggested adding 'visuals' to the information sheets to help parental understanding. Visuals included photos, images, and videos demonstrating the items. As photographing and videoing infants for inclusion in the information sheets were not possible in the time frame left for this PhD, and I was unable to find photos that were royalty free and clearly demonstrated the items, in the next iterations I focused on providing images. Widget[8] style images were suggested due to being 'simple pictures'. Parents and HCPs liked the inclusion of widgets due to them 'very clearly' showing what was meant in the text, and 'because they focus on the position of the child and they take away any other considerations that you might have' (Petra, I10), such as what the child is wearing. Some parents felt that photos of real babies would be better as it 'makes it a bit more like personal (…) It's less clinical it's less [Jess: yeah; Noah: yeah] it's less scary' (Nicole, I9) making it more likely parents will look at the images.

Further clarification was also suggested for the growth section, however unlike the other sections this was to do with changes in health visiting practice due to COVID-19. Due to COVID-19 some NHS trusts stopped Health Visitor appointments taking place in person, with some parents reporting that they had 'never seen their Health Visitors' (Nicole, I9). Initially Health Visitors suggested using clothing sizes as a way for parents to gauge their infant's growth. Although the use of clothing sizes was generally accepted, some participants pointed out that some clothing brands are more stretchy than other brands and that may cause parents to panic. Additionally, during the UK lockdowns some parents had begun

[8] Widgets were created by JB using Paint.Net (version 4.3.4), an opensource image and photo editing tool.

weighing their infants to determine how much food they needed. Because of this, these parents taught themselves about how to use centile growth charts in the Red Book. This resulted in them suggesting to include information about changes in centiles within the long information sheet.

In summary, the participants wanted clear, simple and accessible language which is not open for interpretation. They wanted the atypical items to be supported by examples of what is typical for an infant to do with clear 'visuals' demonstrating what is meant within the text. However, parents were unsure if the images should be widgets or photos. Finally, due to changes in Health visiting appointments, Health Visitors and parents provided suggestions on items based on their experiences.

6.3.5 How much is enough?

Throughout the interviews there was a lack of consensus between the HCPs and between the HCPs and parents on what information should be included on each information sheet. HCPs and parents suggested items that could be included. HCPs based their suggestions on their own experiences. Often the items were straight forward, such as the infant staying in a clothing size longer than expected or including information on typical development. However, some suggestions demonstrated the variability and complexity of the signs some infants demonstrate and how parents can struggle to explain them. For example, Jackie highlighted that parents struggle to describe the variation in feeding ability in one day to the next;

> In the initial stages, initial kind of weeks, um, when any maybe cerebral palsy hasn't kind of. Developed, [Jess: yeah] that's the right word. um, it's often. It's almost babies. Feeding can be quite variable. [Jess: Yeah] So parents describe it as either some days are, absolutely fine, and they can take a bottle or breastfeed in just a matter of minutes, but then the next day it they can be feeding constantly. [Jess: yeah] So it's almost. I mean, I don't think you've put it there, but I think that in variability is is one of the big factors that parents talk about, (…) they know they from my point of view they can feed they, you know they can take things. Uhm, well at times, but it's that it's that it's that variability and sometimes parents are. They don't quite know where to go because. [Jess: It's not always. Yeah,] And typically if I go and watch them feeding that will be the time that they do it really well. [Jess: Yeah] Uhm, and that's not the parents everyday experience. (Jackie, I5).

The issues around feeding also extended to weaning where Jackie pointed out that some infants feed slowly from birth, but when they begin weaning feeding can become difficult for parents. However, there were disagreements between the HCPs over the relevance of the items. Sam pointed out the difficulty weaning 'can mean all sorts couldn't it? It could be like allergy to certain things' (Sam, I13). However, rather than add the stages of weaning, Sam suggested to include more general information such as if your 'baby is struggling or getting any rashes or anything um, speak, speak to someone' (Sam, I13) to prevent the long information sheet from becoming the 'Encyclopaedia of Childhood' (Sam, I13). Similarly, parents suggested items based on their experiences with their own child and other children with CP, which were later debated on. For example, Bethany suggested adding head size as an item as her own daughter and other kids with CP she had interacted with had 'huge heads'. However, HCPs were quick to point head size out as a measure already used at the 6-8 week check up and therefore likely not needed.

However, the inclusion of some of these suggestions led to some participants believing the short information sheet was too long. Suggestions to cut down the short information sheet included removing information about typical development and leaving it as a list of signs parents could look out for, as they could access the long information sheet if they wanted more information. However, depending on their speciality, HCPs argued that other topics should be covered on the short information sheet. For example, Jackie felt feeding difficulties should be elaborated on within the short information sheet as in her experience feeding is 'One of the first things that parents, talk about that, you know. If they're having difficulty' (Jackie, I5). However, the parents of children with CP felt that short information sheet should just contain an introduction, parental instinct, movement, tone, and developmental milestones as between them they 'more or less saying everything' (Claire, I2) without overwhelming new parents.

Similarly, the addition of these items left a couple of the Health Visitors feeling that the long information sheet was not specific enough to CP. Suggestions to increase the long information sheet's specificity included; removing the sections that overlapped with typical development; changing the order of the sections so that the more representative items were first; including parameters such as the signs needing to persist over time and explaining what typical development usually looks like within each section. After changing the section order

and including the parameters, no further comments were made about the long version being too non-specific. Instead, the GPs felt that new parents with concerns 'would need that level of information' (Thomas, I15). They also suggested including non-CP items around what they would look for in clinic, such as advising parents to speak to a GP if their infant started developing diarrhoea and a rash after feeding. However, they did recognise that the long information sheet is 'not supposed to be a parenting manual so' (Sam, I13) the amount and depth of the information included should come down to what the parents want.

Even when asked about removing sections the parents of children with CP did not want anything removing as they felt it was 'best just to have all the information' (Alice, I11). They felt the long information sheet covered all of their concerns and a lot more. They also felt the items were presented in a way that was subtle enough that it was not obvious it was about CP and would not scare new parents. The parents may have wanted this level of information included within the long information due to the difficulties they had in finding information. Parents of children with CP spoke about how they had 'googled' their infant's symptoms and read case studies to identify if their infants' signs were atypical so that they could present this information to their GP as evidence for needing a referral.

However, parents of typically developing infants also described having difficulties with the currently available information presented online as suggesting that their infant was delayed in their development. In particular, Nicole mentioned having used the Wonder Weeks app[9]. Nicole explained how her own typically developing son was 2 weeks delayed according to the app and that she had 'had to pull some people off ledges' (Nicole, I9) as their infants were also being classified as delayed on the app. Nicole mentioned that the app had been developed on a relatively small sample of infants all from white 'upper class families' with 'no diversity'. Although Nicole was aware that the Wonder Weeks App was not representative of every infant's development, she acknowledge that not every parent has the same level of awareness of infant development. As such, Nicole's experiences suggests there is a need for general guidance about typical development which is available to all, in

[9] Wonder weeks initially started as a book on infant development aimed at new parents written by Dr Frans Plooij and Dr Hetty van de Right in 1992. The Wonder weeks content was developed from Plooij and van de Right's own research on infant development. More recently Wonder weeks was developed into an app for IOS and android.

addition to very clear specific information on CP signs that clearly separates what is typical from atypical.

It should be noted that PHCPs felt there needed to be guidance around if they are 'just looking for one of these signs, or is it a combination of signs?' (Thomas, I15). As 'one thing in isolation probably doesn't mean they've got Cerebral Palsy' (Thomas, I15). Yet the parents of children with CP felt differently as they felt that all the items 'if they're handled early enough, can have a, a significant impact on the child's outcome' (Bethany, I1) regardless of whether the item is directly related to CP.

Overall parents in this study wanted to be given as much information as possible, however they did not want it all at once. To begin with they wanted to be given clear descriptions of the key CP signs as to not overwhelm them. Different HCPs had different ideas of what should be counted as a key sign, however everyone agreed that parental instinct, movement, tone, and developmental milestones were the key signs to include on the short information sheet. Although the Health Visitors felt the long information sheet was too general, the GPs and parents did not want to remove any of the content, but instead suggested including more general items that new parents should seek help about.

6.3.6 Managing disagreements

Unlike the previous issues, managing disagreements focuses on how participant suggestions and disagreements were managed. As participants took part in interviews, rather than focus groups, the immediateness of feedback on a participant's idea by other participants was lost. To determine if a participant's suggestion would be worth pursuing, their idea would be presented to the next interview. For example, after the idea of a PHCP tool was first suggested by Alice and Bethany (I1), the idea was then raised with each of the PHCPs.

Sometimes this led to agreement and sometimes this led to disagreement. For example, the parents tended to agree on the topics included in the short and long information sheets. However, despite the parents agreeing that on what should be included in the long information sheet, Lily and Madelyn felt that the long information sheet was 'non-specific' which could lead to the floodgates opening in terms of what 'a parent might worry about which is massive' (Madelyn, I7). When disagreements occurred, participants were presented with the reasoning given by previous participants. Often this led to the current participant(s)

offering compromises. For example, after explaining why the parents of children with CP wanted that level of information presented in the long form, Lily and Madelyn suggested: putting the 'big hitting' items first; signposting to relevant information so that parents could try using the currently available advice before deciding on if they need to seek help; normalising aspects of items that overlap with typical development, such as crying; and explaining how infants usually develop. All four solutions were aimed at alleviating unnecessary worry without removing the information requested by parents.

Sometimes disagreements between participants resulted in the suggestion not being carried forward. For example, Briar suggested including 'strategies' that parents could do with their infant to test their infant's function before seeking help. An example of what one of these strategies may look like was initially added to the information sheet for the next interview. However, the parents in the next focus group completely disagreed with Briar's suggestion, due to the chance of it being overwhelming for new parents and due to the potential for the strategies to cause harm if carried out incorrectly. In response, the idea of including them was dropped.

Finally, some suggestions were not carried forward to the next interview due to disagreement from the research team. For example, Elaine and Grace suggested including signposting to conductive education centres, so that new parents could seek therapy separate to the NHS process they may be going through. The research team disagreed with this suggestion due to conductive education centres not being accessible to all new parents, due to issues such as locality.

Overall, the suggestions participants made were generally presented to the next interviewee(s). Sometimes this led to agreements and further building onto the suggestion. Sometimes it led to compromises, and sometimes it led to removal of the suggestion from the information sheet(s). Occasionally participants gave suggestions that were not taken forward due to the research team identifying issues that would result in the suggestions removal at a later stage.

6.4 Discussion

This chapter developed two new information sheets aimed at screening for infants with CP in the community, based on the contributions of parents of children with CP, parents of typically developing children, PHCPs, and SHCPs. Across the interviews there was a lack of consensus about what information should be included, when to receive the information, and how to receive it.

6.4.1 Information needs of new parents

A screening tool was initially suggested rather than informational resources for three reasons. Firstly, previously collected data showed that some parents felt that whilst they had identified and reported worrying signs in their child, PHCPs had not acted promptly on those concerns (Basu *et al.*, 2015). Nothing in this data set suggested that parents did not have informational resources to support their concerns, therefore a resource could be created to help support PHCPs to identify parental concerns indicative of CP. Secondly, screening tools are already successfully used within primary care. For example, Health Visitors currently use the Edinburgh Postnatal Depression Scale (EPDS) to screen for postnatal depression in new mothers. Health Visitors have described the EPDS to help them in their role due to their lack of knowledge around mental health (Vik *et al.*, 2009). They have also highlighted that the EPDS has allowed them to identify cases that they would not have identified through talking alone. Additionally, they explained that giving the EPDS to all new mothers allowed them to 'sell' the EPDS as creating opportunities to 'create the best conditions for mother and baby' (Vik *et al.*, 2009, p. 241), reducing the stigma around mental health. These same benefits could occur if a screening tool was added to assess early development; reducing the need for training on early CP signs, reducing the need for Health Visitors to identify early signs through talking, and reducing the pressure for parents to speak up about their concerns. The third reason is due to the current recommendations, such as a UK wide CP register and the re-implementation of a screening service, being expensive to implement and, realistically, need to be implemented at the national level (Richardson, 2018; All-Party Parliamentary Group on Cerebral Palsy, 2021). However, the UK government believes that decisions around community based infant care, including screening programs, should be decided at the local level, which have restricted funds (Department of Health and Social Care, 2019). Therefore, a cost-effective approach to early screening in the community was needed to prevent inequality across the UK.

However, the parents in this study did not want another screening tool, but rather access to information on early CP signs. The parents identified that when trying to get help for their infants, they were unable to access resources to help them identify if their infant needed medical attention. Because of the lack of informational resources, some parents in this study did not identify all of their infant's signs. For those who did find resources, they took the resources with them to the GP to ask if what they were seeing could be what the resource described. Some of the parents attributed their 'looping' through the primary care pathway to not having the informational resources available to effectively state their case, aligning with the findings of Chapter 3. This led to the parents wanting informational resources rather than a screening tool.

The suggestion of easy-to-understand information sheets is not new, due to parents having previously been provided with little information on other topics. In 2014, Jones *et al*. carried out semi-structured interviews and focus groups with parents of children under 5 years of age and HCPs treating children under 5 years of age in the UK. The study looked at the informational needs of parents looking after acutely ill children. As with this study, the parents requested the information be presented in an information sheet or on a single website. They suggested including the information within the information packs provided to new parents and that the information is presented using basic language and audio-visual materials, such as pictures and videos. The consistency in the responses given by participants is likely due to the difficulties both sets of parents reported in identifying relevant and easy to understand online resources in a timely manner (Jones *et al.*, 2014; Neill *et al.*, 2015).

The biggest issue to arise was about how specific to CP the long information sheet should be. The parents of children with CP wanted as much information about CP signs to be included as possible. This was because they had experienced difficulties in determining if their infants' signs were atypical and they wanted it to be easier for new parents to identify their infants' signs. Similarly, one parent of a typically developing child highlighted that the apps used by new parents, such as the Wonder weeks (thewonderweeks.com), tell parents what their baby should be doing in that week of their development, with little to no guidance around the typical variation in how babies develop. Across the apps and websites parents mentioned using, none of them contained information about atypical motor development.

Even the NHS website does not contain information around atypical development that is not linked to a specific condition (NHS, no date). This means that parents must have an idea about the condition their infant has before they can look up the symptoms. Although some NHS Trusts do have information around atypical development, such as Kent Community Health NHS Foundation Trust (no date) , this is not standard across the UK.

In contrast to the parents, the Health Visitors did not want parents to be given lots of information, due to the potential of overwhelming services. When conflicts arose, such as this, the reasoning given by the previous participants was given to the current participant(s). In this example, the Health Visitors were informed that the parents of children with CP had asked for all the information to remain as they felt it was important for new parents to be aware of all the potential symptoms. In turn the Health Visitors provided solutions to allow the parents to have this information without potentially overwhelming services.

By taking into consideration all stakeholder opinions, the long information sheet became a more generic information sheet; the need for more information around infant development is not unique to parents of children with CP. A recent Delphi survey of parents and health care professionals in Australia came to a consensus that within the first postnatal year new parents should be provided with information on (but not limited to): choking and suffocation; crying; developmental milestones (such as fine- and gross-motor skills); feeding problems; sleep; and social and community support (such as parenting groups and asking for help) (Cashin, Wroe and Campbell, 2021). Similar findings have also been shown by Slomian *et al.* (2017) in Belgium. Both studies were carried out with the plan to develop perinatal psychoeducational materials and an informational website dedicated to the postnatal period, respectively.

6.4.2 Candidacy

This need for more information around atypical development alongside typical development provides further support for the theory of candidacy (Dixon-Woods *et al.*, 2005; Dixon-Woods *et al.*, 2006). As explained in Chapter 3, candidacy is the dynamic process in which individuals assess their eligibility for medical attention and how they legitimise their interaction and engagement with services. The lack of clear guidance around typical and atypical development together makes it difficult for parents to identify if their infant has any atypical signs. By not knowing what is atypical, parents search for advice

about their concerns rather than seeking medical care, as shown in Chapter 3. Additionally, parents are unable to identify the extent of the signs, with some parents of children with CP commenting that the prototype information sheets developed in this study made them realise their infants had more signs of CP than they had attributed and reported. Again, this adds to the difficulty parents face in identifying their infant's candidacy. Furthermore, by not identifying, and therefore not reporting, all their infant's CP signs, PHCPs are left with a partial picture of the infant's needs. In turn, PHCPs may be delaying referrals to ensure that the care given to these infants is necessary, as expected as part of their gatekeeper role.

6.4.3 Limitations and future research

It should be noted that there was also a lack of consensus between the parents, and between the parents and HCPs. Parents were unsure if they would prefer to receive the short information sheet before, just after, or shortly after giving birth. Similarly, they were unable to decide whether it would be better for the short information sheet to be presented as a leaflet or as part of the Red Book. Similarly, as explained above, parents and HCPs disagreed on the level of information parents should be given. However, once the parents' viewpoint had been explained, the HCPs began to suggest compromises.

There are two reasons these disagreements may have occurred. Firstly, this is most likely a limitation of having used interviews rather than workshops or focus groups. Focus groups are communal activities, typically used to understand different viewpoints and pull together a consensus on a topic (Myers, 1998). In contrast, interviews aim to understand more deeply about an individual's decisions (Miles and Gilbert, 2005). Because of this, it is likely that the participants' ability to scrutinise different viewpoints and bounce ideas off one another was lost, resulting in a lack of consensus. Additionally, the lack of consensus between parents may also be due to the parents not feeling strongly about how and when the information is presented to them, as they are likely to only access the information sheets when they need them. Further research is needed to determine when and how to disseminate the short information sheet. However, this research needs to be carried out collectively. This could be done either through workshops, focus groups, or through pilot testing with all stakeholders working together.

The need for a HCP technical information sheet on CP suggests that ongoing training for PHCPs is needed. As described in Chapters 2 and 3, Action CP (2016; 2018) identified

significant gaps in primary health care training opportunities around CP, with some Health Visitors receiving no CP training after qualifying as a Health Visitor. In particular it was the Health Visitors in this study who suggested that the development of a HCP tool would help aid training of newly qualified Health Visitors. This lack of training may be causing a lack of awareness which is resulting in the delays to referral.

This study has three further limitations; the sample were all white Europeans and none of the parents of children with CP reported their child as having a Tetraplegic CP diagnosis at the time of the study. As explained in Chapter 3, infant motor development is directly affected by the infant's environment (Karasik *et al*., 2015, Touwen, 1976). Because only white Europeans took part in the study, there is the chance that the information included in the information sheets may not reflect the breadth of environments infants in the UK are exposed to.

The lack of tetraplegic CP representation also may affect the information sheets contents. As explained in Chapter 1, CP is an umbrella term, and infants with different topographic limb impairments may present in different ways. This limitation may have been mitigated by the inclusion of SHCPs who have experience working with infants across the CP spectrum and by developing the initial items from survey data that included parents of children with tetraplegic CP. However, there is no guarantee that the information included in the information sheets accurately reflect the initial concerns of parents whose infant has emerging tetraplegic CP.

Similarly to Chapter 2, the parent sample was over-representative of those with university degrees (81.8%) compared to the general population (33.8%) (Office for National Statistics, 2023). In turn, this suggests that the sample is over-representative of higher Social Economic Status (SES) and therefore unlikely to be representative of the wider CP community. Solaski, Majnemer and Oskoui (2014) systematically reviewed studies on SES and CP prevalence from Greece, Ireland, Kosovo, Malta, Sweden, UK, and USA. They identified that SES is negatively correlated with CP, even when confounding variables, such as multiple births, are controlled for. Lower SES is associated with lower income, lower education, poor housing, increased health care needs, and increased barriers to research participation, such as feeling unqualified to take part and requirement for additional carer time to aid participation (National Institue for Health and Care Research, 2020). It is possible that informational tools

may not be what families from lower SES want or need and such tools may not be accessible to those with lower literacy abilities. Additionally, the experiences shared may not reflect the difficulties that those from lower SES face when trying to seek care for their infant.

The lack of individuals from lower SES may be explained by the recruitment strategy and the inherent biases of carrying out interviews. Parent carer groups, Governing bodies, NHS trusts, and Primary care facilities who were asked to share the survey often used private communication, such as private social media pages and email, causing them to act as gatekeepers. The term 'Gatekeepers' in this context refers to an individual who controls access to a privately owned space, and so would have been able to decide on who is made aware of the study. As the gatekeepers used private communications, I am unable to tell if anyone who was eligible to take part in the study was stopped by a gatekeeper either not informing them or coercing them to, or not to take part. However, it is unlikely that coercion did take place. As such I cannot accurately determine the referral rate for this study.

As described above, participants from lower SES often experience more barriers to participating in research. As participants were informed that the interview would last around an hour, some may not have been able to guarantee that time due to work schedules and child care, which are two barriers known to impact those from low SES (National Institue for Health and Care Research, 2020). Future research should aim to recruit participants through the NHS to allow for proportional selection of participants.

Further research is needed to refine, finalise and validate the information sheets, however this research could also be used to overcome these two limitations. Once the short information sheets design is finalised, the sensitivity and specificity of both information sheets for identifying infants with emerging motor difficulties needs to be determined. During both stages, researchers could use purposeful sampling to ensure diversity across the parents, around ethnicity as well other potential factors, like educational attainment, are included and that the CP spectrum is represented.

6.4.4 Conclusion

Parents did not want a list or question like tool, this was due to having difficulties in identifying if their infant needed medical care and then presenting their infant's candidacy to

their PHCPs. During the development of the two information sheets, issues around the availability of information to new parents was raised and the impacts it may have on candidacy considered. Parents feel like the information around CP symptoms is not easily accessible and current guidance does not tie typical and atypical development together. The long information sheet was made more general to reduce potential overwhelming of NHS services where items overlapped with similar issues seen in the typically developing population. However, in some topics, such as how and when to disseminate the short information sheet, there was a lack of consensus. This is likely due to the methodological limitations of interviewing as participants were unable to debate their viewpoints. Further research is required to bring clarity to topics in which consensus was not reached and to validate the two information sheets.

Chapter 7. Final discussion

7.1 Introduction

In this chapter, I will bring together the findings of my thesis and consider their implications. This thesis looked at the concerns parents of children with emerging Cerebral Palsy (CP) have (Chapter 2), their experiences of the primary care referral system (Chapter 3), how motor screening tools for infants age term to 6 months corrected age were developed, if parents were involved in their development (Chapter 4), and how the items within these tools relate to the concerns raised by parents (Chapter 5). Finally, this thesis described the development of two information sheets using participatory design and the issues that arose (Chapter 6).

The first section of this chapter will present the findings of this thesis in relation to their impacts on different audiences. I will begin with the impacts on parents and caregivers of infants with CP. I will then describe the implications for primary health care professionals. I will then also discuss my findings in terms of the broader health care literature around early CP identification. Following this, I will reflect on the research process, discuss the future of CP identification in the community, and finally, I will discuss potential areas for future research.

7.2 Candidacy at the level of the parent

This thesis explored the experiences parents and caregivers of infants with emerging CP have when approaching primary care services with their concerns. Some participants reported having difficulties in identifying their infants' initial symptoms or that they were unaware that their infants' signs were not typical (Chapter 3). As these participants were unable to identify their infants' signs, they did not act, resulting in delays. Although some parents did develop concerns they downplayed their observations, or they doubted the observations they were making and so did not act on their concerns until others, such as family and friends, raised their concerns with them (Chapter 3). Upon developing concerns about their infants some participants reported continuing to observe their infants to confirm their concerns, some reached out to family and friends for advice, while others searched their concerns online, again to confirm their infant warranted medical care. Although these proactive approaches resulted in participants delaying attending primary care, it also built these participants' confidence in seeking out medical care for their infant which potentially resulted in them attending primary care earlier than if they had not been proactive. Finally,

some parents and caregivers who had identified that their infant warranted medical care delayed booking appointments due to their fears of being labelled, such as being a worrier, by HCPs.

However, these reasons for delaying medical care are not unique to CP and suggest underlying factors influencing parental appraisal of their infants' signs and their help-seeking behaviours. Similar reasoning has been described for other paediatric conditions such as Arthritis (Kirkpatrick *et al.*, 2018), Cancer (Dixon-Woods *et al.*, 2001; Clarke *et al.*, 2014; Pedersen *et al.*, 2020), and Diabetes (Usher-Smith, Thompson and Walter, 2013). A likely reason for these underlying factors is Dixon-Woods *et al.* (2005) candidacy. Candidacy is a seven-stage dynamic process in which patients negotiate their need for medical care between themselves and their HCPs (Chapter 3). The seven stages consist of; identification of candidacy, navigation of services, permeability of services, appearance at service, adjudication by HCPs, offers of resistance to services, and operating conditions and local production of candidacy.

In particular, the reasons parents and caregivers delayed attending services can be explained by identification of candidacy and appearance at services. Identification of candidacy describes how individuals come to recognise their symptoms require medical attention (Dixon-Woods *et al.*, 2005). As described above, parents and caregivers struggled to identify their infants' signs and when they did identify them, they doubted their observations or downplayed them. Appearance at services describes an individual's ability to assert their candidacy for medical care (Dixon-Woods *et al.*, 2005). To be able to assert their candidacy, individuals are expected to be able to formulate and articulate their concerns and present them to be credible. Again, as described above, parents and caregivers felt the need to develop their case by asking for advice, seeking out additional information, and continuing to observe their infants' signs to present as credible. The need to present as credible also prevented some participants from reaching out early due to fear.

It should be noted that some parents who have family and friends with experience of CP were able to present their concerns early and received an immediate referral. For example, Violet spoke to a physiotherapist she worked with who referred her daughter on immediately (Chapter 6). Similarly, even though Riley was in denial about her daughters' symptoms her sister, whose son has CP, approached her about the similarities in their

development. By having a knowledgeable person close to them these parents were able to identify their infant's candidacy and then assert their infant's candidacy.

During the interviews reported in Chapter 6 parents of children discussed what would have helped them to identify their infant's candidacy earlier and support them to assert their infant's candidacy. From the list of concerns developed in Chapter 2, the parents felt that new parents should be provided with all the information on the concerns list as well as information about typical development to help new parents decipher what is typical and what should be a concern. This was because these parents had experienced difficulties in finding information about atypical development to legitimise their concerns. However, some parents felt all of the information would be too much but felt that it should still be available for parents who want it. As a result, two information sheets were developed; a short information sheet that contained the key signs of CP that new parents would see first, and a long information sheet that provides further information about all of the signs of CP. To prevent causing parents unnecessary worry, general advice, normalising of typical infant behaviours (such as crying), and signposting to alternative support services were incorporated into the long information sheet. The inclusion of these strategies was appreciated by the parents of children with CP and the parents of typically developing children. In particular, both groups appreciated the addition of information about typical development to help them to decipher what is atypical from typical.

Parents wanting more information is not new. Recent studies of parents and health care professionals have demonstrated parents in their first postnatal year should be provided with information on (but not limited to): choking and suffocation; crying; developmental milestones; feeding problems; sleep; and social and community support (Slomian *et al.*, 2017; Cashin, Wroe and Campbell, 2021). Additionally, multiple studies have shown that some parents of typically developing children report wanting to be given more information about infant development than they currently are (Schuster *et al.*, 2000; Combs-Orme, Holden Nixon and Herrod, 2011; Lång, Tell and Johansen, 2021).

A big issue that was raised in Chapter 6 was the amount of information being given to parents. Health Visitors felt that by giving parents lots of information about CP signs, more parents would raise concerns that could potentially overwhelm services. When the parents' requests were explained to the Health Visitors, the Health Visitors suggested strategies to

include to prevent overwhelming services. However, when Graybill *et al.* (2016) provided parents with an informational booklet on infant developmental milestones, the chances of parents seeking medical attention did not increase. Instead, Graybill *et al*.'s (2016) mixed-methods randomised control trial found that giving parents more information about developmental milestones improved their understanding of child development and reduced their concerns about their child's development. Although some parents reported the informational booklet alerting them to potential areas of their infant's development that could become a concern, the rates at which participants raised concerns remained the same across the groups and between pre- and post-trial. Although Graybill *et al.* (2016) did not measure the impact the booklet had on parental mental health, parents did find the booklet to be helpful and informative, and helped 'empower' them to raise their concerns. As such, providing parents with the information they request may help them to identify their infant's need for medical care and feel empowered to raise their concerns, without potentially increasing service use.

7.2 Candidacy at the level of Health Care Professionals

The findings above describe what candidacy looks like at the level of the parent. This is not always where candidacy occurs as the process is also influenced by the parents' interactions with HCPs.

Chapter 2 identified that infants identified within primary care are significantly more likely to receive therapy and diagnosis later than their counterparts identified within secondary care. Additionally, the earlier parents and caregivers raised their concerns the more likely they were to experience delays in receiving a referral. These findings match those reported in Canada (Hubermann *et al.*, 2016; Boychuck *et al.*, 2020).

In Chapter 3, parents and caregivers described their experiences with PHCPs when reporting their concerns about their infant's development. These parents and caregivers described three reasons for delays that were given by the PHCPs they approached; their concerns were not shared by the HCP, or they felt their concerns were 'brushed off', the PHCP giving an alternative reason for the infant's signs, and the PHCP choosing to take a 'watch and wait' approach. Some of these participants reported attending primary care multiple times before a referral was made. In particular, one parent reported speaking to their GP 5 times before a referral was made. It should be noted that these findings only represent the experiences of

parents and caregivers as GPs and Health Visitors were not surveyed. These findings may also demonstrate gatekeeping by PHCPs as well as issues in communication between the PHCPs and the parents and caregivers. Despite the lack of PHCPs viewpoints, these reasons for delays have also been demonstrated in other paediatric conditions (Dixon-Woods *et al.*, 2001; Clarke *et al.*, 2014; Kirkpatrick *et al.*, 2018; Pedersen *et al.*, 2020), again suggesting underlying factors, such as candidacy, influencing the referral process. However, the parents and caregivers in this study felt that the PHCPs awareness of CP determined whether they received an immediate or a delayed referral.

There are three potential reasons for this: PHCPs have a lack of training around CP, parents are not reporting all their infants' signs and therefore PHCPs need to probe further, or PHCPs are gatekeeping, resulting in some watch and wait approaches being used. Freedom of information requests of UK Local authorities, Clinical Commissioning Groups, and NHS trusts have shown little CP specific training occurring within primary care (Action CP, 2016; Action CP, 2018). Training for CP was often included within generic disability training, with training being reported to occur biannually, sporadically, or only upon request. Similarly, only 19 of the 56 trusts that responded had developed or were developing formal CP pathways in line with NICE (2017a) guidelines. However, it should be noted that the NICE (2017) guidelines of possible early features of CP are not exhaustive and do not demonstrate the breadth of concerns raised by parents and caregivers in Chapter 2. This raises the question do PHCPs have enough training and knowledge around early CP symptoms to be able to question parents about symptoms not raised, and to effectively identify infants with emerging motor difficulties that require therapy. Alternatively, PHCPs may be following good clinical practice by implementing watch and wait approaches to ensure the referrals they make are required. However, these questions cannot be answered until the primary HCPs view is more formally considered through new research .

If PHCPs do not have the required training and knowledge this again leads to issues of candidacy. In the model of candidacy, stages three, permeability of services, and five, adjudication by HCPs, describe the ease in which individuals can access services and how an individual is judged by a HCP on if they deserve medical care respectively. This thesis did find a high level of gatekeeping; however, this may be an artifact of the sample population. Similarly, multiple studies have demonstrated GPs use key signs of a condition to determine

if a referral is needed immediately (Molassiotis *et al.*, 2010; Usher-Smith, Thompson and Walter, 2013; Kostopoulou *et al.*, 2019). Those whose concerns do not meet the GPs criteria receive a delayed referral. Furthermore, as GP's positive predictive value for referral increases, their discrimination ability does not change. Instead, Kostopoulou *et al.* (2019) identified that GPs increase the criteria required for referral.

This thesis has also shown parents do not report all of their infants' signs. While reading through the information sheets, parents of children with CP reported additional CP signs that their child had presented with that they had not associated with the other concerns they had raised (Chapter 6). As explained in the previous section, this is likely due to the limited information available to new parents around infant motor development. Again, by not being able to identify all of their infant's signs, new parents' ability to assert their infants candidacy is reduced. Similarly, PHCPs must then question parents and caregivers who are reporting concerns on aspects of CP. However, if parents and caregivers are unable to answer these questions, their ability to assert their infant's candidacy to gain a favourable adjudication by their HCP is reduced. Alternatively, if PHCPs are not knowledgeable about CP, they may not know the questions to ask, resulting in them making their decisions on limited information. It is likely that it is the combination of HCP awareness, and the limited concerns parents are raising which are resulting in the delays.

Standardised training and the development of materials around CP or just highlighting motor development could help PHCPs to identify and quickly refer on infants with emerging motor difficulties. In 2011, the Royal College of Paediatrics and Child Health (RCPCH) identified that fewer than 25% of GP trainees undertake a paediatric placement during their three-year training programme. To improve the level of care available to infants and children in the community the RCPCH and the Royal College of General Practitioners (RCGP) called for interprofessional training for GPs (O'Dowd, 2016; RCPG and RCPCH 2016). Giving specific training on early CP symptoms may also help to increase awareness and improve the level of care available to these families. Similarly, during the interviews, PHCPs commented on the need for a HCP information sheet on CP to help improve their knowledge, understanding, and ability to identify the earliest signs (Chapter 6). Although this HCP information sheet was not developed, some of the HCPs felt that the short information sheet provided the key signs they would need to look out for and that it would be useful to have access to the same

information given to the parents. As such, versions of the two information sheets could also be deployed to primary HCPs to help with CP identification.

7.3 Implications for wider research

The previous sections have described the key findings in terms of parents and caregivers of infants with CP and the HCPs treating these infants. I will now discuss the findings in reference to their implications for research on early CP identification.

A running theme in this thesis is the lack of parents of children with CP being directly engaged with in research. In the UK, research is expected to include potential participants in the design, conduct, and analysis of the research, rather than only being the subject of the research (Department of Health, 2005). Chapter 2 shows that the breadth of CP signs is not accounted for in the literature due to parents not having been included – or their inclusion not reported on - in these previous publications. Chapter 4 demonstrates only one tool for identifying infants with motor difficulties to have included a parent of a child with CP in the development process. Chapter 5 demonstrates that the tools aimed at completion by parents do not consider all of the concerns a parent of a child with CP may report. The lack of parental involvement is likely due to the research in these publications being carried out prior to the broader trajectory of patient and public involvement in research becoming more mainstream.

Similarly, parents and caregivers also reported feeling unheard by GPs (Chapter 3). Listening to parents is a basic part of communication and if patients are feeling unheard there is likely some breakdown of communication. As parents of children with other paediatric conditions have also reported feeling unheard, there is likely a wider communication issue at play (Dixon-Woods *et al.*, 2001; Usher-Smith, Thompson and Walter, 2013; Clarke *et al.*, 2014; Pedersen *et al.*, 2020).

Similarly, some parents are asking for more information about infant development, both in this study and in the wider literature. The findings of Chapter 3 replicate wider findings that some new parents seek out additional information about typical infant development from their friends, family, and online sources to identify if their infant is developing typically (Price *et al.*, 2017; Aston *et al.*, 2018; Moon *et al.*, 2019; McLeish *et al.*, 2021). When using online sources, these parents are often described as critically analysing sources through their

comparisons with other sources and the beliefs of those around them. Despite having access to reliable services online and through their HCPs, parents still want access to additional information around infant development again from a reliable resource (Schuster *et al.*, 2000; Combs-Orme, Holden Nixon and Herrod, 2011; Lång, Tell and Johansen, 2021).

Across this thesis and this chapter, the theory of candidacy has been prominent. Undeniably, in the UK context, there will always be gatekeeping, as not all parent's and caregiver's concerns will require referral. Additionally, as seen in Chapter 6, there is a lack of consensus about what information new parents require, and how to disseminate this information due to fears of overwhelming new parents and NHS services. As such, regardless of the changes and improvements made, there will never be a perfect system in which all parents and caregivers whose infants have emerging CP receive an immediate referral. However, until the delays in the community are reduced this will remain a problem.

The results of this thesis, along with those from other paediatric conditions (Dixon-Woods *et al.*, 2001; Usher-Smith, Thompson and Walter, 2013; Clarke *et al.*, 2014; Kirkpatrick *et al.*, 2018; Pedersen *et al.*, 2020) demonstrate underlying factors for delay occurring within primary care that need to be tackled. As discussed before, candidacy provides a potential explanation for the delays reported at both the parental and HCP levels (Dixon-Woods *et al.*, 2005). Improvements need to be made that incorporate the voices of parents alongside HCPs.

7.4 Reflections

In this section, I will provide some of my reflections on this thesis, including the limitations and strengths of the research.

The main strength of this research is the inclusion of parents and caregivers of children with CP. The concerns parents of children with CP initially raise have not previously been described in the literature. Additionally, the concerns they raised matched the signs of CP previously reported in the literature by HCPs (Garfinkle *et al.*, 2020) and added additional signs. Similarly, Chapter 4 demonstrated that parents of children with CP had only previously been included in the development of one motor screening tool for infants age from term to 6 months corrected age. During the interviews in Chapter 6, some parents expressed views that more information around typical and atypical development is needed, rather than a

new screening tool. This was so parents could quickly identify if their infant is showing atypical signs and feel confident in presenting their concerns to their PHCPs. The need for more information was also supported by parents of typically developing children. Although the parents felt that a new screening tool was not needed, the distribution of accessible information may act as a proxy screening tool for delayed development, in which CP is one cause.

The biggest limitation of this thesis is the use of retrospective data. The data described in Chapters 2 and 3 relied on participant recall of their earliest concerns and their experiences of the primary care referral process. Retrospective data is often fraught with recall bias and systematic errors that occur when participants do not remember previous events accurately or omit details. This reduces the accuracy of the data collected. In addition, some of the concerns reported used medicalised language not typically used by lay parents, suggesting that the concerns were influenced by the HCPs and experience of the healthcare system around the participants. However, this data was then later used in Chapters 5 and 6. In Chapter 5, day to day concerns were used to compare the items included in the tools to the earliest concerns parents report. In Chapter 6, day to day concerns were also used to form the first version of the tool. However, it should be noted that due to delays that occur between symptom onset and diagnosis for CP, and the infrequency of parents reporting concerns within primary care that go on to be diagnosed with CP, a retrospective approach was needed. Similarly, the legitimacy of the day-to-day concerns were confirmed by parents of children with CP and specialist secondary HCPs during the initial interviews carried out in Chapter 6.

Additionally, the data used in the Andersen Model (Walter *et al.*, 2012) analysis to look at total patient delays was not collected for that purpose (Chapter 3). At the time of data collection, I was unaware of the Andersen model. However, upon reading the responses on why parents and caregivers had a good or bad experience of the primary care referral pathway I noticed that often they were describing types of delays. As such, the delays reported in this thesis may not demonstrate the range of delays experienced by parents and caregivers who raise their concerns in primary care. Similarly, the analysis of this data provided no evidence for treatment delays. Without the data having been collected

specifically for analysing total patient delay it is unclear if treatment delays do not occur due to infants receiving therapy before a diagnosis of CP or if it is an artifact of the data.

The participants who took part in the survey (Chapters 2 and 3) and the interviews (Chapter 6) are subject to self-selection, response bias, and recall bias. As participants were able to self-select, this thesis likely represents the individuals on the more extremes of opinions who wanted to share their stories. Similarly, participants may have reported what they thought the studies were trying to identify rather than their own experiences and opinions. Additionally, the participants in this thesis were largely white females and from higher Social Economic Statuses than is representative of the general population (Office for National Statistics, 2023) and of the CP community. Finally, these studies relied on recall of caregiver's experiences, which given that some participants were describing events from over 20 years previous, it is likely that they information given is not completely accurate. As a result, the results of this thesis likely do not represent the wider experiences of parents and caregivers of infants with CP across the UK. Similarly, Chapter 3 did not include PHCPs opinions on the primary care referral process for CP.

This thesis is also limited by the lack of HCP opinions throughout and the use of realist inductive approach to thematic analysis means that the interpretation of caregiver comments was not considered in terms of the pressures and working conditions HCPs experience. For example, in Chapter 3, the lack of HCP voices means that it is likely that there are alternative reasons for delays such as PHCPs being required to reduce referrals to secondary care. Similarly, in Chapter 6 only two GPs and three Health visitors took part. As different NHS trusts have different processes, it is likely that their experiences do not reflect the wider experience of PHCPS. As such, the experiences reported by parents and caregivers may reflect good practice and/or misunderstandings between the participants and their PHCPs. For the same reasons, the tools developed in Chapter 6 may also not be appropriate for use by all primary HCPs.

There are also methodological issues within Chapter 6 due to the use of interviews. The development of the screening tool was originally planned to happen through focus groups. This was to allow participants to debate their viewpoints and come to a consensus on how best to develop and disseminate the tool. However, due to the COVID-19 pandemic and the pressures the UK lockdown put onto parents and HCPs, the study switched to using

interviews to allow for flexibility. However, interviews do not allow for different viewpoints to be debated directly between participants as they would be in a focus group (Miles and Gilbert, 2005). To overcome this I did present previous participants reasonings to current participants. However, this only allowed the current participant to consider their own view and the view of those before them, rather than being able to discuss the viewpoints. Therefore, it was difficult to develop a consensus between participants due to using interviews. Assuming a consensus on these topics could be met, further research is required. Future research should take some form of synchronous or asynchronous focus groups to allow for a consensus to be produced.

The final limitations focus on the poor historical citing habits in older publications and the lack of time available to trace the citation history of the screening tools included in Chapter 4. Part of the aims of Chapter 4 was to understand where items of motor screening tools were being developed from. However, the older screening tools included in the review provided little to no citations of the work they developed their tools from. For example, Prechtl and Beintema (1964); and Prechtl (1977) did not provide any citations for how they developed the Neonatal examination (NNE), despite it being clear that the NNE was developed based on Prechtl's publications at the time. Because of this, the history of how these older screening tools were developed cannot be traced.

The lack of time available to research the citation pathways of the included tools was due to the age of the publications and their location. This thesis only included the publications cited by the screening tools (layer 1). Further investigation was carried out, however, a large number of publications cited were only accessible within the British Library and inter-library loans were not available. Additionally, the British Library has restrictions on the number of publications you can access per day. Because of this, I would have needed to spend the equivalent of around 3-4 months in the British Library just to review the publications known to me at the time I decided to just use the layer 1 citations. By not further investigating the layer 2 citations (citations made by the publications that were cited by the screening tools) the extent to which the tools developed their items from overlapping literature cannot be identified.

7.5 The future of CP identification in the community

Since starting this PhD, there have been several advancements towards improving identification of infants with emerging CP in the community.

In March 2021, an All-Party Parliamentary Group (APPG) report on Cerebral Palsy in the UK was published (APPG 2021). The report focused on early identification, intervention and care pathways of infants and young children with CP. In total 11 recommendations were made. Of these, 7 directly relate to this thesis. In general, the recommendations incorporated funding and ringfencing streamlined pathways between primary, secondary, and tertiary care across the UK that follow NICE guidelines. They recommended additional funding to increase the Health Visitor workforce and to add in three new standardised Health Visitor appointments at 3-5 weeks, 3 months, and 3.5 years. In particular, the 3-month appointment was recommended to be used to assess infant motor development. Additional recommendations included providing additional training on the early signs of CP and associated neuro-disabilities in infants to all non-specialist PHCPs, the inclusion of information around atypical development within the Red Book, and the development of a national CP register.

Following this report, Action CP released their CP awareness campaign 'If in doubt, check it out' in March 2022 (Action CP, 2022). The campaign aims to raise awareness of CP by providing new parents with the key signs of CP and advising parents to speak to their doctor or Health Visitor if they see any of the signs in their child. The content of the campaign was developed by expert clinicians, senior practitioners, public affairs specialists, trusts and foundations, and, most importantly, parents.

In addition to the campaign, Action CP began to petition to have information about atypical motor development included within the Red Book. This information being put forward by Action CP aligns with the information on typical and atypical development that the parents in Chapter 6 reported wanting to be presented in the red book. Additionally the content of the Red Book is currently under review while the whole of the Red Book is being developed into an online tool. Although some NHS trusts are already using the online Red Book, it is currently thought the Red Book will be fully digitised by 2023. If Action CP are successful, it is likely that the online Red Book will provide new parents with the information they require to determine if to seek further help.

After completing the development of the short information sheet, I have been in contact with Action CP to discuss my findings and the short information sheet that I had developed. Currently, Action CP are looking to use the findings of this thesis to further support needed policy changes to early identification and referral of infants presenting with early signs of CP.

7.6 Areas for future research

The research I carried out opened up additional questions that I feel need answering. This final section introduces some areas I believe would benefit from further research.

Firstly, this thesis found evidence of underlying factors influencing paediatric referrals from primary care suggestive of candidacy (Dixon-Woods *et al.*, 2005). The findings of Chapters 3 and 6 identified 6 of the 7 stages of candidacy occurring in primary care referrals of infants with emerging CP based on parent and caregiver recall. These same stages were identified in paediatric Arthritis (Kirkpatrick *et al.*, 2018), Cancer (Dixon-Woods *et al.*, 2001; Clarke *et al.*, 2014; Pedersen *et al.*, 2020), and Diabetes (Usher-Smith, Thompson and Walter, 2013) literature. However, the data presented in Chapters 3 and 6, and in Kirkpatrick *et al.* (2018), Dixon-Woods *et al.* (2001); Usher-Smith, Thompson and Walter (2013); Clarke *et al.* (2014); and Pedersen *et al.* (2020) was not collected to be analysed for candidacy. Although they demonstrate aspects of candidacy, to fully understand the effects of candidacy, either in one of these conditions specifically, or across paediatric primary care in general, a prospective study should be carried out. This would also allow for identification of aspects of paediatric primary care that could be targeted for intervention to reduce referral delays. A prospective study would likely take the shape of a series of qualitative interviews with families recently referred to paediatric services within secondary care from primary care, and with the PHCPs that referred them. The interviews would be semi-structured with questions focusing on the seven stages of candidacy. Including PHCPs would also allow for a better understanding of the reasons why PHCPs delay referrals, as well as the frequency in which parental concerns, reported within primary care, require referral.

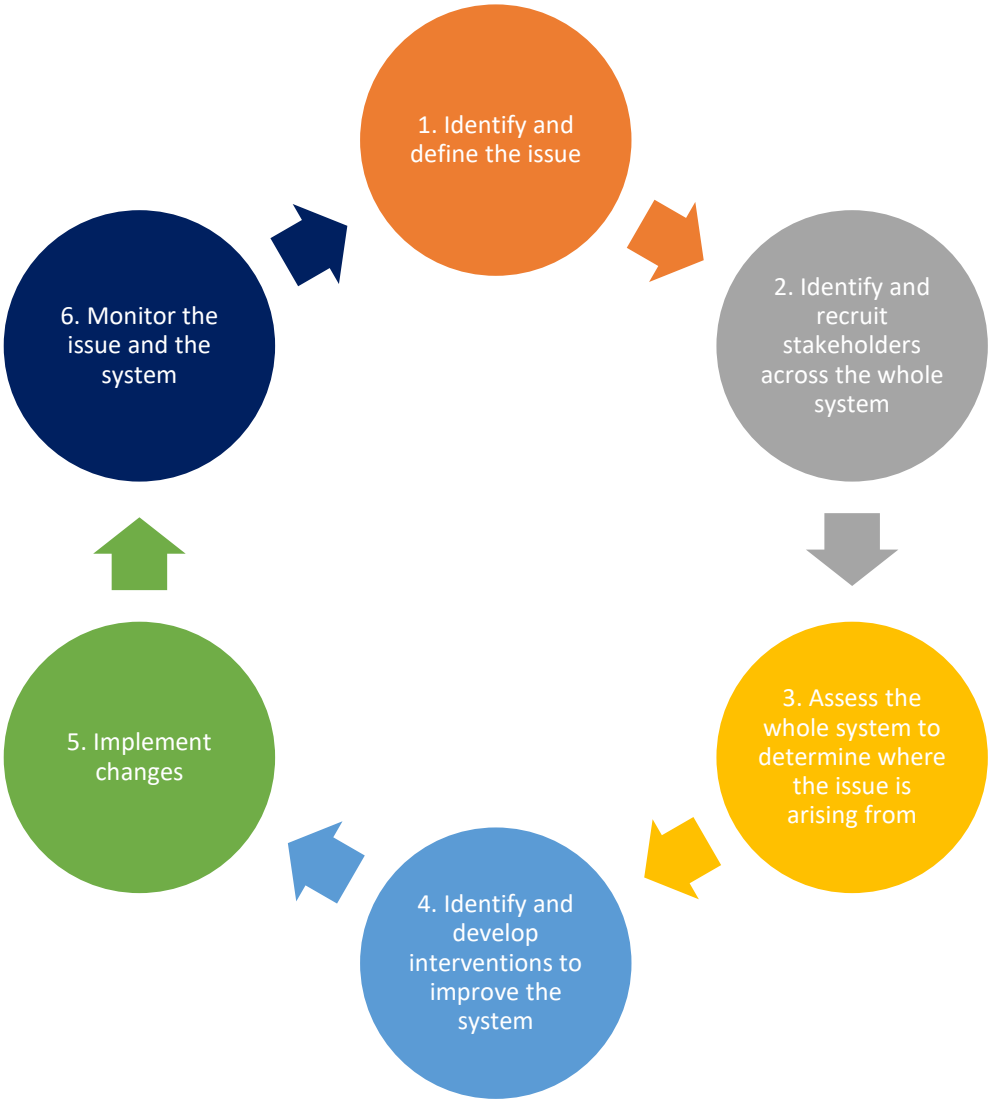
Secondly, this thesis identified that a large proportion of currently available motor screening tools were developed from the same literature. As explained in the last section I was unable to go more than one layer deep into the citation network. As such it is unclear if more of the tools included in the scoping review have used the same original data source. Further exploration into the citation network would allow the original data sources to be identified

and would highlight which motor screening tools were developed from the same data. Further exploration could also examine why so many motors screening tools have been developed.

Thirdly, further piloting and focus groups are required to clarify what information is needed, what format should be used, when would be best to give new parents the short information sheet, and who should receive it. The interviews carried out in Chapter 6 occurred during the COVID-19 pandemic which made recruitment difficult due to the added pressures on parents and on health care professionals. This resulted in what was originally planned to be focus groups turning into interviews. Focus groups had initially been planned to allow for participants to come to a group decision on what the tool should look like, however this was lost as conflicting opinions were not present. To improve recruitment in further piloting, it would be worthwhile recruiting families through the NHS rather than through parent carer groups and social media. This would allow me to directly contact families about the study and allow for purposeful sampling, ensuring a more representative sample. Additionally, I would want to run further piloting using asynchronous focus groups. Asynchronous focus groups are online focus groups that do not require all participants to be present at the same time (Gordon *et al.*, 2021). They work similarly to social media where participants are able to make comments and ask/answer questions posed by the researcher or other participants at a time that is best for them. Asynchronous focus groups have been shown to have a number benefits that may not just help recruitment but agency and the quality of data (Williams *et al.*, 2012; Lally *et al.*, 2018; Wirtz *et al.*, 2019; Gordon *et al.*, 2021). Firstly, they have been shown to make engagement in research feasible for participants with irregular work schedules, family responsibilities, or other needs that may make in-person groups difficult to attend. Secondly, they allow for participants to digest information and questions before responding. Not only does this allow participants time to think, but it can also prevent participants from domineering the conversation. Additionally, it gives space for participants to go on tangents or have 'side-conversations' without disrupting the group. Thirdly, participants can take agency in choosing their own online name/handle and therefore how they are represented in research. In turn, this can increase participant comfort with sharing sensitive information due to the anonymity they can choose to give themselves.

Fourthly, this thesis developed two information sheets for new parents (Chapter 6). Before the information sheets can be distributed, they need to undergo validation within the general population. Additionally, since the parents could not decide when the information sheets should be given this also needs to be assessed. Validation and the timing of distribution could be determined within a single between subjects study, with four arms. The control arm would receive standard care, while the first test arm would receive the short information sheet as part of the maternity pack given to pregnant women. The second test arm would receive the short information sheet alongside the Red Book, and the third test arm would be given the short information sheet a couple of weeks after their infant's birth by their Health Visitor. Randomisation of families to arms could happen through cluster randomisation based on the local NHS trust. Similarly to Graybill *et al.* (2016), this study would then assess the following: parental knowledge of infant development before receiving the information sheet and when the infant turned 1 year old, where parents accessed information from, the rates in which parents developed concerns, spoke to HCPs about their concerns, and initiated referrals. The study would also examine parental perceptions of the information sheets, and their potential effects on parental wellbeing. Together, this information would provide a picture of the current sources of information parents use and the effects of additional information on parental wellbeing, and service use rates.

Finally, the use of a whole systems approach to improving CP identification in the community may provide a better, more sustainable system for CP identification. Whole systems approaches look at the system, the individual parts that make up the system, and how these individual parts work together to make changes that have positive impacts on an agreed upon issue (Charnley, Lemon and Evans, 2011). Whole systems approaches can be split into six stages, as shown in Figure 8. Firstly, the issue must be identified and defined so that the key stakeholders can be identified. Subsequently, stakeholders from across the whole system must be recruited. Next stakeholders need to work collaboratively to assess the whole system, each of its parts, and how those parts work together to identify the reasons that are resulting in the issue. After having identified what requires change, they then need to develop an intervention, or interventions, which would have a positive impact on the issue. Once these interventions are implemented, the system requires continuous monitoring to ensure the intervention is working, to identify if other parts of the system



would benefit from intervention, and to identify if other issues have arisen. Because the system requires monitoring, the whole systems approach results in an iterative cycle of improvement.

Figure 8 The process of a whole system approach to intervention.

This thesis has shown the current system for CP screening in the community to have flaws. Examples include; a lack of PHCPs knowledge and awareness of CP, a lack of tools within primary care to screen for CP, a lack of informational resources for parents, and infants looping through primary care resulting in delays to diagnosis and therapy.

A whole systems approach would require individuals involved in: primary care practices; decision makers at local, regional and national levels of the NHS; local and national government; NICE; as well as the Royal Colleges responsible for training Health Visitors, GPs, and specialists in Paediatric and Child health care; alongside people with CP; their parents and guardians; and the range of voluntary and community sector organisations engaged with CP to collaborate to begin to explore existing and new ways to engage with the issues described above. A whole system collaboration could result in multiple changes that overcome the issues described in this thesis. Examples of the changes that could occur are; changing the training requirements of PHCPs so that infant motor assessment is a key component. Changes in the way primary care services are structured, such as all Health Visitors being able to refer infants to secondary care or to community physiotherapists. Health Visitor roles being revised so that motor assessments become part of their role, such as the GMs. Changes in the requirements for follow up screening in secondary care. Changes to the NICE guidelines to include more accessible screening tools and more descriptive early CP signs. Government policy could be changed to ring-fence funds for the development of time-critical referral pathways to triage services for motor testing, similar to pathways used for patients with suspected Cancer. Finally, the development of a toolkit, encompassing not only the parent information sheets development in this thesis but additional information and tools to help PHCPs.

7.7 Conclusion

Overall, this thesis adds to the previous literature by exploring the factors that result in infants with emerging CP receiving a delayed referral from primary care and by developing new information sheets to overcome these delays. The reasons given by parents and caregivers for delays occurring are reflective of Dixon-Woods *et al.* (2005) theory of Candidacy. They also suggest an issue with communication between HCPs and parents, which aligns with current reports on primary care CP training. Specific research into candidacy within paediatric primary care referral may highlight opportunities in which interventions could be implemented to improve services for all paediatric patients.

The screening tools aimed at motor development do not cover the breadth of parental concerns and only the PEDI-CAT included a parent of a child with CP in the development of the tool. These findings are limited due to poor referencing in the older publications.

However, further research into the citation network may identify why so many tools have been developed and what the original publications used are.

Key stakeholders felt that information sheets would provide the best way to improve the identification of infants with emerging motor difficulties. The information sheets developed consist of; a short form containing the key signs of CP that new parents access first, and a long form containing all CP signs as well as general information about typical infant development and signposting. The biggest issue raised was the amount of information being given to new parents through the long information sheet. However, in addition to the parents included in Chapter 6, the need and want for further information about motor development is also reported by some parents in the wider literature. Further research is needed to validate the information sheets, however the process of implementing information about atypical motor development has started due to campaigning by Action CP.

Chapter 8. References

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Chapter 9. Publications and presentations

9.1 Publications

At the time of writing, I have not submitted any publications for publishing. Following my PhD, I plan to submit five papers based on the outcomes of this thesis.

The first will focus on the concerns parents report as described in Chapter 2. Participants described the same and additional concerns regarding the symptoms of Cerebral Palsy (CP) reported in the literature. Publishing these concerns in the parents' own language may help primary health care professionals identify infants will emerging movement difficulties sooner.

The second paper will report on the delays to referral described in Chapter 3. To my knowledge, the reasons for delays to referral occurring within the community for CP have not been previously published. Similarly, to my knowledge, the similarities in the delays occurring across different paediatric conditions have also not been compared. This publication would aim to fill this gap in the literature.

The third publication will look to report the findings of citation network analysis. In particular, it will aim to raise questions about why there as so many motor screening tools and why are these tools are often developed using the same literature.

The fourth publication will describe the participatory design process of developing the screening tools. In particular, it will focus on the wants and needs of the parents.

The fifth, and arguably the most important publication, will focus on Dixon-Woods *et al.* (2005)'s candidacy. This paper will draw on the participants' experiences reported in Chapters 3 and 6 to demonstrate the ongoing issue of candidacy at the parent and health care professionals' levels.

9.2 Platform Presentations

Baggaley J., Rapley T., & Basu A. *Do the nature of concerns raised by parents influence whether an infant with emerging cerebral palsy is referred?* Presented at the European Academy for Childhood Disorders. November 2020

Baggaley J. *Bridging the communication gap: Utilising parents' earliest concerns of emerging Cerebral Palsy to aid early referral*. Presented at The Institute of Health and Society

Postgraduate Conference. Newcastle-Upon-Tyne, UK. 11th June 2019. Awarded second place in the Best Speaker competition.

9.3 Poster Presentations

Baggaley J., Rapley T. & Basu A. *Which motor screening tools for infants aged 0-6 months included parents in their development process? A scoping review.* Presented at the 34th Annual Meeting European Academy of Childhood Disability. Online. May 2022

Baggaley J., Rapley T. & Basu A. '*Navigating the ocean alone in a reed boat with no map.' Parental experiences of accessing primary care referral for their infants with Cerebral Palsy.* Presented at the 34th Annual Meeting European Academy of Childhood Disability. Online. May 2022

Baggaley J., Rapley T. & Basu A. *Exploration of Delays in referrals to secondary care of infants with suspected Cerebral Palsy.* Better Together 2022. February 2022.

Baggaley J., Rapley T. & Basu A. *'You are navigating the ocean alone in a reed boat with no map or oars.' Parental experiences of accessing primary care referral for their infants with Cerebral Palsy.* Presented at the North East Postgraduate conference. 13th November 2020. Awarded first place in the Best Poster Presentation award.

Baggaley J., Rapley T. & Basu A. *Red Flags: Parent-reported earliest concerns regarding their childs emerging Cerebral Palsy.* Presented at the Royal Collage of Paediatrics and Child Health. Online. November 2020.

Appendix

Table of Contents

A. Early Parental Observations in Infants with Cerebral Palsy Survey

Early Parental Observations in infants with Cerebral Palsy Survey V1.7

Red Flags Draft Survey: Early Parental Observations in Infants with Cerebral Palsy

Introduction

Hello and welcome to the Early Parental Observations Survey.

We are carrying out a survey to understand when parents and carers start to develop concerns about their child's development and what those concerns are.

We want to survey UK-based parents and carers who have a child with a diagnosis of Cerebral Palsy. You may be more familiar with terms such as Monoplegia, Hemiplegia, Diplegia, Triplegia, Tetraplegia, Quadriplegia, Pentaplegia and Dystonic Cerebral Palsy, all of which are forms of Cerebral Palsy. If your child is now an adult you may also take part.

In the survey you will be asked to give details about;

- The relationship you have to the child you are basing your answers on
- The child's current age and diagnosis
- The child's age at diagnosis and at referral
- The earliest observations you, or others, made about the child that caused you concern
- Who you first spoke to about your concerns
- How your first concerns were handled by health care professionals
- Your experiences of going to see health care professionals to discuss your concerns
- Yourself, such as your age and ethnicity

Your responses will go towards developing a new screening tool. The new screening tool aims to reduce the age children get access to appropriate care by taking in to account the early concerns parents and carers have. Unfortunately, participating in this survey will not directly benefit yourself or your child. However, your responses may help other families in the future.

If you are interested in learning more about this research survey and/or participating please click next and you will be taken to the information sheet about the survey. The information sheet will give you further information about the survey and what we will do with your responses. The information sheet will take around 5 minutes to read and the survey will take around 10 minutes to complete. If you are not interested, I would like to thank you for your time.

With thanks

Jess Baggaley

PhD student, Newcastle University
J.Baggaley2@newcastle.ac.uk

Participant Information Sheet

Early Parental Observations in Infants with Cerebral Palsy.

We invite you to take part in a research survey. The survey is being carried out by Jessica Baggaley and is being supervised by Dr Anna Basu at Newcastle University, Professor Tim Rapley at Northumbria University and by Dr Nadja Reissland at Durham University. The Economic Social Research Council's Northern Ireland and North East England Doctoral Training Partnership is funding this survey.

This survey aims to explore the earliest concerns parents and carers have about their infant's development. In particular, we would like to hear from parents/carers whose child now has a diagnosis of cerebral palsy. The overall aim of this research is to develop a new tool for identifying infants at risk of movement problems. We hope that by including parent/carer concerns, the final tool will help support new parents to raise their concerns about their child's development to health care professionals.

In this online survey you will be asked to give some brief details about yourself (such as your relationship to your child), your child (such as their age and diagnosis), the earliest concerning observations you made and your experiences of the referral process. In total there are 26 questions made up of multiple choice and free text questions, in which you may write your own responses. We expect the survey to take you around 10 minutes to complete.

Will my taking part be kept confidential?

We will keep the information you share with us confidential. Only those who you tell will know you took part in this survey.

As researchers, we will keep your information confidential. In this survey we do not ask you to provide any information that could directly identify yourself or others. Any information you do provide that could identify you will be removed before anyone else is able to view your responses. We will store all of your responses anonymously and securely in accordance with the Data Protection Act 1998, the General Data Protection Regulation (GDPR) legislation and the British Psychological Society Code of Ethics.

Once the survey has finished, your responses will be downloaded onto a secure Newcastle University server and stored within password protected computer files that only the research team can access. The responses stored online will then be deleted.

The survey host (JISC Online Surveys) does not use cookies or external tracking software and does not store your IP address. This means that no one will be able to identify you through your computer, tablet or phone. It is unlikely that anyone will ever find out you participated in this study unless you tell them.

At the end of the survey we will ask you to share the survey online with your friends and family who may also wish to take part. Sharing of the survey will not share your responses and sharing the survey is your choice. If you choose not to share the survey, we will not know and it will not affect you in any way.

What will you do with my responses?

Your responses will be used to;

1) Develop a new early screening tool for Cerebral Palsy.

2) Publish papers on the earliest concerns parents develop and the referral process they experience.

3) Only with your permission, support further research by sharing your anonymous, processed responses with other researchers.

Firstly, we will download your responses and store them on a secure Newcastle University server. The Newcastle University server will only be accessible to the research team. Next we will check that you have not given any information which may identify you or another. If you have given identifiable information, we will remove the identifiable information by replacing it for a description of place or person you mention. For example, if you were to say the specific name of the hospital your child attended, this would be replaced with the word "hospital". If you were to give a person's name, the person's name would be changed. Anonymising the data in this way will mean no one will be able to identify you from your responses. After anonymising your responses we will begin analysis. Your responses will be analysed in two ways. The first analysis is called "qualitative analysis", whereby we will look at and compare all of the text based responses. Comparing all of the responses will allow for us to determine the common concerns parents/carers report. Comparing all of the responses will also allow for us to determine the variation in concerns parents/carers report. The second analysis is "quantitative analysis" in which we will compare the multiple choice responses. By comparing all of the multiple choice responses we will be able to determine which factors influence the experiences and observations yourself and others had.

The outcomes of these two analyses will then be used to; 1) Develop a new screening tool based on the earliest parent/carer reported signs of Cerebral Palsy. 2) Publish papers demonstrating and explaining the earliest concerning observations parents, carers and family members make and their experiences with the referral process when their child is developing Cerebral Palsy. Only with your permission will we use your responses in these two ways. For both outcomes we may need to quote your responses, we will only do this if you give us permission. After the study has ended, we intend to share the processed, anonymous data with other researchers to help improve and support further research. We will only share your processed data if you give us permission.

What about my personal data?

Newcastle University is responsible for looking after your data and using it properly. We will not be asking you for any information that may identify you. Any demographic information you give will be used to assess if and how your demographics affected your referral and diagnosis experience.

Newcastle University will be using information from you in order to undertake this research study and will act as the data controller for this study. This means that Newcastle University is responsible for looking after your information and using it properly. Your rights to access, change or move your information are limited, as Newcastle University needs to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, Newcastle University will keep the information about you that has already been obtained. You can find out more about how Newcastle University uses your information at http://www.ncl.ac.uk/data.protection/PrivacyNotice and/or by contacting Newcastle University's Data Protection Officer (Maureen Wilkinson, rec-man@ncl.ac.uk).

We will use your demographic information in order to determine if and how that information affects the observations you made and the referral process you experienced. Individuals at Newcastle University may look at your research data to check the accuracy of the research study.

If you agree to take part in the research study, anonymised information provided by you will be stored for 10 years after the study has ended, as according to Newcastle University's Data policy. Your anonymised information may also be shared with researchers running other research studies at Newcastle University and in other organisations via the UK Data Service, in line with the Economic Social Research Council's data policy. These organisations may be universities and NHS organisations. Your information will only be used by organisations and researchers to conduct research.

This information will not identify you and will not be combined with other information in a way that could identify you. The information will only be used for the purpose research, and cannot be used to contact you. It will not be used to make decisions about future services available to you.

Are there any risks involved in participation?

We believe there are no known risks associated with this survey.

As with all research, participation may cause distress. As with all online activity, a data breach is always a risk. We will do our best to keep your data and participation in this survey confidential and we will only publish anonymous data. We have minimised any risks by not requesting any identifiable information and all data will be stored within password protected files only accessible to the research team.

This study was approved by the Faculty of Medical Sciences Research Ethics Committee, part of Newcastle University's Research Ethics Committee. This committee contains members who are internal to the Faculty, as well as one external member. This study was reviewed by members of the committee, who must provide impartial advice and avoid significant conflicts of interests.

Can I withdraw from the survey?

Yes.

Participation is completely voluntary. You may withdraw your data from the survey at any point by closing the online window. By closing the online window you will destroy any answers you have previously given. To move on from a page you will have to answer every question.

At the end of the survey you will be shown a finish button. Due to your responses being anonymous, clicking the finish button will submit your answers and you will no longer be able to change or withdraw your responses from the survey.

Contact us

If you have any complaints, questions, or if anything is unclear, please do not hesitate to get in touch. You can reach our team by emailing Jessica Baggaley at J.Baggaley2@newcastle.ac.uk or through phoning: 01912 821 378.

Thank you for reading.

Early Parental Observations in infants with Cerebral Palsy Survey V1.7

Consent Form

Please read through the following terms and conditions.

Please note, we define Cerebral Palsy as an umbrella term for a group of lifelong conditions that affect movement and co-ordination caused by a problem with the brain occurring before, during or soon after birth. Monoplegia, Hemiplegia, Diplegia, Triplegia, Tetraplegia, Quadriplegia, Pentaplegia and Dystonic Cerebral Palsy are all conditions which are classified under the term Cerebral Palsy.

1. I confirm that I have read and understood the information sheet
2. I confirm I have had the opportunity to consider the information sheet, ask questions and have had these answered satisfactorily
3. I understand that my participation is voluntary and that I am free to withdraw at any time by closing the window without having to give a reason
4. I understand that by clicking "Finish" at the end of the survey I can no longer change or withdraw my responses
5. I agree for this consent form and the data collected to be kept at Newcastle University
6. I give permission for anonymised quotes from the survey to be used in publications and presentations related to the study
7. I understand that the information I supply may be used to support other research in the future and may be shared anonymously to other researchers.
8. I confirm that I am looking after, or have looked after, an infant who now has a diagnosis of Cerebral Palsy.
9. I have a good understanding of the English Language and will be able to complete the survey
10. I live within the United Kingdom and Ireland
11. I am willing and consent to take part in this study

I have read and agree to the terms and conditions above

Yes No

Early Parental Observations in infants with Cerebral Palsy Survey V1.7

Demographics

Please answer these questions in relation to your child (or the child you care/cared for) who developed Cerebral Palsy.

Please note, we define Cerebral Palsy as an umbrella term for a group of lifelong conditions that affect movement and co-ordination caused by a problem with the brain occurring before, during or soon after birth. Monoplegia, Hemiplegia, Diplegia, Triplegia, Tetraplegia, Quadriplegia, Pentaplegia and Dystonia are all conditions which are classified under the term Cerebral Palsy.

1. What relationship do you have to the child in question?

   Mother

   Father

   Grandmother

   Grandfather

   Brother

   Sister

   Carer

   Other: Please describe

   _____

2. Which parts of the body does your child's Cerebral Palsy affect? *Tick all that apply.*

   Head and Neck

   Left Arm

   Right Arm

   Left Leg

   Right Leg

   Trunk/Body

3. Please state if your child has any other diagnosed conditions (such as epilepsy, autism, blindness, deafness, etc). If your child does not have any other diagnosed conditions please put N/A.

_____
_____
_____

4. Which age group is your child currently in?
    a. Under 2 Years
        i. Please skip forward to question 10, page 13
    b. 2-3 years
        i. Please skip forward to question 5, page 8.
    c. 4-5 years
        i. Please skip forward to question 6, page 9.
    d. 6-11 years
        i. Please skip forward to question 7, page 10
    e. 12-17 years
        i. Please skip forward to question 8, page 11
    f. Over 18 years
        i. Please skip forward to question 9, page 12

2-3 years

5. Please read the following and select the description that best represents your child's movement abilities.

1	**Has difficulty controlling head and trunk posture in most positions** ***and*** uses specially adapted seating to sit comfortably ***and*** has to be lifted by another person to move about
2	**Can sit on own when placed on the floor and can move within a room** ***and*** uses hands for support to maintain sitting balance ***and*** usually uses adaptive equipment for sitting and standing ***and*** moves by rolling, creeping on stomach or crawling
3	**Can sit on own and walk short distances with a walking aid** (such as a walker, rollator, crutches, canes, etc.) ***and*** may need help from an adult for steering and turning when walking with an aid ***and*** usually sits on floor in a "W-sitting" position ***and*** may need help from an adult to get into sitting and may pull to stand ***and*** cruise short distances and prefers to move by creeping and crawling
4	**Can sit on own and usually moves by walking with a walking aid** ***and*** may have difficulty with sitting balance when using both hands to play ***and*** can get in and out of sitting positions on own ***and*** can pull to stand and cruise holding onto furniture ***and*** can crawl, but prefers to move by walking
5	**Can sit on own and moves by walking without a walking aid** ***and*** is able to balance in sitting when using both hands to play ***and*** can move in and out of sitting and standing positions without help from an adult ***and*** prefers to move by walking

Now please skip to question 10, page 13.

1. years

6. Please read the following and select the description that best represents your child's movement abilities.

1	**Has difficulty sitting on their own and controlling head and body posture in most positions** ***and*** has difficulty achieving any voluntary control of movement ***and*** needs a specially-adapted supportive chair to sit comfortably ***and*** has to be lifted or hoisted by another person to move
2	**Can sit on their own but does not stand or walk without significant support and adult supervision** ***and*** may need extra body / trunk support to improve arm and hand function ***and*** usually needs adult assistance to get in and out of a chair ***and*** may achieve self-mobility using a powered wheelchair or is transported in the community
3	**Can walk on their own using a walking aid** (such as a walker, rollator, crutches, canes, etc.) ***and*** can usually get in and out of a chair without adult assistance ***and*** may use a wheelchair when travelling long distances or outside ***and*** finds it difficult to climb stairs or walk on an uneven surface without considerable help
4	**Can walk on their own without using a walking aid, but has difficulty walking long distances or on uneven surfaces** **and** can sit in a normal adult chair and use both hands freely **and** can move from the floor to standing without adult assistance **and** needs to hold the handrail when going up or down stairs **and** is not yet able to run and jump
5	**Can walk on their own without using a walking aid, including fairly long distances, outdoors and on uneven surfaces** **and** can move from the floor or a chair to standing without using their hands for support **and** can go up and down stairs without needing to hold the handrail **and** is beginning to run and jump

Now please skip to question 10, page 13.

6-11 years

7. Please read the following and select the description that best represents your child's movement abilities.

1	**Has difficulty sitting on their own and controlling head and body posture in most positions** **and** has difficulty achieving any voluntary control of movement **and** needs a specially supportive chair to sit comfortably **and** has to be lifted or hoisted by another person to move
2	**Can sit on their own but does not stand or walk without significant support and adult supervision** **and** therefore relies mostly on wheelchair at home, school and in the community **and** often needs extra body / trunk support to improve arm and hand function **and** may achieve self-mobility using a powered wheelchair or is transported in the community
3	**Can stand on their own and only walks using a walking aid** (such as a walker, rollator, crutches, canes, etc.) ***and*** finds it difficult to climb stairs, or walk on uneven surfaces ***and*** may use a wheelchair when travelling long distances or outside
4	**Can walk on their own without using a walking aid, but needs to hold the handrail when going up or down stairs** ***and*** often find it difficult to walk on uneven surfaces, slopes or in crowds
5	**Can walk on their own without using a walking aids, and can go up or down stairs without needing to hold the handrail** ***and*** walks wherever they want to go (including uneven surfaces, slopes or in crowds) ***and*** can run and jump although their speed, balance, and coordination may be slightly limited

8.

Now please skip to question 10, page 13.

12-17 years

8. Please read the following and select the description that best represents your child's movement abilities.

1	**Has difficulty sitting on their own and controlling head and body posture in most positions** **and** has difficulty achieving any voluntary control of movement **and** needs a specially adapted chair to sit comfortably and be transported anywhere **and** has to be lifted or hoisted by another person or specialist equipment to move
2	**Can sit with some pelvic and trunk support but does not stand or walk without significant support** **and** therefore always relies on wheelchair when outdoors **and** can achieve self-mobility using a powered wheelchair **and** can crawl or roll to a limited extent to move around indoors
3	**Can stand on their own and only walks using a walking aid** (such as a walker, rollator, crutches, canes, etc.) ***and*** finds it difficult to climb stairs, or walk on uneven surfaces without support ***and*** uses a variety of means to move around depending on the circumstances ***and*** prefers to use a wheelchair to travel quickly or over longer distances
4	**Can walk on their own without using walking aids, but needs to hold the handrail when going up or down stairs** ***and*** therefore walks in most settings ***and*** often find it difficult to walk on uneven surfaces, slopes or in crowds ***and*** may occasionally prefer to use a walking aid (such as a cane or crutch) or a wheelchair to travel quickly or over longer distances
5	**Can walk on their own without using walking aids, and can go up or down stairs without needing to hold the handrail** ***and*** walks wherever they want to go (including uneven surfaces, slopes or in crowds) ***and*** can run and jump although their speed, balance, and coordination may be slightly limited

9.

Now please skip to question 10, page 13.

18+ years

9. Please read the following and select the description that best represents your child's movement abilities.

1	**Has difficulty sitting on their own and controlling head and body posture in most positions** **and** has difficulty achieving any voluntary control of movement **and** needs a specially adapted chair to sit comfortably and be transported anywhere **and** has to be lifted or hoisted by another person or specialist equipment to move
2	**Can sit with some pelvic and trunk support but does not stand or walk without significant support** **and** therefore always relies on wheelchair when outdoors **and** can achieve self-mobility using a powered wheelchair **and** can crawl or roll to a limited extent to move around indoors
3	**Can stand on their own and only walks using a walking aid** (such as a walker, rollator, crutches, canes, etc.) ***and*** finds it difficult to climb stairs, or walk on uneven surfaces without support ***and*** uses a variety of means to move around depending on the circumstances ***and*** prefers to use a wheelchair to travel quickly or over longer distances
4	**Can walk on their own without using walking aids, but needs to hold the handrail when going up or down stairs** ***and*** therefore walks in most settings ***and*** often find it difficult to walk on uneven surfaces, slopes or in crowds ***and*** may occasionally prefer to use a walking aid (such as a cane or crutch) or a wheelchair to travel quickly or over longer distances
5	**Can walk on their own without using walking aids, and can go up or down stairs without needing to hold the handrail** ***and*** walks wherever they want to go (including uneven surfaces, slopes or in crowds) ***and*** can run and jump although their speed, balance, and coordination may be slightly limited

10.

Diagnosis and Referral

10. How old was your child when your child received a diagnosis of Cerebral Palsy?

    a. Under 1 month
    b. 1 Month
    c. 2 Months
    d. 3 Months
    e. 4 Months
    f. 5 Months
    g. 6 Months
    h. 7 Months
    i. 8 Months
    j. 9 Months
    k. 10 Months
    l. 11 Months
    m. 12 months to 17 months
    n. 18 months to 23 months
    o. 2 Years
    p. 3 Years
    q. 4 Years
    r. 5 Years
    s. 6+ Years

11. How old was your child when your child was first referred for therapy? Either for concerns about their development or for their Cerebral Palsy.

    a. Under 1 month
    b. 1 Month
    c. 2 Months
    d. 3 Months
    e. 4 Months
    f. 5 Months
    g. 6 Months
    h. 7 Months
    i. 8 Months
    j. 9 Months
    k. 10 Months
    l. 11 Months
    m. 12 months to 17 months
    n. 18 months to 23 months
    o. 2 Years
    p. 3 Years
    q. 4 Years
    r. 5 Years
    s. 6+ Years

12. Please describe the earliest observations you made about your child's development that caused you concern?

    You may wish to describe when you first made each observation, what you and the child were doing, where you were and what about the observation concerned you or did not concern you at the time. You may also include when others told you about observations they made that concerned them. If possible please also give the age your child was at the time you made each of the observations. You may wish to bullet point each observation you describe to help you keep track.

    More Info: *This can include any concerns you had about your child's arm and leg movements, feeding, hearing, playing, vision, sounds they did or did not make, milestones.*

_____

_____

The Referral Process

13. Who was the first person to become concerned about your child's development?

Myself

The Child's Mother

The Child's Father

The Child's Grandparent

The Child's Sibling

Other family member (please describe)

A friend

GP

Nurse Practitioner

Nurse

Health Visitor

Midwife

Paediatrician

Obstetrician

Neonatologist

Other Health Care Professional (please describe)

_____

Other (please describe)

_____

14. What was the nature of the concern?

_____

_____

_____

_____

_____

15. Which health care professional did you, or another, first raise your concern too?

GP

Nurse Practitioner

Nurse

Health Visitor

Midwife

Paediatrician

Obstetrician

Other; please describe

_____

a. How old was your child when the first concern was raised to, or made by, a health care professional?

i. Under 1 month
ii. 1 Month
iii. 2 Months
iv. 3 Months
v. 4 Months
vi. 5 Months
vii. 6 Months
viii. 7 Months
ix. 8 Months
x. 9 Months
xi. 10 Months
xii. 11 Months
xiii. 12 months to 17 months
xiv. 18 months to 23 months
xv. 2 Years
xvi. 3 Years
xvii. 4 Years
xviii. 5 Years
xix. 6+ Years

b. Did the health care professional refer the child for further assessment the first time concerns were raised?

i. Yes
ii. No

c. If no, what happened next?
Please tell us about how many appointments were made with primary health care professions (such as GPs and Health Visitors) and which health care professionals you spoke to before the child was referred. Please give the reasons you were given as to why the child was not referred and how long it took for the child to be referred. If you are not sure about the amount of appointments, who you saw, the reasons you were given and the length of time you waited for the referral please give your best estimations.

16. Overall how do you think the health care services handled your concerns and observations?
    i. Well

ii. Okay

iii. Poorly

17. In terms of the service you received from the NHS,

i. What was done well? If you have no comments please answer no comment. If you do not wish to answer please type N/A

1. More information: you may wish to comment on the speed of referral/diagnosis, being given appropriate literature, sign posting to peer support groups, receiving counselling

_____
_____
_____
_____
_____
_____
_____
_____
_____
_____
_____

ii. What could have been done better? If you have no comments please type no comment. If you do not wish to answer please type N/A

1. More information: you may wish to comment on the speed of referral/diagnosis, being given appropriate literature, sign posting to peer support groups, receiving counselling

_____
_____
_____
_____
_____
_____
_____
_____

iii. What are your suggestions to improve the early referral/diagnostic service you experienced? If you have no comments please answer no comment. If you do not wish to answer please type N/A

1. More information: you may wish to comment on the speed of referral/diagnosis, being given appropriate literature, sign posting to peer support groups, receiving counselling

18. Please provide any other information that you think may be relevant.

_____
_____
_____
_____

20

Your demographics

Finally, we need to ask some personal questions. We only ask these questions to understand who is taking part in our study.

19. What is your age?

    _____

20. What is your Ethnicity?

    a. White/European
    b. Pakistani
    c. Chinese
    d. Indian
    e. Bangladeshi
    f. Asian other
    g. Black African
    h. Black Caribbean
    i. Black Other
    j. Mixed (Please specify)

       _____

    k. Other (Please specify)

       _____

21. What Country do you live in?

    a. UK
    b. Other, please state

       _____

22. What is the highest level of education you have achieved?

    a. No formal qualifications
    b. GCSE level or equivalent
    c. A levels or equivalent
    d. University degree (eg. BSc, MA, PhD or equivalent)
    e. Other, *please describe*

    _____

23. What is your current employment status?

    a. Employed full time
    b. Employed part time
    c. Full-time carer
    d. Full-time homemaker
    e. Unemployed and looking for work
    f. Unemployed due to health
    g. Retired
    h. Doing voluntary work
    i. Full-time student
    j. Maternity/paternity leave
        i. Was your Maternity/Paternity leave from a full time or part time job
            1. Full time
            2. part time
    k. Other (please specify)

    _____

24. Has this changed since you developed concerns about the child's development?

    a. Yes
    b. No

If yes: What was your employment status when you became concerned about your child's development?

a. Employed full time
b. Employed part time
c. Full-time carer
d. Full-time homemaker
e. Unemployed and looking for work
f. Unemployed due to health
g. Retired
h. Doing voluntary work
i. Full-time student
j. Maternity/paternity leave
    i. Was your Maternity/Paternity leave from a full time or part time job
        1. Full time
        2. part time
k. Other (please specify)
    _____

2. What is your marital status?
    a. Married/civil partnership/co-habiting with long term partner
    b. Divorced/separated
    c. Single
    d. Widowed

Debrief

Thank you for volunteering your time to take part in this survey. Your responses will be used to help further our understand Cerebral Palsy, potentially helping other families. This page is to provide you with more information about our research and why we are doing this study. Please feel free to ask any questions or to comment on any aspect of the study.

You have just participated in the Red Flags Survey conducted by Miss Jessica Baggaley, Dr Anna Basu, Professor Tim Rapley and Dr Nadja Reissland.

The point of this study is to determine what the earliest concerning observations parents and carers make when their child is developing Cerebral Palsy. Your responses will now be analysed and used in the development of a screening tool. We hope that the screening tool will help identify infants at risk of developing Cerebral Palsy earlier and will result in earlier referral to services providing help. The next stage of the research is to carry out focus groups to determine which observations you made will be the best to identify infants at risk of developing Cerebral Palsy.

We will do our best to keep your responses confidential and we will ensure that your responses are anonymised and stored securely.

As you know, your participation in this study is voluntary. As you have now submitted your responses we are no longer able to withdraw your data from the study.

We do hope you will share this study with friends and family who may also wish to participate in the study. You can do this by sharing the study URL https://newcastle.onlinesurveys.ac.uk/parent-survey via email, text, social media and by talking about the study.

You may keep a copy of this debriefing for your records by printing the page through your web browser.

If you have any questions about the research or please contact Miss Jessica Baggaley at J.Baggaley2@newcastle.ac.uk or 01912 821 378. If, as a result of your participation in this study, you have experienced any adverse reactions, please get in touch with Miss Jessica Baggaley at J.Baggaley2@newcastle.ac.uk or 01912 821 378 or contact your GP.

Thank you again for taking part in this survey. If you would like to continue following our research, you can follow our research team on Twitter at @JessBaggaley1, @AnnaBasu1 and @JanicepeaOT.

This study was approved by the Faculty of Medical Sciences Research Ethics Committee, part of Newcastle University's Research Ethics Committee. This committee contains members who are internal to the Faculty, as well as one external member. This study was reviewed by members of the committee, who must provide impartial advice and avoid significant conflicts of interests.

B. Early Parental Observations in Infants with Cerebral Palsy recruitment poster

Durham University

NINE Doctoral Training in the Social Sciences North East of England and Northern Ireland

E·S·R·C Economic & Social Research Council

Newcastle University

Did you notice a problem with your child's early movements?

Are you the parent of a child diagnosed with
Cerebral Palsy, Monoplegia, Hemiplegia, Diplegia, Triplegia, Tetraplegia, Quadriplegia OR Pentaplegia?

We need you!

If so, we are interested in hearing about the first concerns **you** had about **your child's** movement and **your experiences** of the referral and diagnosis process.

About the project

We only ask that you complete our online survey. The survey contains 25 questions and should take you around 10 minutes to complete.

Why take part?

We are developing a new tool to identify infants at risk of developing movement problems and to help support new parents when raising concerns about their child's movement. Your answers will inform what we include in the new tool we are developing.

How do I get involved?

To find out more or to take part go to
https://newcastle.onlinesurveys.ac.uk/parent-survey
Or contact Jessica Baggaley at J.Baggaley2@newcastle.ac.uk

C. Break down of the signs of Cerebral Palsy identified in Garfinkle *et al.*, (2020) compared to the caregiver concerns identified in Chapter 2.

Concerns identified by Garfinkle et al., (2020)	*Signs of CP as described by Garfinkle et al. (2020)*	*Parental concerns split by the type of concern*		
		Day to day concerns	*Developmental milestones*	*Troubling medical history*
Clinical features	Early handedness	Hand posture		
	Irritability, including; jitteriness, jumpy behaviour, excessive crying, and easy startling	Temperament, and movement. Reflexes and reactions (startle reflex)		
	Reduced level of consciousness, including Lethargy, lack of alertness, and irregular sleep patterns	Sleep, and movement.		
	Feeding problems: Poor sucking, poor swallowing, excessive drooling, and oral hypersensitivity	Feeding problems		
	Stiffness when handled	Tone		
	Difficulty diapering	Tone		

Concerns identified by Garfinkle et al., (2020)	*Signs of CP as described by Garfinkle et al. (2020)*	*Parental concerns split by the type of concern*		
		Day to day concerns	*Developmental milestones*	*Troubling medical history*
	Seizures			Epilepsy/ Seizures/ Convulsions
	Strabismus	Eye gaze		
	Recurrent infections			Infections
Developmental milestones	Volitional rolling delays beyond 4-6 months			
	Paradoxical early rolling at 1-2 months		Delayed rolling	
	Siting delayed beyond 7-9 months		Delayed sitting	
	Walking delayed beyond 15-18 months		Delayed walking	
Neurological examination	Persistence of primitive reflexes, beyond 4-6 months, such as the Moro, crossed extensor, and the suprapubic extensor reflex. Beyond 8 months the Galant response and the Asymmetrical tonic neck reflex.	Reflexes and reactions – asymmetric tonic neck reflex		

Concerns identified by Garfinkle et al., (2020)	*Signs of CP as described by Garfinkle et al. (2020)*	*Parental concerns split by the type of concern*		
		Day to day concerns	*Developmental milestones*	*Troubling medical history*
	Absence of primitive reflexes when they should be present, such as the Moro before 4 months and the plantar grasp reflexes before 6 months of age			
	Delay or failure to acquire postural reflexes, such as propping and parachute reflexes			
	Asymmetry in popliteal angle, hand position, Scalf sign, kicking in vertical suspension, plantar grasp response, tone and movements	Asymmetrical movements Tone Posture		
	Hypotonia in the early phase followed by hypertonia, particularly after 6 months.	Tone		
	A head lag beyond 5-6 months		Delayed head control	
	Fisting	Hand posture		
	Scissoring	Leg posture		
	Clonus			
	Babinski sign			

Concerns identified by Garfinkle et al., (2020)	*Signs of CP as described by Garfinkle et al. (2020)*	*Parental concerns split by the type of concern*		
		Day to day concerns	*Developmental milestones*	*Troubling medical history*
	Toe walking		Atypical walking	
	Abnormal crawling, including scooting, commando crawling, non alternating crawling, asymmetrical crawling.		Atypical crawling	
	Wide based and pigeon toed posturing.		Atypical standing	

Red is used to highlight the signs of Cerebral Palsy identified by Garfinkle *et al*., (2020) that were not identified by the caregivers.

D. Scoping review search stratagies

D.1 Embase

1.	"sensitivity and specificity"/
2.	bayley scales of infant development.mp. or "Bayley Scales of Infant Development"/
3.	child development/
4.	denver developmental screening test.mp. or Denver Developmental Screening Test/
5.	psychomotor performance/
6.	motor performance/
7.	harris infant neuromotor test.mp.
8.	Peabody developmental motor scale.mp.
9.	alberta infant motor scale.mp.
10.	(ages and stages questionnaire).mp. [mp=title, abstract, heading word, drug trade name, original title, device manufacturer, drug manufacturer, device trade name, keyword, floating subheading word, candidate term word]
11.	mullen scales of early learning.mp.
12.	hand assessment of infants.mp.
13.	infant/
14.	prematurity/
15.	postmaturity/
16.	newborn/
17.	general movements.mp.

18.	"movement (physiology)"/
19.	reproducibility/
20.	psychometry/
21.	2 or 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 12 or 17
22.	13 or 14 or 15 or 16
23.	concurrent validity of the alberta infant motor scale to detect delayed gross motor development in preterm infants.m_titl.
24.	Development of the Hand Assessment for Infants.m_titl.
25.	(The Structured Observation of Motor Performance in Infants has convergent and discriminant validity in preterm and term infants).m_titl.
26.	23 or 24 or 25
27.	exp validity/
28.	1 or 19 or 20 or 27
29.	21 and 22 and 28
30.	26 and 29

D.2 Medline

1	"Sensitivity and Specificity"/ or Mass Screening/
2.	Child Development/ or bayley scales of infant development.mp.
3.	bayley scales of infant development.mp.
4.	denver developmental screening test.mp.
5.	psychomotor performance/ or motor skills/
6.	harris infant neuromotor test.mp.
7.	Peabody developmental motor scale.mp.
8.	alberta infant motor scale.mp.
9.	(ages and stages questionnaire).mp. [mp=title, abstract, original title, name of substance word, subject heading word, floating sub-heading word, keyword heading word, protocol supplementary concept word, rare disease supplementary concept word, unique identifier, synonyms]
10.	mullen scales of early learning.mp.
11.	hand assessment of infants.mp.
12.	INFANT, VERY LOW BIRTH WEIGHT/ or INFANT, EXTREMELY PREMATURE/ or INFANT, POSTMATURE/ or INFANT, SMALL FOR GESTATIONAL AGE/ or INFANT, EXTREMELY LOW BIRTH WEIGHT/ or INFANT, PREMATURE/ or INFANT/ or INFANT, NEWBORN/
13.	general movements.mp.
14.	MOVEMENT/ or movement.mp.
15.	3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 or 11 or 13 or 14

16.	validation studies/
17.	"Reproducibility of Results"/ or Psychometrics/
18.	1 or 16 or 17
19.	12 and 15 and 18
20.	limit 19 to (english language and humans and ("all infant (birth to 23 months)" or "newborn infant (birth to 1 month)" or "infant (1 to 23 months)"))

D.3 Psycinfo

1	concurrent validity of the alberta infant motor scale to detect delayed gross motor development in preterm infants.m_titl.
2.	Development of the Hand Assessment for Infants.m_titl.
3.	(The Structured Observation of Motor Performance in Infants has convergent and discriminant validity in preterm and term infants).m_titl.
4.	1 or 2 or 3
5.	*test validity/ or test sensitivity/ or test specificity/
6.	test reliability/ or interrater reliability/ or test standardization/
7.	5 or 6
8.	exp "Bayley Scales of Infant Development"/ or bayley scales of infant development.mp.
9.	denver developmental screening test.mp.
10.	harris infant neuromotor test.mp.
11.	Peabody developmental motor scale.mp.
12.	alberta infant motor scale.mp.
13.	(ages and stages questionnaire).mp. [mp=title, abstract, heading word, table of contents, key concepts, original title, tests & measures]
14.	mullen scales of early learning.mp.
15.	hand assessment of infants.mp.
16.	general movements.mp.
17.	*infant development/

18.	exp MOTOR SKILLS/ or exp MOTOR CONTROL/ or exp MOTOR DEVELOPMENT/
19.	8 or 9 or 10 or 11 or 12 or 13 or 14 or 15 or 16 or 17 or 18
20.	infan*.mp.
21.	newborn.mp.
22.	neonat*.mp.
23.	preterm.mp.
24.	20 or 21 or 22 or 23
25.	7 and 19 and 24
26.	4 and 25

D.4 Pubmed

(("infant") AND ((((((((((("bayley scales of infant development") OR "denver developmental screening test") OR "harris infant neuromotor test") OR "peabody developmental motor scale") OR "alberta infant motor scale") OR ("ages and stages questionnaire")) OR "mullen scales of early learning") OR "hand assessment of infants") OR "general movements") OR "movement") OR "motor development")) AND ((((("sensitivity") OR "specificity") OR "validation") OR "reproducibility of results") OR "psychometrics") OR "reliability")

Limits: Infants (0-23months)

D.5 Web of Knowledge

TS=(("infant") AND ("bayley scales of infant development" OR "denver developmental screening test" OR "harris infant neuromotor test" OR "peabody developmental motor scale" OR "alberta infant motor scale" OR "ages and stages questionnaire" OR "mullen scales of early learning" OR "hand assessment of infants" OR "general movements" OR "movement" OR "motor development") AND ("sensitivity" OR "specificity" OR "validation" OR "reproducibility of results" OR "psychometrics" OR "reliability")

E. Methods used to include expert opinions in the development of items for screening tools

	Tool	*Expert involvement*
1	ASQ3	Items were reworded based on feedback from project staff, interventionists, parents, nurses, and paediatricians using the questionnaires in clinic and research environments.
2	AIMS	Reviewed by paediatric physiotherapists for appropriate content and clinical importance. A mail inquiry also carried out on 291 members of the Paediatric division of the Canadian Physiotherapy Association. 6 international experts attended a 2 day work session as part of the content validation process.
4	Bayley III	Semi-structured survey used in all phases of test development. Experts and examiners asked to rate the scale qualities (developmental appropriateness, user-friendliness and clinical utility). The results fed back to the advisory panel and clinical measurement consultants. Series of focus groups with N=71 assessment professionals working in child development. Advisory panel of nationally recognised experts in cognitive, language, motor and social emotional development were assembled to work in the dev team throughout the dev of BSID III. Clinical measurement consultants also gave feedback.
6	cDMAT	Focus groups were used to discuss the cDMAT results after the cDMAT's performance reference charts and reliability had been assessed.

	Tool	*Expert involvement*
7	CREDI	After pilot testing, items that either 1) had more than 10% 'don't know' response rate; 2) were not understood less than 80% of participants in qualitative interviews; 3) have a test-retest reliability below .40; and 4) scores vary across, low, middle, and high income countries were removed. Item response theory was then used on the remining items to so that only the 20 items that had the least standard error of the measurement were included for each 6-month age band.
11	GRAB	Expert review panel, consisting of four senior occupational therapists, whose experience in paediatrics ranged from 15 to over 30 years; (ii) a senior physiotherapist with over 30 years' experience in paediatrics; and (iii) a child neurologist with 13 years' experience in paediatrics. The panel confirmed that the test items and structured play session of the GRAB should: (i) detect asymmetries in unimanual/bimanual reach and grasp behaviours between ULs in infants with asymmetrical brain injury; and (ii) detect differences in unimanual/bimanual reach and grasp behaviours between term-born TD infants and infants with asymmetrical brain injury. The expert panel reviewed the initial scoring criteria of 15 assessment occasions by (a combination of randomly selected term-born TD infants and infants with asymmetrical brain injury at 14, 16 and 18 weeks C.A.). As scoring difficulties were identified in differentiating between unimanual contacts as 'palmar' or 'dorsal' based on hand orientation; the scoring was modified

	Tool	*Expert involvement*
12	HINE	Used in clinical practice for 'a number of years' and underwent 'several' modifications (Mercuri, Haataja and Dubowitz, 2007).
13	HNNE	User feedback; items removed due to difficulty in eliciting them, only giving limited information and upsetting the parent.
15	HINT	Reviewed by 26 international experts, including 13 physical therapists, 4 occupational therapists, a paediatrician, a psychologist, and an early childhood special educator.
17	IYCD	A working group was set up to form a consensus on the items that should be included.
20	MDAT	An expert panel reviewed the pilot study results and decided which items should remain, which should be modified, and which should be removed.
21	Milani-Comparetti Developmental	Used by colleagues before publishing
23	Movement quality measure	Convenience sample of 8 paediatric physical therapists took part in in depth structured interviews. Seven paediatric physical therapist participated in the NGT phase. The NGT lasted up to 2 hours. 61 paediatric physical therapists, identified through expert sampling, took part in the Delphi survey.

	Tool	*Expert involvement*
27	PediTrac	An interdisciplinary team of nine faculty and clinical professionals from two universities and the private sector provided consensus opinion regarding the core conceptual constructs (domain map) to be included in the PediaTrac survey using the Delphi method. To further validate the content of PediaTrac v.1.0, a novel panel of three subject matter experts with 12–40 years of clinical experience were selected based on expertise from a sample of possible rater's nominated by the multidisciplinary team. The experts evaluated the items to ensure a representative sample of the items and domains of interest, appropriateness of the timing of the items, and clarity of the items.
28	PEDI-CAT	Feedback on the original PEDI. Focus groups with Physical therapy, occupational therapy, speech and language clinicians, and parents of children with disabilities to provide feedback on the expanded set of items and response scales. The expanded set of items and response scales were also sent to a group of physical and occupational therapy clinicians for feedback. Following the additions and revisions to the pool of items, structured, individual, cognitive interviews were conducted to finalize the items for calibration. finalize the items for calibration.

	Tool	*Expert involvement*
32	Rapid Neurodevelopmental Assessment	Items selected based on being age-references in worldwide assessment procedures and from the research teams own clinical experiances.
36	TIMP	Consultation with the rating clinicians resulted in revising definitions of misfitting items to eliminate ambiguities in descriptors that were identified as potentially responsible for misfit or to better reflect the dev sequence in the rach analysis.
38	Denver II	A consultant speech pathologist reviewed the DDST items, leaving 82 items unchanged, 21 revised, and adding in 43 new items.

F. Items identified from screening tools by their item groups

F.1 Eyes

Conjugate gaze (regards), Convergence of eyes, Corneal reflex, eye movements, Eye muscle control, Hypertonia of the levator muscles of the upper eyelids, Inspects own hand, Looks for…, Nystagmus, Optical blink reflex, Pupils, Roving eye movements, Setting sun sign, Strabismus, Visual following, Visual scanning.

F.2 Locomotion

Active trunk and leg movements while crawling, Actively lifts pelvis, buttocks and unweighted leg, Arm movements, Arm posture, axial rolling, Balance, Climbs, Crawl progression, Crawls up stairs, Cruses, Foot placement, Four point kneeling, Gait, Gets into four point kneeling, Leg movement, Leg posture, Reciprocal limb movements, Steps over things, Trunk movement, Trunk posture, walks independently, Walks on tiptoes, Walks up stairs, Walks with support, Weight shift.

F.3 Movement

Abnormal movements of the arms, Appropriate movements, Asymmetry, Athetoid posture and movements, Automated movements, Clonic movements, Co-ordinated movements, Dyskinetic movements of the limbs, Fluency, Involuntary movement, Limb movements, Movement quality, Movement quantity, Quality, Quantity, Segmental arm movements, Segmental leg movements, Speed, Spontaneous motor activity, Stereotyped movements, Strength regulation, Variation, Variation in leg movement, Volitional movement, Ankle movements.

F.4 Prone

Arm posture, Arm posture, Chin tuck, Extended arms, Extending the elbows with pronation at the wrists, Forearm support, Head lift, Head righting, Head turn, Hip posture, Leg posture, Movement quality, Pelvis posture, Pivoting, Places head gently down, Posture, Propped side lying, Reaching, Rolling, Trunk posture, Weight shift.

F.5 Reaching

Arm movements, Brings hands together, Brings to midline, Dumps objects, Grasp type, Grasping, Hand activity not related to objects, Hand motility, Hand posture, Hands to midline, Hands to mouth, Holds toys, Lets go, Manipulates objects, Moves fingers, Palmer

grasp, Plays/reaches with/for toy, Prehensile movements, Pulls/pushes object, Reaching, Rolls object, Rotates wrist, Throws ball, Touches, Touches own face, Transfers toys, Unimanual activity, Uses arm, Voluntary.

F.6 Reflexes

Clonus, Crossed adduction, Extensor thrust, Slip through, Withdrawel, Abdominal reflex, Acoustic blink reflex, Adductor reflex, Adductors angle, Anal reflex, Antigravity posture, Ankle jerk, ATNR, Axillary hanging, Babinski, Bauer, Bicep reflex, Branchioradialis jerk, Chaddock sign, Chovstek's reflex, Cremaster reflex, Crossed extensor reflex, Defensive movements, Derotative righting, Dorsal suspension posture, Equilibrium in sitting, Equilibrium reaction, Flapping of the foot, Flapping of the hand, Galent reflex, Glabella tap, Hip abduction reaction, Hoffman sign, Horizontal suspension, Jaw Jerk, Knee jerk, Landau reflex, Lateral propping reaction, Lip reflex, Magnet reflex, Mass reflex, Moro reflex, Neck righting, Optical placing, Optical righting of head, Optokinetic nystagmus, Palmo-mental response, Parachute reaction, Passive shoulder elevation, Passive trunk rotation, Pectoralis jerk, Placing response, Plantar grasp, Positive support reaction, Prone suspension posture, Protective extension, Reaction to tactile stimuli, Resistance against passive movements, Response to rotation, Rooting, Scarf sign, Schaltenbrandt reaction, Sensory motor responses, Square window, Startle, Suck reflex, Symmetrical tonic neck reflex, Tendon reflexes, Threat reflex, Tilting reflexes, Tonic labyrinth reflex, Turns to auditory stimulation, Ventral suspension posture, Vertical suspension, Walking reaction.

F.7 Sitting

Active extension, Actively pulls to sit (arm flexion), Arm flexion to traction, Arm posture, Can be left alone/ sits independently, Chin tuck (Head lag), Free head movements, Leg posture, Leg tone, Lifts and maintains head, Motility, Moves in and out of sitting, Scapular adduction, Sits in chair, Sits when placed, Sits with support, Sits without support, Sitting position, Supports weight on arms, Trunk posture, Unsustained sitting, Weight shift.

F.8 Standing

Active trunk control, Balancing, Can bend over while standing, Controlled lowering, Foot placement, Four point kneeling to standing, gets in and out of Standing, Half kneeling, Head in midline, Hip and knee extension, Hip and knee flexion, Hip posture, Jumps, Kicks a ball, Kneeling, Leg posture, Pulls to stand, Rotation of trunk and pelvis, Shifts weight, Squats,

Stand up from sitting, Standing, Stands alone, Stands on toes, Supported standing, Supports own weight, Trunk movement, Trunk posture, Variation in standing behaviour.

F.9 Supine

Arm and leg movements, Arm posture, Body lying in supine, Chin tuck, Fluency, Foot to foot contact, Hand to knee contact, Head movements, Head rotation, Hip posture, Holds feet, Holds knees, Kicking, Leg movement variability, Leg posture, Lifts arms to 'come here', Mobility, Mouths feet, Mouths hand, Mouths object, Movement quality, Rolling, Supine arm protection, Supine suspension, Trunk posture.

F.10 Tone

Arm pronation and supination, Arm recoil, Arm traction, Dorsiflexion angle of the foot, Extension of the trunk, Extensor tone, Flexor tone, Heel to ear, Lateral flexion of the trunk, Leg abduction, Leg recoil, Leg traction, Limb posture asymmetry, Muscle tone - lower extremities, Muscle tone - upper extremities, Neck posture, Neck tone, Popliteal angle, Posture, Resting posture, Symmetrical posture, Toe posture, Tone distribution, Tone in general, Trunk tone, Ventral flexion of the trunk.

F.11 Other

Alertness, Behaviour quantification, Cognitive processes, Consolability, Contractures, Convulsions, Crying, Dressing, Dribbling, Drinks from an open cup, Excitement bursts, Facial appearance, Feeding behaviours, Finds object under sheet, Gets…, Getting dressed, Habituation, Home tasks, Imitation, Irritability, Keeping clean, Measurements, Motor tasks, Mouth movement, Moves quickly/ keeps up, Opens and closes doors, Opisthotonos, Placing of hands and feet, Pumps legs, Respiration, Skin, Sleep, Sucks finger/thumb, Supporting with hands and feet, Swimming, Tickling, Tongue movements, Tremor, Twitching, Vocalisation.G

G. Items tools included based on item groups developed in Chapter 5.

Tool	*Prone*	*Supine*	*Sitting*	*Standing*	*Locomotion*	*Hand use*	*Eyes*	*Movement quality and posture*	*Reflexes and reactions*	*Tone*	*Other*
ASQ3	Yes	Yes	Yes	Yes	-	Yes	-	-	Yes	-	-
AIMS	Yes	Yes	Yes	Yes	Yes	-	-	-	Yes	-	-
Amiel-Tison neurological examination	-	Yes	Yes	Yes	-	Yes	Yes	Yes	Yes	Yes	Yes
Bayley III	Yes	Yes	Yes	Yes	Yes	Yes	Yes	-	Yes	Yes	Yes
BINS	-	-	Yes	-	Yes	Yes	Yes	Yes	-	Yes	Yes
cDMAT	Yes	Yes	Yes	Yes	-	Yes	Yes	Yes	-	-	-
CREDI - short	Yes	Yes	Yes	Yes	Yes	Yes	Yes	-	-	-	Yes
CREDI - long	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	-	-	Yes
EMPP	-	-	Yes	Yes	-	Yes	-	-	Yes	Yes	-
EMI	Yes	Yes	Yes	Yes	Yes	Yes	-	-	-	-	-
GRAB	-	-	-	-	-	Yes	-	-	-	-	Yes
HINE	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes

Tool	*Prone*	*Supine*	*Sitting*	*Standing*	*Locomotion*	*Hand use*	*Eyes*	*Movement quality and posture*	*Reflexes and reactions*	*Tone*	*Other*
HNNE	Yes	Yes	Yes	-	-	Yes	Yes	Yes	Yes	Yes	Yes
HAI	-	-	-	-	-	Yes	-	-	-	-	-
HINT	Yes	Yes	Yes	-	-	Yes	Yes	-	-	-	-
IYCD	Yes	Yes	Yes	Yes	Yes	Yes	Yes	-	-	-	Yes
Infant motor profile	Yes	Yes	Yes	Yes	Yes	Yes	-	Yes	Yes	-	Yes
INFANIB	Yes	Yes	Yes	Yes	-	Yes	-	Yes	Yes	Yes	Yes
Infant neuromotor assessment	Yes	Yes	Yes	-	-	-	-	-	Yes	Yes	-
MDAT	Yes	Yes	Yes	Yes	Yes	Yes	Yes	-	-	-	Yes
Milani-Comparetti	-	-	-	-	-	-	-	Yes	Yes	Yes	Yes
MAI	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Movement quality measure	-	-	-	-	-	Yes	-	Yes	-	Yes	-

Tool	*Prone*	*Supine*	*Sitting*	*Standing*	*Locomotion*	*Hand use*	*Eyes*	*Movement quality and posture*	*Reflexes and reactions*	*Tone*	*Other*
NNNS	Yes	Yes	Yes	-	Yes	Yes	Yes	Yes	Yes	Yes	Yes
NNDE	Yes	Yes	Yes	-	-	-	Yes	-	Yes	Yes	Yes
NMSDA	Yes	Yes	Yes	Yes	-	Yes	Yes	Yes	Yes	Yes	-
PediaTrac	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	-	-	-
PEDI-CAT	Yes	Yes	Yes	Yes	Yes	Yes	-	Yes	-	-	Yes
PEDS:DMS	Yes	Yes	-	-	-	Yes	Yes	Yes	-	-	Yes
GMs	-	-	-	-	-	-	-	Yes	-	-	-
GMs optimality (Pre-term GMs and writhing movements)	-	-	-	-	-	-	-	Yes	-	-	Yes
GMs optimality (3 months+)	-	Yes	-	-	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Rapid Neurodevelopmental Assessment	Yes	Yes	Yes	Yes	Yes	Yes	-	Yes	Yes	Yes	Yes
STEP	Yes	Yes	Yes	Yes	-	Yes	Yes	-	-	Yes	-

Tool	*Prone*	*Supine*	*Sitting*	*Standing*	*Locomotion*	*Hand use*	*Eyes*	*Movement quality and posture*	*Reflexes and reactions*	*Tone*	*Other*
Standardized Infant NeuroDevelopmental Assessment Neurological scale	Yes	Yes	Yes	-	-	Yes	Yes	-	-	-	-
Structured observation of motor performance	Yes	Yes	Yes	Yes	Yes	Yes	-	-	Yes	-	-
TIMP	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	-	-
TIMPSI	-	-	-	-	-	-	-	-	-	-	-
Test of motor and neurological function	-	-	Yes	-	-	-	-	-	Yes	Yes	-
Denver II	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	-	-	-
Neoneuro	-	Yes	Yes	-	-	Yes	Yes	Yes	Yes	Yes	Yes
NNE	Yes	-	Yes	-	Yes	-	Yes	Yes	Yes	Yes	Yes

Tool	*Prone*	*Supine*	*Sitting*	*Standing*	*Locomotion*	*Hand use*	*Eyes*	*Movement quality and posture*	*Reflexes and reactions*	*Tone*	*Other*
Touwen	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes	Yes

H. Information sheets used in the interview study described in Chapter 6.

H.1 Information sheet for a parent of a child with Cerebral Palsy

Jessica Baggaley
PhD Candidate
Population Health Sciences Institute
Newcastle University
Email: J.Baggaley2@newcastle.ac.uk
Tel: 01912 821 378

Participant information sheet for the Red Flags: Developing a questionnaire to facilitate early detection of Cerebral Palsy, based on early parental observations.

We would like to invite you to take part in a focus group as part of a research study. We are giving you this information pack before the focus group to help you make a decision on if you wish to take part. At the focus group one of our team will go through this information sheet with you. If you have any questions you may either contact Jessica Baggaley now (Tel:01912 821 378; Email: J.Baggaley2@newcastle.ac.uk) or ask at the focus group. This study is being undertaking towards obtaining a PhD qualification.

Background to the research

Not all infants with emerging signs of cerebral palsy (CP) are identified and referred promptly for further assessment and intervention. Parents sometimes comment that they have had worries about their child's development which have not been "heard" by healthcare professionals. One way to help improve this situation is to develop a Screening Tool to identify infants with emerging CP, using information from parents about the first concerning features they identified in their own children. We are developing a Screening Tool to help bridge the communication gap between parents and health care professionals. We hope that the Screening Tool will reduce the time taken for infants with emerging cerebral palsy to be referred for further assessment and input. We would like your help with the design and content of the Screening Tool.

Who can take part?

We are looking for parents and/or carers of children with a diagnosis of cerebral palsy. To take part;

- **Your child** must have a formal diagnosis of cerebral palsy.
- **You must** have been one of your child's main carers in the first 6 months of your child's life.
- **You must** be able to have a good ability to understand and speak English.
- **You must** be able to access Microsoft Teams
- **You must** have access to a microphone

What will happen if I agree to take part?

If you agree to take part we will ask you to take part in an online focus group and to answer a short questionnaire about yourself and your circumstances (such as your age, occupation and marital status). You can find an example of the questionnaire in this pack. A focus group is a guided group discussion involving

around 5-9 people. This group will be filled with parents and/or carers like yourself. We will ask you to give verbal consent to take part in the study. For verbal consent, you will be asked if you agree or disagree to each of the statements provided on the consent form. Your verbal consent and the focus group will both be audio recorded. There is an example of the consent form included in this pack. We will then ask you about your experiences with your child's referral, the earliest concerns you had about your child's development and about the Screening Tool we are developing. The focus group is expected to take between 1 hour to 1½ hours. We would like to audio record the focus group so that we do not miss anything important that you or another participant says. The audio recordings will be typed up word for word into a password protected Microsoft Word Document by a member of the research team. A member of the research team will then remove any identifiable information before we begin to analyse the data you provided.

Is the focus group confidential?

All participants will be reminded that anything said during the session should not be repeated elsewhere, nor should they identify people who participated to others. The information you give as a group will not be shared outside of our research group without your permission and any information shared will be anonymous. You may choose to give us permission to share your data by verbally agreeing to statement 5 of the consent form, an example consent form is included in your pack. The data that we analyse will also be anonymised and stored on a secure Newcastle University server only accessible to the research team. If any disclosures are made during the focus group which suggest malpractice, misconduct or that someone is in danger of harm we will discuss with you our need to share this information with the appropriate personnel. All the data we collect will be stored securely within Newcastle University in line with the Data Protection Act 1998, the General Data Protection Regulation (GDPR) legislation and the British Psychological Society Code of Ethics.

Once the study is over, we plan to upload the anonymous data generated in this study to the UK Data Service. The UK Data Service is an online platform, which allows researchers to share their data with other researchers, policy makers, students and teachers. The data generated by this study will only be available to the UK Data Service users who have signed the UK Data Service's End User Licence. In short, those who agree to the UK Data Service's End User Licence (2018) agree to '*to preserve the confidentiality of, and not attempt to identify, individuals, households or organisations in the data*' and that failure to comply with the Agreement will result in the termination of their access to the data and may result in legal action. A full version of the End User License is available at https://ukdataservice.ac.uk/media/455131/cd137-enduserlicence.pdf. We will only upload your anonymised data if you give us permission to share your data by initialling part 6 of the consent form.

Newcastle University will be using information from you in order to undertake this research study and will act as the data controller for this study. This means that Newcastle University is responsible for looking after your information and using it properly. When we use personally-identifiable information from people who have agreed to take part in research, we ensure that it is in the public interest. Your rights to access, change or move your information are limited, as Newcastle University needs to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, Newcastle University will keep the information about you that has already been obtained. To safeguard your rights, the minimum personally-identifiable information will be used. You can find out more about how Newcastle University uses your information at http://www.ncl.ac.uk/data.protection/PrivacyNotice and/or by contacting Newcastle University's Data Protection Officer (Maureen Wilkinson, rec-man@ncl.ac.uk).

We will use your name and contact details (Telephone number, address and email) to contact you about the research study. We will use information on yours and your child's demographics in order to ensure the

participants in the focus groups come from a range of backgrounds. Individuals at Newcastle University may look at your research data to check the accuracy of the research study. The only individuals at Newcastle University who will have access to information that identifies you will be individuals who need to contact you to clarify what you spoke about or audit the data collection process.

If you agree to take part in the research study, information provided by you may be shared with researchers running other research studies at Newcastle University and in other organisations. These organisations may be universities and NHS organisations. Your information will only be used by organisations and researchers to conduct research. This information will not identify you and will not be combined with other information in a way that could identify you. The information will only be used for the purpose research, and cannot be used to contact you. It will not be used to make decisions about future services available to you.

What will happen if I change my mind?

If you decide during the focus group that you no longer wish to take part, you may leave the room or not say anything further. You also do not need to answer all of our questions and you do not have to give any reasons for this. If you wish to retract something you said during the focus group or withdraw from the study after the focus group is over, you may do so without giving a reason. However, we will not be able to retract any data that has been analysed prior to your withdrawal.

What are the benefits and risks of taking part?

Unfortunately you will not directly benefit from taking part in this study, however you will be providing valuable information which may benefit others in the future. We will refund any travel expenses occurred due to focus group participation.

We appreciate that the topic being discussed may be upsetting. We will be sensitive and supportive to this throughout the focus group. The research team are also available to talk to you on a one-to-one basis. You may call or email the research team at any time to discuss any issues or needs for further support.

What will happen at the end of the study?

We will use your feedback alongside feedback collected from other focus groups to develop and improve a new screening tool for early cerebral palsy that we are developing. The outcomes of the focus group will be published as part of a PhD thesis and we do intend to publish an article about your experiences of the referral process and the concerns you presented to your child's health care professionals.

What if there is a problem?

Any complaints about how you dealt with during the study and any possible harm you suffer will be addressed. If you have any concerns or complaints about any aspect of this study, you should contact the Newcastle University Research Strategy and Development Team, who are independent of the research team, at res.policy@newcastle.ac.uk

Who is organising, funding and undertaking the research?

The study is funded through the Economic Social Research Councils Northern Ireland and North East Doctorial Training Programme to be carried out by Jessica Baggaley (PhD Student) under the supervision of

Dr Anna Basu (Clinical senior lecturer and honorary consultant child neurologist), Professor Tim Rapley (Medical sociologist) and Professor Nadja Reissland (Developmental psychologist).

Who has reviewed the study?

All research carried out in the NHS is reviewed by an independent group of people, called the Research Ethics Committee. The study has been reviewed and given ethical permission by Wales Research Ethics Committee 7.

Contact details for further information

If you would like any further information or if you have any questions or complaints please contact Jessica Baggaley (01912 821 378) at J.Baggaley2@newcastle.ac.uk or Dr Anna Basu at Anna.Basu@newcastle.ac.uk

Thank you for taking your time to read through this information sheet and for considering taking part in this study

H.2 Information sheet for parents of typically developing children

Jessica Baggaley
PhD Candidate
Population Health Sciences Institute
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Email: J.Baggaley2@newcastle.ac.uk
Tel: 01912 821 378

Participant information sheet for Red Flags: Developing a questionnaire to facilitate early detection of Cerebral Palsy, based on early parental observations.

We would like to invite you to take part in a focus group as part of a research study. We are giving you this information pack before the focus group to help you make a decision on if you wish to take part. At the focus group one of our team will go through this information sheet with you. If you have any questions you may either contact Jessica Baggaley now (Tel:01912 821 378; Email: J.Baggaley2@newcastle.ac.uk) or ask at the focus group. This study is being undertaking towards obtaining a PhD qualification.

Background to the research

Not all infants with emerging signs of cerebral palsy (CP) are identified and referred promptly for further assessment and intervention. Parents sometimes comment that they have had worries about their child's development which have not been "heard" by healthcare professionals. One way to help improve this situation is to develop a Screening Tool to identify infants with emerging CP, using information from parents about the first concerning features they identified in their own children. We are developing a Screening Tool to help bridge the communication gap between parents and health care professionals. We hope that the Screening Tool will reduce the time taken for infants with emerging cerebral palsy to be referred for further assessment and input. We would like your help with the design and content of the Screening Tool.

Who can take part?

We are looking for parents and/or carers of typically developing children. To take part;

- **Your child** must not have a formal diagnosis of cerebral palsy.
- **You must** have been one of your child's main carers in the first 6 months of your child's life.
- **You must** be able to have a good ability to understand and speak English.
- **You must** be able to access Microsoft Teams
- **You must** have access to a microphone

What will happen if I agree to take part?

If you agree to take part we will ask you to take part in an online focus group and to answer a short questionnaire about yourself and your circumstances (such as your age, occupation and marital status). You can find an example of the questionnaire in this pack. A focus group is a guided group discussion involving

around 5-9 people. This group will be filled with parents and/or carers like yourself. We will ask you to give verbal consent to take part in the study. For verbal consent, you will be asked if you agree or disagree to each of the statements provided on the consent form. Your verbal consent and the focus group will both be audio recorded. There is an example of the consent form included in this pack. We will then ask you about your experiences with your child's development and about the Screening Tool we are developing. The focus group is expected to take between 1 hour to 1½ hours. We would like to audio record the focus group so that we do not miss anything important that you or another participant says. The audio recordings will be typed up word for word into a password protected Microsoft Word Document by a member of the research team. A member of the research team will then remove any identifiable information before we begin to analyse the data you provided.

Is the focus group confidential?

All participants will be reminded that anything said during the session should not be repeated elsewhere, nor should they identify people who participated to others. The information you give as a group will not be shared outside of our research group without your permission and any information shared will be anonymous. You may choose to give us permission to share your data by verbally agreeing to statement 5 of the consent form, an example consent form is included in your pack. The data that we analyse will also be anonymised and stored on a secure Newcastle University server only accessible to the research team. If any disclosures are made during the focus group which suggest malpractice, misconduct or that someone is in danger of harm we will discuss with you our need to share this information with the appropriate personnel. All the data we collect will be stored securely within Newcastle University in line with the Data Protection Act 1998, the General Data Protection Regulation (GDPR) legislation and the British Psychological Society Code of Ethics.

Once the study is over, we plan to upload the anonymous data generated in this study to the UK Data Service. The UK Data Service is an online platform, which allows researchers to share their data with other researchers, policy makers, students and teachers. The data generated by this study will only be available to the UK Data Service users who have signed the UK Data Service's End User Licence. In short, those who agree to the UK Data Service's End User Licence (2018) agree to '*to preserve the confidentiality of, and not attempt to identify, individuals, households or organisations in the data*' and that failure to comply with the Agreement will result in the termination of their access to the data and may result in legal action. A full version of the End User License is available at https://ukdataservice.ac.uk/media/455131/cd137-enduserlicence.pdf. We will only upload your anonymised data if you give us permission to share your data by initialling part 6 of the consent form.

Newcastle University will be using information from you in order to undertake this research study and will act as the data controller for this study. This means that Newcastle University is responsible for looking after your information and using it properly. When we use personally-identifiable information from people who have agreed to take part in research, we ensure that it is in the public interest. Your rights to access, change or move your information are limited, as Newcastle University needs to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, Newcastle University will keep the information about you that has already been obtained. To safeguard your rights, the minimum personally-identifiable information will be used. You can find out more about how Newcastle University uses your information at http://www.ncl.ac.uk/data.protection/PrivacyNotice and/or by contacting Newcastle University's Data Protection Officer (Maureen Wilkinson, rec-man@ncl.ac.uk).

We will use your name and contact details (Telephone number, address and email) to contact you about the research study. We will use information on yours and your child's demographics in order to ensure the

participants in the focus groups come from a range of backgrounds. Individuals at Newcastle University may look at your research data to check the accuracy of the research study. The only individuals at Newcastle University who will have access to information that identifies you will be individuals who need to contact you to clarify what you spoke about or audit the data collection process.

If you agree to take part in the research study, information provided by you may be shared with researchers running other research studies at Newcastle University and in other organisations. These organisations may be universities and NHS organisations. Your information will only be used by organisations and researchers to conduct research. This information will not identify you and will not be combined with other information in a way that could identify you. The information will only be used for the purpose of research, and cannot be used to contact you. It will not be used to make decisions about future services available to you.

What will happen if I change my mind?

If you decide during the focus group that you no longer wish to take part, you may leave the room or not say anything further. You also do not need to answer all of our questions and you do not have to give any reasons for this. If you wish to retract something you said during the focus group or withdraw from the study after the focus group is over, you may do so without giving a reason. However, we will not be able to retract any data that has been analysed prior to your withdrawal.

What are the benefits and risks of taking part?

Unfortunately you will not directly benefit from taking part in this study, however you will be providing valuable information which may benefit others in the future. We will refund any travel expenses occurred due to focus group participation.

We appreciate that the topic being discussed may be upsetting. We will be sensitive and supportive to this throughout the focus group. The research team are also available to talk to you on a one-to-one basis. You may call or email the research team at any time to discuss any issues or needs for further support.

What will happen at the end of the study?

We will use your feedback alongside feedback collected from other focus groups to develop and improve a new screening tool for early cerebral palsy that we are developing. The outcomes of the focus group will be published as part of a PhD thesis and we do intend to publish an article about your experiences of the referral process and the concerns you presented to your child's health care professionals.

What if there is a problem?

Any complaints about how you dealt with during the study and any possible harm you suffer will be addressed. If you have any concerns or complaints about any aspect of this study, you should contact the Newcastle University Research Strategy and Development Team, who are independent of the research team, at res.policy@newcastle.ac.uk.

Who is organising, funding and undertaking the research?

The study is funded through the Economic Social Research Councils Northern Ireland and North East Doctorial Training Programme to be carried out by Jessica Baggaley (PhD Student) under the supervision of

Dr Anna Basu (Clinical senior lecturer and honorary consultant child neurologist), Professor Tim Rapley (Medical sociologist) and Professor Nadja Reissland (Developmental psychologist).

Who has reviewed the study?

All research carried out in the NHS is reviewed by an independent group of people, called the Research Ethics Committee. The study has been reviewed and given ethical permission by Wales Research Ethics Committee 7.

Contact details for further information

If you would like any further information or if you have any questions please contact Jessica Baggaley (01912 821 378) at J.Baggaley2@newcastle.ac.uk or Dr Anna Basu at Anna.Basu@newcastle.ac.uk

Thank you for taking your time to read through this information sheet and for considering taking part in this study

H.3 Information sheets for primary health care professionals

Jessica Baggaley
PhD Candidate
Population Health Sciences Institute
Newcastle University
Email: J.Baggaley2@newcastle.ac.uk
Tel: 01912 821 378

Participant information sheet for the Red Flags: Developing a questionnaire to facilitate early detection of Cerebral Palsy, based on early parental observations.

We would like to invite you to take part in a focus group as part of a research study. We are giving you this information pack before the focus group to help you make a decision on if you wish to take part. At the focus group one of our team will go through this information sheet with you. If you have any questions you may either contact Jessica Baggaley now (Tel:01912 821 378; Email: J.Baggaley2@newcastle.ac.uk) or ask at the focus group. This study is being undertaking towards obtaining a PhD qualification.

Background to the research

Not all infants with emerging signs of cerebral palsy (CP) are identified and referred promptly for further assessment and intervention. Parents sometimes comment that they have had worries about their child's development which have not been "heard" by healthcare professionals. One way to help improve this situation is to develop a Screening Tool to identify infants with emerging CP, using information from parents about the first concerning features they identified in their own children. We are developing a Screening Tool to help bridge the communication gap between parents and health care professionals. We hope that the Screening Tool will reduce the time taken for infants with emerging cerebral palsy to be referred for further assessment and input. We would like your help with the design and content of the Screening Tool.

Who can take part?

We are looking for primary health care professionals who work with infants. To take part;

- **You must** be involved in early referral or be involved in early developmental assessments of infants
- **You must** be able to have a good ability to understand and speak English.
- **You must** be able to access Microsoft Teams
- **You must** have access to a microphone

What will happen if I agree to take part?

If you agree to take part we will ask you to take part in an online focus group and to answer a short questionnaire about yourself and your circumstances (such as your age, occupation and marital status). You can find an example of the questionnaire in this pack. A focus group is a guided group discussion involving around 5-9 people. This group will be filled with primary health care professionals who work with infants,

like yourself. We will ask you to give verbal consent to take part in the study. For verbal consent, you will be asked if you agree or disagree to each of the statements provided on the consent form. Your verbal consent and the focus group will both be audio recorded. There is an example of the consent form included in this pack. We will then ask you about the parent concerns, or "red flags", you would refer an infant onto therapy for and about the Screening tool we are developing. The focus group is expected to take between 1 to 1½ hours. We would like to audio record the focus group so that we do not miss anything important that you or another participant says. The audio recordings will be typed up word for word into a password protected Microsoft Word Document by a member of the research team. A member of the research team will then remove any identifiable information before we begin to analyse the data you provided.

After the focus group, we would like to send you a copy of the final draft of the screening tool. We hope that you will be able to provide us with some more feedback about the screening tools design and to let us know how relevant you think the final questions are. However, we will only do this if you give us permission too.

Is the focus group confidential?

All participants will be reminded that anything said during the session should not be repeated elsewhere, nor should they identify people who participated to others. The information you give as a group will not be shared outside of our research group without your permission. You may choose to give us permission to share your data by verbally agreeing to statement 5 of the consent form, an example consent form is included in your pack. The data that we analyse will be anonymised and stored on a secure Newcastle University server only accessible to the research team. If any disclosures are made during the focus group which suggest malpractice, misconduct or that someone is in danger of harm we will discuss with you our need to share this information with the appropriate personnel. All the data we collect will be stored securely within Newcastle University in line with the Data Protection Act 1998, the General Data Protection Regulation (GDPR) legislation and the British Psychological Society Code of Ethics.

Once the study is over, we plan to upload the anonymous data generated in this study to the UK Data Service. The UK Data Service is an online platform, which allows researchers to share their data with other researchers, policy makers, students and teachers. The data generated by this study will only be available to the UK Data Service users who have signed the UK Data Service's End User Licence. In short, those who agree to the UK Data Service's End User Licence (2018) agree to '*to preserve the confidentiality of, and not attempt to identify, individuals, households or organisations in the data*' and that failure to comply with the Agreement will result in the termination of their access to the data and may result in legal action. A full version of the End User License is available at https://ukdataservice.ac.uk/media/455131/cd137-enduserlicence.pdf. We will only upload your anonymised data if you give us permission to share your data by initialling part 6 of the consent form.

Newcastle University will be using information from you in order to undertake this research study and will act as the data controller for this study. This means that Newcastle University is responsible for looking after your information and using it properly. When we use personally-identifiable information from people who have agreed to take part in research, we ensure that it is in the public interest. Your rights to access, change or move your information are limited, as Newcastle University needs to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, Newcastle University will keep the information about you that has already been obtained. To safeguard your rights, the minimum personally-identifiable information will be used. You can find out more about how Newcastle University uses your information at http://www.ncl.ac.uk/data.protection/PrivacyNotice and/or by contacting Newcastle University's Data Protection Officer (Maureen Wilkinson, rec-man@ncl.ac.uk).

We will use your name and contact details (telephone number and email address] to contact you about the research study. We will use your demographic information, such as your job title, in order to ensure that the

focus groups are attended by health care professionals with a wide range of relevant experience. Individuals at Newcastle University may look at your research data to check the accuracy of the research study. The only individuals at Newcastle University who will have access to information that identifies you will be individuals who need to contact you to attend another focus group, to clarify something you said during the focus group or audit the data collection process.

If you agree to take part in the research study, information provided by you may be shared with researchers running other research studies at Newcastle University and in other organisations. These organisations may be universities and NHS organisations. Your information will only be used by organisations and researchers to conduct research.

This information will not identify you and will not be combined with other information in a way that could identify you. The information will only be used for the purpose research, and cannot be used to contact you. It will not be used to make decisions about future services available to you.

What will happen if I change my mind?

If you decide during the focus group that you no longer wish to take part, you may leave the room or not say anything further. You also do not need to answer all of our questions and you do not have to give any reasons for this. If you wish to retract something you said during the focus group or withdraw from the study after the focus group is over, you may do so without giving a reason. However, we will not be able to retract any data that has been analysed prior to your withdrawal.

What are the benefits and risks of taking part?

Unfortunately you will not directly benefit from taking part in this study, however you will be providing valuable information which may benefit your practice and/or help others in the future. We will refund any travel expenses occurred due to focus group participation.

What will happen at the end of the study?

We will use your feedback alongside feedback collected from other focus groups to develop and improve a new screening tool for early cerebral palsy that we are developing. The outcomes of the focus group will be published as part of a PhD thesis and we do intend to publish an article about your experiences of the referral process and the concerns you presented to your child's health care professionals. You will not be identifiable in any of these publications.

What if there is a problem?

Any complaints about how you dealt with during the study and any possible harm you suffer will be addressed. If you have any concerns or complaints about any aspect of this study, you should contact the Newcastle University Research Strategy and Development Team, who are independent of the research team, at res.policy@newcastle.ac.uk.

Who is organising, funding and undertaking the research?

The study is funded through the Economic Social Research Councils Northern Ireland and North East Doctorial Training Programme to be carried out by Jessica Baggaley (PhD Student) under the supervision of Dr Anna Basu (Clinical senior lecturer and honorary consultant child neurologist), Professor Tim Rapley (Medical sociologist) and Professor Nadja Reissland (Developmental psychologist).

Who has reviewed the study?

All research carried out in the NHS is reviewed by an independent group of people, called the Research Ethics Committee. The study has been reviewed and given ethical permission by Wales Research Ethics Committee 7.

Contact details for further information

If you would like any further information or if you have any questions or complaints please contact Jessica Baggaley (01912 821 378) at J.Baggaley2@newcastle.ac.uk or Dr Anna Basu at Anna.Basu@newcastle.ac.uk

Thank you for taking your time to read through this information sheet and for considering taking part in this study

H.4 Information sheet for secondary health care professionals

Information sheet for Secondary Health Care Professionals V1.5 10/06/2020 IRAS Number 261048

Jessica Baggaley
PhD Candidate
Population Health Sciences Institute
Newcastle University
Email: J.Baggaley2@newcastle.ac.uk
Tel: 01912 821 378

Participant information sheet for Red Flags: Developing a questionnaire to facilitate early detection of Cerebral Palsy, based on early parental observations.

We would like to invite you to take part in a focus group as part of a research study. We are giving you this information pack before the focus group to help you make a decision on if you wish to take part. At the focus group one of our team will go through this information sheet with you. If you have any questions you may either contact Jessica Baggaley now (Tel:01912 821 378; Email: J.Baggaley2@newcastle.ac.uk) or ask at the focus group. This study is being undertaking towards obtaining a PhD qualification.

Background to the research

Not all infants with emerging signs of cerebral palsy (CP) are identified and referred promptly for further assessment and intervention. Parents sometimes comment that they have had worries about their child's development which have not been "heard" by healthcare professionals. One way to help improve this situation is to develop a Screening Tool to identify infants with emerging CP, using information from parents about the first concerning features they identified in their own children. We are developing a Screening Tool to help bridge the communication gap between parents and health care professionals. We hope that the Screening Tool will reduce the time taken for infants with emerging cerebral palsy to be referred for further assessment and input. We would like your help with the design and content of the Screening Tool.

Who can take part?

Health care professionals who either treat or diagnose infants developing cerebral palsy. To take part;

- **You must** either provide treatment to or diagnose infants developing cerebral palsy
- **You must** be able to have a good ability to understand and speak English.
- **You must** be able to access Microsoft Teams
- **You must** have access to a microphone

What will happen if I agree to take part?

If you agree to take part we will ask you to take part in an online focus group and to answer a short questionnaire about yourself and your circumstances (such as your age, occupation and marital status). You can find an example of the questionnaire in this pack. A focus group is a guided group discussion involving around 5-9 people. This group will be filled with health care professionals with relevant experience in early cerebral palsy, like yourself. We will ask you to give verbal consent to take part in the study. For verbal

consent, you will be asked if you agree or disagree to each of the statements provided on the consent form. Your verbal consent and the focus group will both be audio recorded. There is an example of the consent form included in this pack. We will then ask you about the concerns parents raise to you and the "red flags" you see in their child's development and about the Screening Tool we are developing. The focus group is expected to take between 1 to 1½ hours. We would like to audio record the focus group so that we do not miss anything important that you or another participant says. The audio recordings will be typed up word for word into a password protected Microsoft Word Document by a member of the research team. A member of the research team will then remove any identifiable information before we begin to analyse the data you provided.

After the focus group, we would like to send you a copy of the final draft of the screening tool. We hope that you will be able to provide us with some more feedback about the screening tools design and to let us know how relevant you think the final questions are. However, we will only do this if you give us permission too.

Is the focus group confidential?

All participants will be reminded that anything said during the session should not be repeated elsewhere, nor should they identify people who participated to others. The information you give as a group will not be shared outside of our research group without your permission. You may choose to give us permission to share your data by verbally agreeing to statement 5 of the consent form, an example consent form is included in your pack. The data that we analyse will be anonymised and stored on a secure Newcastle University server only accessible to the research team. If any disclosures are made during the focus group which suggest malpractice, misconduct or that someone is in danger of harm we will discuss with you our need to share this information with the appropriate personnel. All the data we collect will be stored securely within Newcastle University in line with the Data Protection Act 1998, the General Data Protection Regulation (GDPR) legislation and the British Psychological Society Code of Ethics.

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What will happen if I change my mind?

If you decide during the focus group that you no longer wish to take part, you may leave the room or not say anything further. You also do not need to answer all of our questions and you do not have to give any reasons for this. If you wish to retract something you said during the focus group or withdraw from the study after the focus group is over, you may do so without giving a reason. However, we will not be able to retract any data that has been analysed prior to your withdrawal.

What are the benefits and risks of taking part?

Unfortunately you will not directly benefit from taking part in this study, however you will be providing valuable information which may benefit your practice and help others in the future. We will refund any travel expenses occurred due to focus group participation.

What will happen at the end of the study?

We will use your feedback alongside feedback collected from other focus groups to develop and improve a new screening tool for early cerebral palsy that we are developing. The outcomes of the focus group will be published as part of a PhD thesis and we do intend to publish an article about your experiences of the referral process and the concerns you presented to your child's health care professionals.

What if there is a problem?

Any complaints about how you dealt with during the study and any possible harm you suffer will be addressed. If you have any concerns or complaints about any aspect of this study, you should contact the Newcastle University Research Strategy and Development Team, who are independent of the research team, at res.policy@newcastle.ac.uk.

Who is organising, funding and undertaking the research?

The study is funded through the Economic Social Research Councils Northern Ireland and North East Doctorial Training Programme to be carried out by Jessica Baggaley (PhD Student) under the supervision of Dr Anna Basu (Clinical senior lecturer and honorary consultant child neurologist), Professor Tim Rapley (Medical sociologist) and Professor Nadja Reissland (Developmental psychologist).

Who has reviewed the study?

All research carried out in the NHS is reviewed by an independent group of people, called the Research Ethics Committee. The study has been reviewed and given ethical permission by Wales Research Ethics Committee 7.

Contact details for further information

If you would like any further information or if you have any questions or complaints please contact Jessica Baggaley (01912 821 378) at J.Baggaley2@newcastle.ac.uk or Dr Anna Basu at Anna.Basu@newcastle.ac.uk

Thank you for taking your time to read through this information sheet and for considering taking part in this study

H.5 Consent form for Parents and caregivers of children with Cerebral Palsy

Parent/Carer Consent form V1.4, 10/06/2020 IRAS Number: 261048

Participant Consent Form for the Red Flags: Developing a questionnaire to facilitate early detection of Cerebral Palsy, based on early parental observations.

If you would like to take part in the Red Flags: Focus Group, please place your initials in each of the boxes below and sign and date this form.

All of the information on this form will be kept confidential and will not be released to anyone outside of the research team.

	Please *initial* each box
1. I confirm I have read and understood the information sheet [Parent and Carer Information Sheet V1.4, 10/06/2020] for the above study	☐
2. I have had the opportunity to consider the information, ask questions and I have had my questions answered satisfactorily.	☐
3. I understand that I am free to withdraw from the study at any time without giving a reason and without any adverse consequences.	☐
4. I give permission for my contact details to be kept by the research team to inform me on how the study is progressing.	☐
5. I give permission for anonymised quotes from the focus group to be used in publications and presentations related to the study.	☐
6. I understand that the research collected about me may be used to support other research in the future and can be shared anonymously with other researchers.	☐
7. I understand that records relating to me will be kept confidential and that no information will be released or printed that would identify me without my permission, unless required by law.	☐
8. I agree to not share any information given by other participants during the focus groups with others	☐
9. I agree to not identify the other focus group participants after the focus group	☐
10. I confirm that I am willing to allow the focus group to be audio recorded	☐
11. I agree to take part in the focus group	☐

Name of Participant (*please print*)	Date (*DD/MM/YY*)	Participant's Signature

Name of person taking consent (*please print*)	Date (*DD/MM/YY*)	Person taking consent's Signature

When completed: 1 for participant; 1 for Newcastle University

H.6 Consent form for parent and carers of typically developing children

Parent/Carer Consent form V1.4, 10/06/2020 IRAS Number: 261048

Participant Consent Form for the Red Flags: Developing a questionnaire to facilitate early detection of Cerebral Palsy, based on early parental observations.

If you would like to take part in the Red Flags: Focus Group, please place your initials in each of the boxes below and sign and date this form.

All of the information on this form will be kept confidential and will not be released to anyone outside of the research team.

	Please *initial* each box
1. I confirm I have read and understood the information sheet [Parent and Carer Information Sheet TD Children V1.4, 10/06/2020] for the above study	
2. I have had the opportunity to consider the information, ask questions and I have had my questions answered satisfactorily.	
3. I understand that I am free to withdraw from the study at any time without giving a reason and without any adverse consequences.	
4. I give permission for my contact details to be kept by the research team to inform me on how the study is progressing.	
5. I give permission for anonymised quotes from the focus group to be used in publications and presentations related to the study.	
6. I understand that the research collected about me may be used to support other research in the future and can be shared anonymously with other researchers.	
7. I understand that records relating to me will be kept confidential and that no information will be released or printed that would identify me without my permission, unless required by law.	
8. I agree to not share any information given by other participants during the focus groups with others	
9. I agree to not identify the other focus group participants after the focus group	
10. I confirm that I am willing to allow the focus group to be audio recorded	
11. I agree to take part in the focus group	

Name of Participant (*please print*)	Date (*DD/MM/YY*)	Participant's Signature
Name of person taking consent (*please print*)	Date (*DD/MM/YY*)	Person taking consent's Signature

When completed: 1 for participant; 1 for Newcastle University

H.7 Consent form for primary health care professionals

Primary Health Care Professionals Consent form V1.5 10/06/2020 IRAS Number: 261048

Participant Consent Form for the Red Flags: Developing a questionnaire to facilitate early detection of Cerebral Palsy, based on early parental observations.

If you would like to take part in the Red Flags: Focus Group, please place your initials in each of the boxes below and sign and date this form.

All of the information on this form will be kept confidential and will not be released to anyone outside of the research team.

	Please *initial* each box
1. I confirm I have read and understood the information sheet [Primary Health Care Professional Information Sheet V1.5, 10/06/2020] for the above study	
2. I have had the opportunity to consider the information, ask questions and I have had my questions answered satisfactorily.	
3. I understand that I am free to withdraw from the study at any time without giving a reason and without any adverse consequences.	
4. I give permission for my contact details to be kept by the research team to inform me on how the study is progressing	
5. I give permission for my contact details to be kept by the research team to send me the final draft copy of the screening tool so that I may provide feedback on it.	
6. I give permission for anonymised quotes from the focus group to be used in publications and presentations related to the study.	
7. I understand that the research collected about me may be used to support other research in the future and can be shared anonymously with other researchers.	
8. I understand that records relating to me will be kept confidential and that no information will be released or printed that would identify me without my permission, unless required by law.	
9. I agree to not share any information given by other participants during the focus groups with others	
10. I agree to not identify the other focus group participants after the focus group	
11. I confirm that I am willing to allow the focus group to be audio recorded	
12. I agree to take part in the focus group	

Name of Participant (*please print*)	Date (*DD/MM/YY*)	Participant's Signature
Name of person taking consent (*please print*)	Date (*DD/MM/YY*)	Person taking consent's Signature

When completed: 1 for participant; 1 for Newcastle University

H.8 Consent form for secondary health care professionals

Secondary Health Care Professionals Consent form V1.4, 10/06/2020 IRAS Number: 261048

Durham University NINE Doctoral Training in the Social Sciences North East of England and Northern Ireland E·S·R·C ECONOMIC & SOCIAL RESEARCH COUNCIL Newcastle University

Participant Consent Form for Red Flags: Developing a questionnaire to facilitate early detection of Cerebral Palsy, based on early parental observations.

If you would like to take part in the Red Flags: Focus Group, please place your initials in each of the boxes below and sign and date this form.

All of the information on this form will be kept confidential and will not be released to anyone outside of the research team.

	Please *initial* each box
1. I confirm I have read and understood the information sheet [Secondary Health Care Professionals Information Sheet V1.5, 10/06/2020] for the above study	
2. I have had the opportunity to consider the information, ask questions and I have had my questions answered satisfactorily.	
3. I understand that I am free to withdraw from the study at any time without giving a reason and without any adverse consequences.	
4. I give permission for my contact details to be kept by the research team to inform me on how the study is progressing	
5. I give permission for my contact details to be kept by the research team to send me the final draft copy of the screening tool so that I may provide feedback on it.	
6. I give permission for anonymised quotes from the focus group to be used in publications and presentations related to the study.	
7. I understand that the research collected about me may be used to support other research in the future and can be shared anonymously with other researchers.	
8. I understand that records relating to me will be kept confidential and that no information will be released or printed that would identify me without my permission, unless required by law.	
9. I agree to not share any information given by other participants during the focus groups with others	
10. I agree to not identify the other focus group participants after the focus group	
11. I confirm that I am willing to allow the focus group to be audio recorded	
12. I agree to take part in the focus group	

Name of Participant (*please print*)	Date (*DD/MM/YY*)	Participant's Signature
Name of person taking consent (*please print*)	Date (*DD/MM/YY*)	Person taking consent's Signature

When completed: 1 for participant; 1 for Newcastle University

H.9 Sociodemographic questionnaire for parents and caregivers

Parent/Carer SES V1 28.11.2018

Red Flags: Focus Groups

Sociodemographic questionnaire

Name			
Age			
Gender			
Number of children at home under 18			
Postcode			
Ethnicity	White/European	Pakistani	Chinese
	Black African	Indian	Black other
	Black Caribbean	Bangladeshi	Asian other
	Mixed (Please specify):		
	Other (Please specify):		
	Prefer not to say		
What is the highest level of education you have completed?	No formal qualifications		GCSE level or equivalent
	A levels or equivalent		University degree (eg. BSc, MA, PhD or equivalent)
	Other:		
	Prefer not to say		
Employment Status	Employed Full-time	Employed Part time	Full time homemaker
	Full time carer	Unemployed and looking for work	Unemployed due to health
	Retired	Doing voluntary work	Full time student
	Other (please specify)		
Occupation:			
Household income	Less than 14,000	14,000 - 17,999	18,000 - 30,999
	31,000 - 51,999	52,000 – 100,000	Greater than 100,000
	Do not know	Prefer not to say	
Marital status	Married/civil partnership/co-habiting with long term partner		Divorced/separated
	Single		Widowed

H.10 Sociodemographic questionnaire for health care professionals

2ndry HCPs SES V1 28.11.2018

Red Flags: Focus Groups

Sociodemographic questionnaire

Name			
Age (years)			
Gender			
Postcode			
Ethnicity	White/European	Pakistani	Chinese
	Black African	Indian	Black other
	Black Caribbean	Bangladeshi	Asian other
	Mixed (Please specify):		
	Other (Please specify):		
	Prefer not to say		
What is the highest level of education you have completed?	No formal qualifications		GCSE level or equivalent
	A levels or equivalent		University degree (eg. BSc, MA, PhD or equivalent)
	Other:		
	Prefer not to say		
Employment Status	Employed Full-time	Employed Part time	Full time homemaker
	Full time carer	Unemployed and looking for work	Unemployed due to health
	Retired	Doing voluntary work	Full time student
	Other (please specify)		
Occupation:			
Years of working experience relevant to Infant development			
Household income	Less than 14,000	14,000 - 17,999	18,000 - 30,999
	31,000 - 51,999	52,000 – 100,000	Greater than 100,000
	Do not know	Prefer not to say	
Marital status	Married/civil partnership/co-habiting with long term partner		Divorced/separated
	Single		Widowed

H.11 Recruitment e-flyer

Durham University

NINE Doctoral Training in the Social Sciences North East of England and Northern Ireland

E·S·R·C Economic & Social Research Council

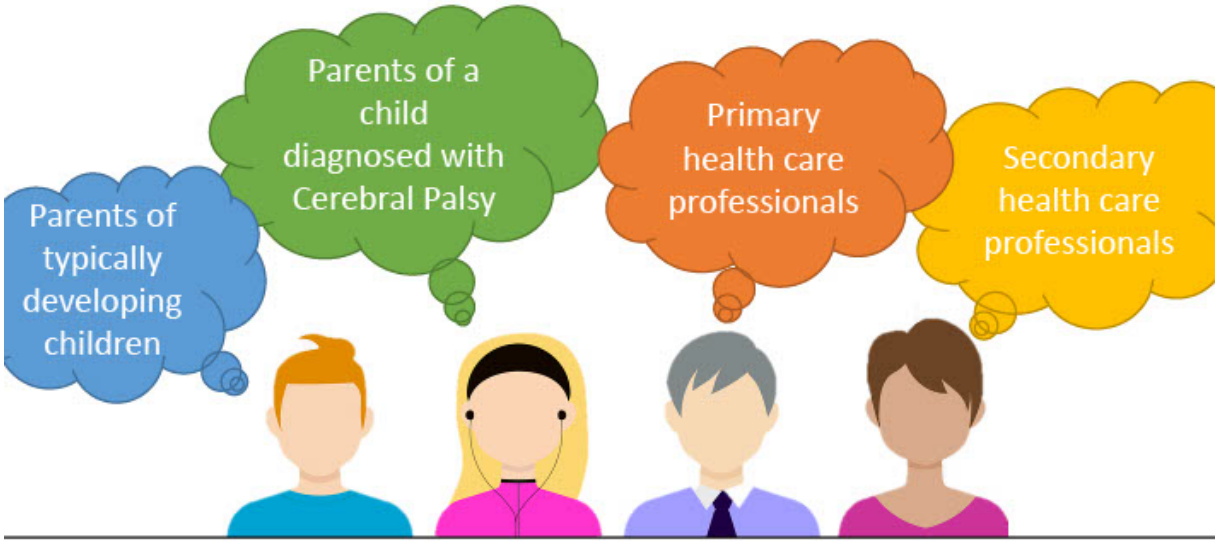
Newcastle University

Help us to

SUPPORT NEW PARENTS

to report their concerns about their child's development

We are developing a new tool to detect infants at risk of developing movement problems, but we need your help!
Please come along to one of our online focus groups:



Focus groups 1 & 5
For Parents of a child diagnosed with Cerebral Palsy
Focus group 2
For Secondary health care professionals
Focus group 3
For Parents of typically developing children
Focus group 4
For Primary health care professionals

To find out more and to register your interest please contact Jessica Baggaley at
J.Baggaley2@newcastle.ac.uk
or at
01912 821 378

I. Short information sheet, final version

Baby development, what usually happens and what to look out for.

This guide is to give you some information on how babies usually develop. Every baby develops differently and they learn new skills in their own time. Often one baby can take along time to learn a skill that another baby learnt quickly, so comparing your baby's development to another's may not be helpful. Instead try to use this guide to check on how your baby is developing.

This guide also contains some things to look out for, such as when your baby should be learning new skills and some behaviors that could be a cause for concern. Although it is unlikely that your baby will have any concerning behaviours, it is important that if your baby does have them we catch them early. These behaviors could happen when your baby is in the bath, when they are playing, or when they are sleeping. If you spot your baby doing one of the concerning behaviors in this guide or you are just worried about your baby, you should contact your Health Visitor or General Practitioner to get you baby checked out and put your mind at rest.

About this guide

The guide is broken up into the types of concerns you may have, this includes;

You just have a concern, Your baby's movement, Stiffness and floppiness, Skill development, and Further information.

Sometimes this guide talks about your baby's due date. Your baby's due date is the date you were told your baby was going to be born, not the date they were actually born. Concerns and skills related to your baby's age are highlighted in coloured boxes.

Infections, injuries, and medical conditions can slow your baby's development, meaning that your baby may begin to do age related things, like learning to sit, when they are older than suggested in this guide. If your baby has had an infection or injury, or has a medical condition, you may want to talk to your GP, Health Visitor, or Consultant about what to expect with your baby's development.

How to use this guide

There are three ways you can use this guide.

If you have concerns,

- You can use the sections of the guide to identify if your baby is showing concerning behaviour
- You can look at the age related skills highlighted in the coloured boxes to see if your baby is developing as expected

If you do not have concerns,

- You can use the sections or the age related skills highlighted in the coloured boxes to learn how babies usually develop and find further advice on how to support your baby's development.

You just have a concern

You may just have a feeling that something in not right with your baby. If so you should speak to your GP or Health Visitor to check it out and help put your mind at rest.

Your baby's movement

Babies usually move both sides of their body about the same amount and are able to do the same things on each side. As babies grows their movements usually begin to look more fluid and wigglier, they begin to hold their hands together, they begin to hold their feet, and they begin to use both of their hands together to hold larger objects such as a ball. You should speak to your health visitor or GP if you notice one of the following persistently occurring:

Baby moving one side of their body

- Your baby always moves one side of their body more than the other, such as;
  - Rarely using one of their arms for tasks such as picking toys up or has a clear hand preference
  - Rarely kicking or moving one leg compared to the other, or has a clear leg preference
- Your baby's movements appear jittery, stiff, rigid, or jerky, and do not appear to become more fluid and wiggly
- Your baby does not hold their hands or bring both feet to their chest. Babies usually start to do these from around 4 months after their due date*.
- Your baby does not use two hands together, such as to hold larger objects. Babies usually do this from around 6 months after their due date*.
- Your baby is often startling and you cannot figure out why. All babies usually startle when they hear a loud sound, see a sudden movement or feel like they are falling. Every baby is born with a startle reflex that begins to fade away from 6 weeks past their due date and disappears by the time your baby is 6 months past their due date.

Baby bringing their hands and feet into their chest

Stiffness or Floppiness

As babies grow and develop strength they usually begin to mould to their parents when being held or during feeding. When moving your baby's limbs you may feel some resistance, however this usually does not stop parents gently moving their baby's limbs. Sometimes babies can feel floppy, such as when they are asleep or just after birth. You should speak to your health visitor or GP if you notice one of the following persistently occurring:

- Your baby does not mould to you when you hold them or you need to continue to give them extra support when you hold them 3-4 months after their due date*.
- Your baby's body, arms, or legs feel stiff or rigid making tasks such as changing your baby's nappy or clapping your baby's hands together difficult.
- Your baby's fist(s) do not become looser over time and you may struggle to open their hand(s) for them

* Your baby's due date is the date you were told your baby was going to be born, not when they were actually born.

- Your baby may hold their arms and legs in the same position, such as always pulling their arm into their chest, or always crossing their legs. Your baby may have difficulties changing their position on their own and you may also have difficulties when positioning your baby
- Your baby's body, arms, or legs feel floppy even when they are awake and you need to continue to support your baby's head when you hold them 3-4 months after their due date*.
- Your baby feels both stiff and floppy in different areas of their body

Skills Development

As babies grow they usually begin to learn new skills such as sitting, crawling, walking, grasping, and talking. Babies learn these skills in their own time, so comparing your baby's development to another babies development may not be helpful. If you do notice any of the below you should contact your GP or Health Visitor.

- Your baby begins to learn new skills but never fully learns them. For example your baby may learn to roll in one direction but does not try to learn to roll in the other direction or your baby may not try to crawl without dragging one of their limbs behind them.

Baby grasping or holding a toy in one hand

8 weeks after your baby's due date*

- Your baby has not began to fix and follow toys or people with their eyes, even for just a couple of seconds.

6 months after your baby's due date*

- Your baby has not began to recognize their own name, show emotions like excitement, or started babbling
- Yor baby has not began to shown any signs of learning how to grasp toys in one or both hands without you placing toys in their hand.

Baby sitting on their own

9 months after your baby's due date*

- Your baby has not began to sit without you supporting them

14 months after your baby's due date*

- Your baby has not began to show any signs of learning to move, such as crawling or walking,
- Your baby has not said their first word (such as Mama or Dada) and does not seem to understand the meaning of any words you say to them, such as drink.

Baby crawling

Further information

This is just a short guide to some of the concerns you may have. More information about the concerns in this guide and concerns about your baby's Hearing and vision, Feeding, Growth, Temperament and Sleep, and where you can find additional help and information can be found at

[website address]

[QR code].

* Your baby's due date is the date you were told your baby was going to be born, not when they were actually born.

J. Long information sheet, final version

Baby development, what usually happens and what to look out for

This guide is to give you some information on how babies usually develop. Every baby develops differently and they learn new skills in their own time. Often one baby can take along time to learn a skill that another baby learnt quickly, so comparing your baby's development to another's may not be helpful. Instead try to use this guide to check on how your baby is developing.

This guide also contains some things to look out for, such as when your baby should be learning new skills and some behaviors that could be a cause for concern. Although it is unlikely that your baby will have any concerning behaviors, it is important that if your baby does have them we catch them early. These behaviors could happen when your baby is in the bath, when they are playing, or when they are sleeping. If you spot your baby doing one of the concerning behaviors in this guide or you are just worried about your baby, you should contact your Health Visitor or General Practitioner to get your baby checked out and to put your mind at rest.

About this guide

The guide is broken up into the types of concerns you may have, this includes;

Parental instinct, Your baby's movement, Posture, Stiffness and floppiness, Reflexes and reactions, Skill development, Speaking and smiling, Eye gaze, Temperament and Sleep, Feeding difficulties, and Growth.

Each section contains a brief introduction explaining what the section is about, gives an example of what babies usually do, and explains what concerning things to look out for. Some sections also have weblinks and/or phone numbers to further information and advice.

Some of the things to look out for talk about your baby's due date. Your baby's due date is the date you were told your baby was going to be born, not the date they were actually born. Concerns and skills related to your baby's age are highlighted in coloured boxes.

Infections, injuries, and medical conditions can slow your babies development, meaning that your baby may begin to do age related things, like learning to sit, when they are older than suggested in this guide. If your baby has had an infection or injury, or has a medical condition, you may want to talk to your GP, Health Visitor, or Consultant about what to expect with your baby's development.

How to use this guide

There are three ways you can use this guide.

If you have concerns,

- You can use the sections of the guide to identify if your baby is showing concerning behaviour and to find further information on how to support your baby and who to contact.
- You can look at the age related skills highlighted in the coloured boxes to see if your baby is developing as expected

If you do not have concerns,

- You can use the sections or the age related skills highlighted in the coloured boxes to learn how babies usually develop and find further advice on how to support your baby's development.

* Your baby's due date is the date you were told your baby was going to be born, not when they were actually born.

Parental instinct

- You have a feeling that something in not right with your baby.

Sometimes parents have a feeling that something is not quite right about their baby, even when their friends and family tell them that their baby is okay and to try not to worry. It may be helpful to look through the following concerns to help you identify what is concerning you. It may be that you are not able to identify what is concerning you and that is okay, you should still contact your Health Visitor or General Practitioner.

Your baby's movement

Babies usually move both sides of their body about the same amount and can do the same things on each side. As babies grows their movements usually begin to look more fluid and wigglier, they begin to hold their hands together and bring both feet into their chest and they begin to use both of their hands together to hold larger objects such as a ball. You should speak to your health visitor or GP if you notice one of the following persistently occurring:

- Your baby always moves one side of their body more than the other, such as;
    - Rarely using one of their arms for tasks such as picking toys up or has a clear hand preference
    - Rarely kicking or moving one leg compared to the other
- Your baby's movements appear jittery, stiff, rigid, or jerky and do not appear to become more fluid and wigglier
- Your baby appears to have a lack of strength, or a general lack of movement compared to other babies.


Baby moving one side of their body

- Your baby does not hold their hands or bring both feet to their chest. Babies usually start to do these from 4 months after their due date*.
- Your baby does not use two hands together, such as to hold larger objects. Babies usually do this from 6 months after their due date*.


Baby bringing their hands and feet into their chest

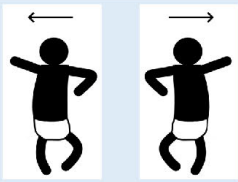
Posture

Usually, babies can change the position their body is in by themselves. When you hold your baby, you may notice that they do not mould to you or that you need to give them extra support. Below are examples of things you may notice when holding your baby or when your baby is sitting or is laid on their back. You should speak to your health visitor or GP if you notice one of the following persistently occurring or suddenly starts happening:

- **Arm** – Your baby's arm(s) is often kept in the same position. Such as raised by their head, beside the body, tucked or pulled into their chest, turned outwards, or twisted behind their back.
- **Body** – Your baby's body is twisted, arched, extend backwards, or in an unusual position. This occurs even when the baby is not crying.

* Your baby's due date is the date you were told your baby was going to be born, not when they were actually born.

- **Face –** One side of your baby's face droops
- **Foot –** Your baby's foot (or feet) turns inwards, turn outwards, or curl up. Your baby's toes may also curl up or their big toes point upwards a lot of the time.
- **Hand –** Your baby has their hand fisted, clenched, or in a claw like position. Your baby's thumb may be held within the fist. Your baby struggles to open their hands and stretch out their fingers.
- **Head –** Your baby always look or turns their head to face the same direction or always has their head tilted in the same direction. They struggle to turn their head to face the other direction. Your baby may also struggle to lift their head on their own and require head support when you hold them.
- **Leg –** Your baby's legs are positioned in an odd, or awkward way. Their legs may appear to be twisting inwards, or they may be crossing over each other.



Babies often enter into this posture before they are 6 months past their due date*. When an baby is laying on their back and faces in one direction (as shown by the arrows), the limbs they are not facing bend. Sometimes babies can get stuck in this position. You can help your baby out of this position by turning their head to face the ceiling. If you notice your baby is still getting into this posture around 6 months after their due date you should speak to your health visitor or GP.

Stiffness and floppiness

As babies grow and develop strength they usually begin to mould to their parents when being held or during feeding. When moving your babies' limbs you may feel some resistance, however this usually does not stop the parent gently moving their baby's limbs. Sometimes babies can feel floppy, such as when they are asleep. You should speak to your health visitor or GP if you notice one of the following persistently occurring:

- Your baby does not mould to you when you hold them, or you need to continue to give them extra support when you hold them 3-4 months after their due date*.
- Your baby's body, arms, or legs feel stiff or rigid making tasks such as changing your baby's nappy or clapping your baby's hands together difficult.
- Your baby's body, arms, or legs feel floppy even when they are awake, and you need to continue to support your baby's head when you hold them 3-4 months after their due date*.
- Your baby feels both stiff and floppy in different areas of their body

Reflexes and Reactions

Usually, babies are born with a set of reflexes and reactions. Over time some of these reflexes fade while their reactions develop. If you do notice any of the below you should contact your GP or Health Visitor.

- **Startle Reflex –** Every baby is born with a startle reflex that begins to fade away from 6 weeks past their due date* and disappears by the time your baby is 6 months past their due date*. All baby's usually startle when they hear a loud sound, see a sudden movement, or feel like they are falling. If you notice that your baby is often startling, and you cannot figure out why you should contact your Health Visitor or General Practitioner.
- **Reaction to sound –** Babies usually react to sounds around them, such as startling, changing their movement or becoming quiet. If your baby shows no reaction to sounds around them,

* Your baby's due date is the date you were told your baby was going to be born, not when they were actually born.

or sounds presented to one side of them, such as not turning to face the sound, you should contact your Health Visitor or General Practitioner.

- **Reaction to touch, textures, and temperatures –** Usually babies will respond to your touch. For example if you stroke a baby's cheek they will usually turn their head in response. If you notice that your baby shows no, or reduced, reaction to touch on certain parts of their body (such as only in one arm) or your baby dislikes being touched by certain touches, textures, or temperatures you should contact your Health Visitor or General Practitioner.

- **Response to your voice –** As babies grow they usually begin to respond to the sound of their parents voice by looking at their parents, smiling, going quiet, and/or or cooing. If when you speak to your baby your baby has not started to respond to you in some way by 8 weeks after their due date*, you should contact your Health Visitor or General Practitioner.

Skills development

As babies grow, they usually begin to learn new skills such as sitting, crawling, walking, grasping, and talking. Babies learn these skills in their own time, so comparing your baby's development to another baby's development may not be helpful. If you do notice any of the below you should contact your GP or Health Visitor.

- Your baby begins to learn new skills but never fully learns them. When babies begin to learn a new skill, they usually take their time to perfect it and can begin doing things such as only rolling in one direction or when they crawl your baby drags one of their limbs behind them as they practice. Babies usually grow out of this as their skills develop. If your baby does not seem to continue to progress in their skills, such as not trying to roll in the other direction or continues to drag a limb behind them you should contact your GP or Health Visitor.

Baby grasping or holding a toy in one hand

6 months after your baby's due date*

- Your baby has not begun to recognize their own name, show emotions like excitement, or started babbling
- Yor baby has not began to show any signs of learning how to grasp toys in one or both hands without you placing toys in their hand.

Baby sitting on their own

9 months after your baby's due date*

- Your baby has not begun to sit without you supporting them

Baby crawling

14 months after your baby's due date*

- Your baby has not begun to show any signs of learning to move, such as crawling or walking,
- Your baby has not said their first words (such as Mama or Dada) and does not seem to understand the meaning of any words you say to them, such as drink

There are more skills that your baby will develop that you may want to look out for and record. These are located towards the back of your Red Book. You can find out more about the skills your baby will develop and how you can support your baby's learning at https://www.bbc.co.uk/tiny-happy-people/science-and-facts

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Speaking and smiling

As babies grow they usually learn how to communicate with their parents though smiling and making non-crying noises. They begin to learn these skills from 8 weeks after their due date*. If you notice that your baby has not begun learning any of the following things, 8 weeks after their due date*, you should speak to your Health Visitor or GP.

- **Smiling –** When you smile at your baby, your baby does not smile back at you.
- **Making sounds** - Your baby does not try to make noises other than crying, such as cooing and babbling.
- **Response to your voice –**When you speak to your baby, your baby does not respond to you with a smile or by making a noise that is not crying.

Temperament and sleep

Every baby is different, some babies cry a lot while others cry a little. Some will cry when they are hungry, wet/dirty, or when they are unwell, while others will cry for no particular reason. At 2 months past their due date* it can be normal for a baby to cry up to 6 hours a day, this is usually not all at once. Please remember that crying is normal, that you are not a bad parent, you and your baby are still getting to know each other and over time you will both begin to learn how to communicate with each other. Below are some simple calming techniques from https://iconcope.org that you can try with your baby. Some may work for your baby and some may not, and some may work one day but not the next, just remember that it can take time for your baby to settle.

Safety Point

It is okay to walk away from your crying baby for a couple of minutes if you feel you need to calm down as long as your baby is in a safe space.

Never ever shake or hurt your baby.

- Try talking calmly to your baby. Stroke them gently. Try placing your baby face down on your lap or hold them against you and try stroking their back rhythmically.
- Hum or sing to your baby. Let them hear a repeating, constant and soothing sound. Classical music including piano and guitar can sometimes soothe some babies.
- Hold them close, such as skin to skin
- Take your baby for a walk or gently rock them in their pram or in a sling.
- Try giving them a warm bath

If you notice your baby is doing one of the below things, or you are struggling to cope with your baby's crying you should contact your health visitor or GP. You can also find further support at https://iconcope.org. Never ever shake or hurt your baby.

- Your baby seems distressed most of the time and is very difficult to calm or comfort. This occurs in different situations such as at home and at the GPs office. At 2 months past their due date* it can be normal for a baby to cry up to 6 hours a day, this is usually not all at once. If they are crying more than this or it is taking more than 30 minutes to calm your baby you should speak to your Health Visitor or GP.

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- Your baby may seem too quiet, may not cry often, or may attempt to cry but not have the strength, or be very passive.

- Usually babies get upset during dressing, tummy time and bathing, or in certain environments, such as noisy or bright environments, or when there are lots of people. If you notice that your baby is consistently or suddenly starts to become very distressed in a specific situations and is very difficult to settle afterwards you may want to speak to your Health Visitor or GP. Furthermore, your baby may only settle if they are laid down in a particular position, such as on their side.

- **Sleep** –Babies can wake up for many reasons and some may try to fight to stay awake, this is usual behavior in babies. Babies also wake up every few hours, needing feeding, attention, and changing. Around 6 months past their due date* babies usually begin to self-sooth and begin to sleep for longer periods of time. If you notice that your baby consistently has difficulties settling to sleep, he or she has difficulty staying asleep once they have fallen asleep, he or she appears sleepy much of the time, or is not waking up to feed, you may want to speak to your Health Visitor or GP.

A baby laying down on their tummy for tummy time. Tummy time is where your baby spends time on their tummy. This is something you can do with your baby for a couple of minutes each day. Tummy time helps babies to strengthen their neck, shoulder, and trunk muscles. Over time this allows them to lift their head up off of the floor and prop themselves up using their arms. How often your baby has tummy time will effect how quickly they develop these skills.

Usually 1 month past the baby's due date babies are able to turn their head during tummy time and may have begun to lift their head off of the ground for a couple of seconds. From 3 months past their due date babies are usually beginning to use their arms to help them lift their head during tummy time.

  - For advice on how you can help your baby to sleep and how to help your baby get into a sleeping routine visit https://www.nhs.uk/conditions/baby/caring-for-a-newborn/helping-your-baby-to-sleep/ or speak to your Health Visitor or GP.

Eye Gaze

As babies grow they usually begin to be able to focus on the world around them, watching the people around them and the toys presented to them. As they get older they usually become able to watch for longer and longer.

- Babies usually begin to fix their eyes on a toy and follow the toy as it moves 4 weeks after their due date*. They may only fix and follow for a couple of seconds as babies can lose interest quickly in the toy or can become overwhelmed and look away. If you notice that your baby struggles to fix their eyes on a toy and follow the toy as you move it you should contact your Health Visitor or GP.
- Babies usually begin to focus on faces in front of them from around 8 weeks after their due date*. They may not look for long before they look away. If your baby does not purposefully look at your face or others faces who care for them, even for just a couple of seconds you should contact your Health Visitor or GP.
- Babies usually begin to start making direct eye contact from 6 weeks after their due date*. If your baby is unable to maintain purposeful eye contact, even for just a couple of seconds you should contact your Health Visitor or GP.
- Usually, babies eye movements look purposeful. If you notice that your baby's eyes look vacant, or their eyes wander or roll in different or the same directions, or that their eye movements do not look purposeful you should contact your Health Visitor or GP.

6

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Feeding difficulties

Feeding, both bottle and breast feeding, is a skill that can take time to get the hang of for both you and your baby, so try not to worry if it takes you longer than you expected for both of you to get the hang of it. It can take weeks for babies to learn how to latch on efficiently. For advice on how to support your baby learning to feed visit https://www.nhs.uk/conditions/baby/breastfeeding-and-bottle-feeding/

Sometimes people choose not to breastfeed or are unable to breastfeed, and that is okay. If you do breast feed it may hurt to begin with but this will go with practice. When breastfeeding it can take up to 5 days for your milk to come in. For further advice around breastfeeding and answers to common breastfeeding concerns visit https://www.laleche.org.uk/

Below are some things that can occur in healthy babies and may just need time or a change in the way you and your baby do them. If you continue to notice them over time and they continue despite making changes you should contact your health visitor, GP, the National Breastfeeding helpline on 0300 100 1212, or your local breastfeeding support service. You can find your local service at https://www.nhs.uk/service-search/other-services/Breastfeeding-support-services/LocationSearch/360.

- Your baby becomes tired during feeding.
- Your baby's ability to feed varies. For example, your baby may feed fine one day and then the next day your baby may struggle to latch on or may feed for a prolonged period of time.
- Your baby struggles to latch on, sucking, or swallowing.
- Your baby has difficulty weaning.
- Your baby coughs, gags or chokes when swallowing liquids and/or foods.
- Your baby suffers from reflux (brings food back up).
- Your baby is unable to eat or drink without being sick.
- Your baby looks uncomfortable during feeding.

Safety Point

If you ever notice your baby choking please call 999 or 112.

You may also want to try to give your baby first aid while you wait. Guidance on how to stop a baby from choking is available from the NHS at:

https://www.nhs.uk/conditions/baby/first-aid-and-safety/first-aid/how-to-stop-a-child-from-choking/

Video instructions are available from the Saint Johns Ambulance Service at:

https://www.sja.org.uk/get-advice/first-aid-advice/choking/baby-choking/

- Your baby develops a rash after feeding. If you notice this please speak to your GP or Health Visitor straight away.
- Your baby becomes breathless during feeding or their lips begin to turn blue. If you notice this please speak to your GP or Health Visitor straight away.

Growth

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As babies grow and get older, they outgrow their clothes. Different babies grow at different speeds and it can be normal for a baby to be in a larger or smaller size than their age. Usually babies lose weight after birth but then regain it within 2 weeks before continuing to grow at a steady rate.

Your baby's growth should be tracked on the weight centile chart within your red book by your Health Visitor or GP. The weight centile chart shows how babies born at different weights usually gain weight over time. Usually as babies grow their weight follows one of the lines (also known as a centile) on the weight centile chart.

If your baby is ill it is likely that their weight gain will slow down until they are better. If you have one of the following concerns you should speak to your GP or Health Visitor.

- Your baby has stayed in the same sized clothing for longer than you expected.
- Your baby's weight has gone down two or more centiles on the weight centile chart within your red book.
- Your baby is going through clothing sizes faster than you expect or has suddenly gone up through clothing sizes
- Your baby's weight has gone up two or more centiles on the weight centile chart within your red book.
- Your baby's head grows very suddenly

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