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Parent-Child Interaction and the Developmental Profile of Children following Neonatal Encephalopathy

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Supervised by

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Declaration

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Chelo Del Rosario

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Summary

Neonatal Encephalopathy (NE) is a clinical condition characterised by disturbed neurological functioning in newborn infants. It is estimated that NE occurs in 3 in 1000 births worldwide (Kurinczuk et al., 2010) and is linked with unfavourable outcomes including developmental delay (Marlow et al., 2021; Robertson et al., 1989). The grade of NE (graded from mild to severe) is considered a reliable predictor for the development of those born with NE. However, inconsistencies in the literature have been observed and research has established that even those diagnosed with mild NE are at-risk of developmental delay (Conway et al., 2018).

The first chapter of this thesis describes the clinical nature of NE and provides a summary of the existing literature on the development of children impacted by the condition. Standardised assessments of development are the most common tool used to assess the development of children born with NE and are typically used as end-point assessments for neonatal trials. While standardised assessments allow us to measure the current developmental abilities of children, the developmental profile of children in this clinical group remains relatively undefined and the underlying processes that influence child development are underexplored. Identifying developmental processes may help explain variability in outcomes and can advance our understanding of the development of children born with NE.

The observation of parent-child interactions is a well-established approach that has been used to study developmental processes that impact child development. Chapter 2 reviews the literature on parent-child interactions and discusses how the behaviours of children and their parents during interaction are found to have a meaningful influence on child development. Research has demonstrated that dyads that include children with clinical conditions might be susceptible to characteristically different parent-child

interactions when compared to typically developing children and their parents (Festante et al., 2019; Wan et al., 2013). However, parent-child interactions have yet to be investigated in the context of those born with NE.

The present research sought to examine characteristics of dyadic interaction such as joint attention and the speech used by parents and children and how they are associated with children's cognitive, language and motor abilities. The parent-child interactions of mother-child dyads where children were born with NE were compared to dyads with children born without complications in order to better characterise the developmental profile of children born with NE.

Chapter 3 details the methods used in the empirical studies conducted in this thesis. A group of preschool-aged children born with NE and a group of children born without complications and their parents were invited to the Infant and Child Research Lab at Trinity College Dublin. Naturalistic interactions of the parent-child dyads at play were recorded and the children's developmental abilities were assessed using a standardised assessment of development.

Chapter 4 aimed to characterise the developmental profile of the group of children born with NE. The developmental assessment scores of children born with NE were compared with those of the children born without complications. Developmental risk factors associated with NE such as neonatal risk, autism traits, executive functioning and sleep difficulties were measured in those born with NE and the associations with developmental outcomes scores were examined. Overall, children born with NE performed significantly worse on assessments of language abilities and processing speed. A subset of children born with NE were identified to exhibit autism traits and EF difficulties. Children who exhibited these behavioural difficulties were also likely to receive below average scores on developmental assessments.

Chapter 5 consists of three studies investigating parent-child interactions and the development of children in clinical groups. Study one is a preliminary investigation of parent-child interactions and developmental outcomes in a closely related clinical group, children with an elevated likelihood of autism. This exploratory study was conducted as a precursor to investigating the parent-child interactions of dyads following NE. It examined the association between global characteristics of parent-child interactions and children's developmental outcomes between children with an elevated likelihood of autism and typical likelihood of developing autism. Study two examined the characteristics of joint attention episodes of mother-child dyads with children born with NE compared to children born without complications. Further, associations between characteristics of joint attention and the developmental abilities of children born with NE were explored. No differences in joint attention episodes were observed between the two groups. However, significant associations between joint attention and the developmental abilities of children born with NE were established. Mothers of children with poorer processing speed abilities used more contingent bids to successfully initiate joint attention. However, children with poor processing speed abilities terminated joint attention episodes more frequently.

The language abilities of the cohort of children born with NE in this study were significantly poorer compared to children born without complications. Therefore, Study three used language sample analysis to examine in detail characteristics of parent and child speech. Transcripts of parent-child interactions for both groups were analysed to measure and compare quantitative and qualitative features of the parents' child-directed speech and the speech produced by the children. In addition, associations between speech and other domains of developmental functioning of children born with NE were investigated. Subtle differences in parent and child speech were observed. Children born

with NE used more nouns in their speech and their mothers used fewer words per turn. A number of positive associations between child speech and children's developmental abilities were observed in children born with NE when they were age 4 years and older.

Chapter 6 discusses the practical and theoretical implications related to the findings of this research. The strengths and limitations of this study are outlined and suggestions for future research are provided.

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List of Abbreviations

ABC: Autism Behaviour Checklist

ADHD: Attention Deficit Hyperactivity Disorder

ADOS-G: Autism Diagnostic Observation Schedule-Generic

AQ: Autism Quotient

Bayley: Bayley Scales of Infant and Toddler Development

BGT: Basal Ganglia and Thalamus

BRIEF-P: Behaviour Rating Inventory of Executive Function, Preschool Version

CDS: Child-Directed Speech

CHAT: Codes for Human Analysis of Transcripts

CLAN: Computerised Language Analysis

CP: Cerebral Palsy

EF: Executive Function

EL: Elevated Likelihood

FRI: Fluid Reasoning Index

GMFCS: Gross Motor Function Classification System

Griffiths: Griffiths Mental Development Scales

HIE: Hypoxic Ischemic Encephalopathy

JA: Joint Attention

M-ABC: Movement Assessment Battery for Children

MACI: Manchester Assessment of Caregiver and Infant Interaction

M-CHAT-R: The Modified Checklist for Autism in Toddlers, Revised

MLT: Mean Length of Turn

MLU: Mean Length of Utterance

MLUm: Mean Length of Utterance using Morphemes

MRI: Magnetic Resonance Imaging

MSEL: Mullen Scales of Early Learning

NICU: Neonatal Intensive Care Unit

NE: Neonatal Encephalopathy

PLIC: Posterior Limb of the Internal Capsule

PSI: Processing Speed Index

SES: Social Economic Status

TH: Therapeutic Hypothermia

TL: Typical Likelihood

TTR: Type-Token Ratio

VCI: Verbal Comprehension Index

VSI: Visual Spatial Index

VOCD: Vocabulary Diversity

WMI: Working Memory Index

WPPSI: Wechsler Preschool and Primary Scale of Intelligence

List of Publications and Presentations

Publications

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Presentations

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Chapter One

The Developmental Profile of Children Born with Neonatal Encephalopathy

Defining Neonatal Encephalopathy

Encephalopathy is a general term that describes a disorder of the brain that alters its structure or function. Neonatal Encephalopathy (NE) is a clinical syndrome that results in neurological damage or dysfunction during the neonatal period. It may occur in term born infants from the 36th week of gestation through to birth following a normal pregnancy (The American College of Obstetricians and Gynaecologists; ACOG, 2014). The clinical presentation of an infant diagnosed with NE includes an altered state of consciousness, typically accompanied with difficulty initiating or maintaining respiration, depressed or abnormal tone and reflexes, seizures and difficulty feeding (Volpe, 2012).

NE is a condition with multifactorial aetiology therefore a number of terms are used interchangeably with NE such as neonatal brain injury, Hypoxic Ischemic Encephalopathy (HIE) and perinatal asphyxia. The term NE is most preferred as it is an overarching term that describes central nervous system dysfunction in infants from all causes (ACOG, 2014; Molloy & Beaer, 2018). The most commonly reported cause of NE is HIE, with 50 to 80% of NE cases being classified as HIE (Volpe, 2012). HIE typically describes encephalopathy that occurs due to hypoxia, a deficiency in the amount of oxygen reaching cell tissues, and consequently ischemia, a shortage of blood flow to the brain and other organs. Alternative or co-occurring causes of NE may include metabolic disorders, infection, stroke in the infant, intracranial haemorrhage and perinatal trauma (Leviton, 2013).

The causes of NE are still poorly understood but multiple risk factors have been linked to the occurrence of NE including placental pathology (Mir et al., 2015; Vik et al., 2018), maternal haemorrhage, nuchal cord and other cord complications (MacLennan,

1999), uterine rupture (Badawi et al., 1998), fetal bradycardia, maternal fever (Impey et al., 2001; Parker et al., 2018) and prolonged labour and birth trauma (Eden et al., 2018; Lijestom et al., 2018; Liu et al., 2018).

Epidemiology of NE

The neonatal period is an incredibly sensitive stage of life for the infant with 40% of deaths in children under the age of 5 years occurring during this time (Setiawan, 2013). NE is one of the biggest contributors to this statistic accounting for 29% of these deaths. Thus, NE causes an estimated 900,000 deaths per year globally (Lawn et al., 2007) and occurs in approximately three in 1000 births that take place globally (Kurinczuk et al., 2010). This places it amongst the top ten diseases with the highest lifelong global burden (Global Burden of Diseases Study, 2013).

Diagnosing NE

Due to our poor understanding of the aetiology of NE, a definitive test that accurately diagnoses the condition or predicts infant outcomes has yet to be developed. However, a number of clinical markers have been established to help diagnose the condition in infants. According to the American College of Obstetricians and Gynaecologists (2014), one or more of the following markers should be identified to consider a diagnosis of NE, including: an Apgar score of less than 5 at 5 minutes and at 10 minutes, a fetal umbilical cord blood pH below 7.0, a requirement for assisted ventilation, neuroimaging evidence of brain injury, or the presence of multisystem organ failure. The Apgar scale is typically used to assess the health of a newborn infant, based on five criteria including Appearance, Pulse, Grimace, Activity and Respiration. Scores below 3 are regarded as critically low, 4 to 6 are fairly low and scores between 7 and 10 are regarded as normal (Apgar, 1953).

If one or more clinical symptoms of neurological dysfunction are present, the Sarnat Grading Scale may be used to confirm the diagnosis of NE and to classify the grade of the insult into one of three categories of severity. The classification of NE is established by the observation of six clinical characteristics including: alertness, muscle tone, seizures, pupils, respiration and duration. An infant classified with mild grade I NE may appear hyperalert, to the point where feeding problems may arise, the infant will have normal or increased muscle tone, dilated yet reactive pupils, normal respiration, with no seizures reported. The duration of this appearance would typically be less than 24 hours. Those classified with moderate grade II NE will appear lethargic and hypotonic, seizures may arise frequently, pupils are small and reactive, maintenance of respiration would be periodic, with the state lasting between 2 to 14 days. In the case of a severely affected individual, the infant will be in a state of coma, with flaccid muscle tone, unequal and often unreactive pupil dilation, and unable to independently sustain respiration. The observed state may often last for a few weeks (Sarnat & Sarnat, 1976).

Treatment for NE

The primary treatment method available for NE is Therapeutic Hypothermia (TH). TH is the standard treatment offered to infants diagnosed with moderate to severe NE. It is an important therapy that has been found to provide neuroprotection for those at-risk of brain damage. TH for those diagnosed with NE involves lowering the infant body temperature from a normal temperature of 37 degrees Celsius to 33 to 34 degrees Celsius. The infant may undergo whole body cooling which involves wrapping the infant in a blanket filled with cooled liquid. Alternatively, selective head cooling may be administered where only the head is cooled using a cap filled with cold water. TH is most effective when administered within 6 hours of birth (Gunn & Bennet, 2015) for

approximately 72 hours (Jacobs et al., 2010). Because of the small window for treatment time, it is imperative that NE is detected as soon as possible.

A number of theories have been proposed that explain the neuroprotective mechanism of TH. Following NE, an inflammatory response is induced in the body causing an exhaustion of cell energy stores which is associated with brain damage (Ziemka-Nalecz et al., 2017). TH reduces the inflammatory response and slows down the process of cell death (Gonzalez-Ibarra et al., 2011). TH has been found to modify cells programmed for apoptosis. Apoptosis is a process that the body typically uses to eliminate unneeded superfluous cells and causes cell death. However, NE may cause inappropriate apoptosis resulting in excessive cell death. Following TH, there is a decrease in the number of apoptotic cells, thus reducing the rate of brain damage (Edwards, 1995). TH also protects neurons by reducing cerebral metabolic rate and slows down the release of excitatory amino acids such as dopamine and glutamate. This lowers the production of neurotoxins and free radicals in the brain that otherwise cause cell damage (Globus, 1995).

In addition to being easy and inexpensive to administer, several clinical studies and randomised controlled trials have found TH to be incredibly effective. TH reduces detrimental outcomes such as disability or death in almost half of moderate to severely affected infants who receive the intervention (Azzopardi et al., 2014; Jacobs et al., 2010). However, this intervention has not been found to be as effective in improving outcomes in populations who experience mild NE (Azzopardi et al., 2014). Thus, it is not standard practice to administer TH to those diagnosed with mild NE. TH is the only treatment available for NE and there are currently no standard treatment options available to those who fall within the mild category.

Short-Term Clinical Outcomes of NE

Immediate clinical outcomes caused by NE are variable and depend on factors such as the cause of the NE, time of onset, duration, magnitude and repetition of insult. NE is classified into three grades of severity ranging from mild to severe (Sarnat & Sarnat, 1976). Depending on the severity of the encephalopathy endured, outcomes may range from typical development, to moderate disability to death. NE has a mortality rate of approximately 15% and a further 25% will exhibit neurological disabilities (Miller et al., 2004). NE is associated with the onset of conditions such as cerebral palsy (De Vries et al., 2010), other motor disorders (Adhikari & Rao, 2017; Mercuri & Barnett, 2003) and seizure disorders (Miller et al., 2004). Sensory impairments have also been observed in infants born with NE (Pin et al., 2009; Volpe, 2018). Major sensory disorders that occur as a result of NE include visual disturbances (Liu et al., 2017) and hearing impairment (Fitzgerald et al., 2018). Abnormal sensory functioning is found to negatively impact cognitive, motor and communication development (Lickliter, 2000). Measures of sensory processing serve as predictive indicators of neuromotor and cognitive development in children born with NE, as delayed sensory processing is associated with lower IQ scores (Kontio et al., 2013). Children born with NE spend the early days of their life in Neonatal Intensive Care Units (NICU). NICUs are bright and noisy environments resulting in excessive sensory stimulation for newborn infants. Excessive sensory exposure in NICU environments is found to have adverse effects on children's future language and motor outcomes (Pineda et al., 2014).

Overall, those who experience difficulties as a result of NE may encounter constraints in their development due to sensory impairments and the reduced ability to explore the environment (Von Hofsten, 2009). Disabilities that occur as a result of NE

may restrict the range of experiences that promote early learning in infants and children (Lewis, 2002).

Neurodevelopmental Outcomes of Children Born with NE

Several studies have investigated the developmental outcomes of children born with NE (Finder et al., 2019; Murray et al., 2016; Pappas et al., 2015; Robertson & Finer, 1985; Van Handel et al., 2007). Such studies highlight the importance of monitoring long term outcomes in this clinical cohort regardless of the severity of NE endured. NE is associated with a number of outcomes that impact neurodevelopment and consequently developmental outcomes including intellectual disability (Dixon et al., 2002), memory impairment (Van Handel et al., 2012), language delay (Shapiro et al., 2017; Adhikari & Rao., 2017; Martinez et al., 2014), attention problems (Spencer et al., 2021; Tonks et al., 2019) and behaviour disorders (Lindström et al., 2006; Robertson et al., 1993). Those born with NE have also been found to have an increased risk of developing neurodevelopmental disorders such as autism (Badawi et al., 2006) and attention deficit hyperactivity disorder (ADHD; Smith et al., 2016).

Standardised assessments are designed to measure aspects of child development such as cognitive, language and motor functioning. These assessments are typically made up of tasks designed for children and follow specific guidelines for administration and scoring to ensure consistency and reliability (Del Rosario et al., 2020). Standardised assessments establish norm scores based on data collected from a representative sample of children. This allows us to determine how well an individual child performs on the assessment relative to children of the same age. These assessments are beneficial for those impacted by NE as they identify developmental delays, help monitor children's progress and inform intervention planning. However, limitations on the reliability and standardisation of such assessments have been reported. Standardised assessments have

been found to overestimate developmental outcomes. This has been attributed to the recruitment of high-achieving normative samples and inaccurate administration and/or scoring (Anderson et al., 2010). These assessments are also considered to be blunt as they tend to assess each developmental domain globally. Therefore, subtle deficits in subdomains of development may be missed (Brito et al., 2019).

Despite these limitations, the majority of studies that examine the developmental outcomes of children born with NE measure cognitive, motor and language development using standardised assessments of development. Outcomes in these core domains and how they may be impacted in children born with NE will be discussed in the following section.

Cognitive Development of Children Born with NE

General cognitive functioning is a broad construct that refers to how individuals think, understand and interact with the world around them. Particular aspects of cognitive functioning include perception, learning, memory, understanding, awareness, reasoning, judgement, intuition and language (VandenBos, 2015). Proficiency in cognitive skills can help facilitate higher order thought processes such as problem solving, information processing and decision making.

Biomarkers of NE and their Association with Cognitive Development

Research has demonstrated the extent of neural injury to be a significant predictor of future cognitive outcomes in children born with NE (Shankaran et al., 2015; Skranes et al., 2017). Magnetic Resonance Imaging (MRI) is the gold standard technique used to detect patterns of neural injury caused by NE (Rutherford et al., 2010). Conventional MRI is commonly used as the technique demonstrates good diagnostic ability to identify brain damage by the end of the first week of life. Advancements in neuroimaging techniques have allowed the development of Diffusion Weighted Imaging and Magnetic

Resonance Spectroscopy. These methods can identify brain damage within the first few days of life; however, these methods are not widely available for use in all hospitals (Khong et al., 2004). In circumstances where MRI is not available for use, such as those receiving treatment in a community hospital or in developing countries, ultrasound can also be used to identify brain injury in infants born with NE (Annick et al., 2020).

A number of biological markers associated with cognitive outcomes among children born with NE have been identified. The outcomes of children born with NE depend on lesion severity and the pattern of neural infarction. MRI studies have found higher levels of brain injury to be associated with lower scores on cognitive assessments (Shankaran et al., 2015). Mild to moderate injury typically involves damage to the watershed regions of the brain. The watershed region borders the major cerebral arteries. Their proximity to the brain blood supply makes these regions vulnerable to events such as ischemia; thus, watershed infarction is a common clinical outcome in those born with NE (Steinman et al., 2009). A watershed pattern of injury is generally considered milder compared to damage in the deeper subcortical regions of the brain. Whilst watershed brain injury is categorised as mild to moderate, evidence suggests that cognitive impairment can occur in children who sustained injury to the watershed region (Lee et al., 2022; Miller et al., 2005; Trivedi et al., 2017).

Severe NE commonly results in damage to subcortical regions of the brain such as the Posterior Limb of the Internal Capsule (PLIC; Hayes et al., 2016) or the Basal Ganglia and Thalamus (BGT; Miller et al., 2005). These structures are made up of diverse neural formations located deep within the brain, influencing a multitude of brain functions. The PLIC is a white matter structure that facilitates communication between areas of the cerebral cortex and the brainstem. It coordinates cognitive, motor and sensory pathways in the brain (Emos et al., 2022). Research has demonstrated damage to the

PLIC is almost always linked to cognitive deficits in children born with NE (Hayes et al., 2016).

The BGT is a group of subcortical nuclei primarily responsible for motor control but its connection to the prefrontal cortex suggests that it may also influence cognitive processes such as working memory, rule-based learning and planning behaviour (Bostan et al., 2018). Damage to the BGT as a result of NE is associated with the worst outcomes including disability or death. Even those with mild damage to the BGT are found to exhibit poor outcomes (Hayes et al., 2016). BGT injury is associated with severely impaired cognitive outcomes in children born with NE when assessed at 30 months of age (Miller et al., 2006).

NE that occurs as a result of neonatal infection has been found to be associated with future cognitive outcomes in children. Those born with NE exhibiting signs of neonatal sepsis showed an increased likelihood of sustaining neural injury to the watershed region, thus, increasing the risk of adverse cognitive outcomes in children (Jenster et al., 2014). Other clinical outcomes caused by NE such as the presence of seizures at birth have been found to be predictive of future cognitive outcomes (Chang et al., 2020; Miller et al., 2004).

Although neural damage has been demonstrated to be a reliable predictor of long-term cognitive outcomes, research suggests that children born with NE remain at an increased risk of cognitive deficits even if they have a normal MRI scan indicating no neural injury. Up to 30% of children born with NE who do not sustain brain damage may still go on to develop a disability or exhibit poor developmental outcomes (Barnett et al., 2002; Hayes et al., 2016; Shankaran et al., 2015).

Cognitive Outcomes of Children Born with NE

NE has been associated with impaired cognitive outcomes and poor academic performance in childhood (Lindström et al., 2008; Schreglmann et al., 2020), but the cognitive profile of children born with NE is still not clearly defined. Studies of children born with NE generally conclude that future developmental outcomes can be determined by the grade of NE endured at birth (Barnett et al., 2002; Robertson et al., 1989; Van Handel et al., 2007). Children born with severe NE almost always suffer from significant cognitive delay and poor academic performance even with the absence of disability. Those born with a moderate grade of NE have mixed outcomes, they tend to receive scores within the normal range but score significantly lower than those born with mild NE.

The literature on mild NE is mixed, some find children with mild NE to receive below average scores on assessments of cognition (Conway et al., 2018; Van Kooij et al., 2010), others do not (Finder et al., 2019; Robertson et al., 1989). Despite receiving developmental outcomes scores within the normal range, recent research has identified those born with mild NE to have significantly lower cognitive scores when compared to a healthy control group (Finder et al., 2019; Murray et al., 2016).

Standardised assessments of development are commonly used to measure cognitive outcomes of children born with NE. The Bayley Scales of Infant and Toddler Development (Bayley; Bayley, 1993; 2006), and the Griffiths Mental Development Scales (Griffiths; Luiz et al., 2006) are most often used when assessing children aged 24-42 months. Each tool uses play-based activities to assess cognitive functioning in young children and cognition is measured broadly using these assessments. The Bayley scales include items that assess sensorimotor development, manipulation and exploration of objects, concept formation, memory and other aspects of cognitive processing. The

Griffiths scales include a Foundations of Learning Subscale. The scale evaluates aspects of cognition in children including memory, thought sequencing, attention, processing speed, and executive functioning (Stroud, 2016).

Studies that assess the cognitive functioning of children born with NE using the Bayley and Griffiths scales find that the presence of a disability can influence the cognitive functioning of children born with NE. Almost all children born with moderate NE who were diagnosed with a disability such as cerebral palsy were severely developmentally delayed. Children with moderate NE without disability performed in the average range or above, although the range of scores within the NE cohort varied widely (Van Handel et al., 2007). However, more recent research has observed 25-63% of children without disability to demonstrate impairments in general cognitive abilities. This indicates that a substantial proportion of children born with NE are at risk of cognitive impairments even if they do not have a disability (Schreglmann et al., 2020).

Some studies have found the Bayley and the Griffiths scales to be good predictors of impairment at school age. Children who received a low cognitive composite score on the Bayley or the Griffiths at 12 and 24 months of age were also likely to receive below average scores on developmental assessments at 5 to 6 years of age (Barnett et al., 2004; O'Connor et al., 2017; Pappas et al., 2015). However, findings are mixed as other studies reported cognitive deficits at 6 to 8 years were not predicted by developmental outcomes scores collected at 18 months of age (Azzopardi et al., 2014; Jary et al., 2019). Recent research suggests that school readiness can be predicted from Bayley assessment scores collected at multiple time points rather than a single assessment score (Neel et al., 2023).

When children reach school age, the Wechsler Intelligence Scales are commonly used to assess aspects of cognitive functioning. Different versions of the assessment are administered depending on the age of the individual. The Wechsler Preschool and

Primary Scale of Intelligence (WPPSI; Wechsler, 2013) measures cognition in children aged 2.6 to 7.7 years of age. The WPPSI measures subdomains of intellectual functioning such as visual spatial skills, verbal skills, fluid reasoning, working memory and processing speed. An overall score is then calculated to determine the child's Full-Scale IQ.

The extent of neural damage sustained from NE has been found to have a significant impact on WPPSI scores. The cognitive skills of school aged children born with NE were assessed in children with a normal MRI and in those who sustained neural damage as a result of NE. The majority of children who received a normal MRI result performed in the average range on the WPPSI assessment. However, the majority of children who sustained significant neural damage had below average full-scale IQ scores (Shankaran et al., 2015).

The severity of NE sustained at birth has also been found to have a significant impact on WISC scores (Wechsler Intelligence Scale for Children; Weschler, 2014) in children when assessed at 9-10 years of age. It was found that 23% of children born with mild NE went on to receive an IQ below 85, whereas 66% of those born with moderate to severe NE had an IQ below 85 (Van Kooij et al., 2010).

The Wechsler scales measure a number of cognitive subdomains and studies have found children born with NE to receive below average scores on the short-term verbal memory and processing speed scales (O'Connor et al., 2017). However, only a small number of studies report scores on each subdomain of the Wechsler scales. The majority of studies only report the full-scale IQ score as an indicator of cognitive development. Future studies should continue to report scores by subdomain in order to better characterise the cognitive profile of children born with NE.

Examination of Specific Cognitive Skills in NE

Cognition is a broad construct that comprises a number of skills. Standardised assessment of development typically produces a cognitive composite score that provides an overview of an individual's global cognitive functioning. Composite scores are a single numerical value that represents a child's performance across the different cognitive domains. However, these scores do not describe performance on each cognitive domain.

A small number of studies have measured specific aspects of cognition in children born with NE such as memory, attention and processing speed (Annick et al., 2018; O'Connor et al., 2017; Tonks et al., 2019; Van Handel et al., 2012; Zorn et al., 2018). One study measured the memory and processing speed skills of 8-12 month old infants born with NE (Zorn et al., 2018). Their performance on attention shifting and episodic memory tasks were compared to a control group born without complications. Overall, no differences in speed of processing or memory between infants born with and without NE were observed.

However, when the same domains of cognition are assessed in later childhood, deficits become more apparent. Research has demonstrated verbal working memory, verbal and visuospatial long-term memory and learning to be negatively impacted in 9-10 year old children born with NE (Annick et al., 2018; Van Handel et al., 2012). Some cases have been associated with differences in neural volumes such as reduced hippocampus size (Annick et al., 2018), but memory problems have also been observed in those without disability and with a normal MRI (Van Handel et al., 2012). Deficits in attention skills and processing speed have also been observed in later childhood (O'Connor et al., 2017; Tonks et al., 2019). In a study of 6 to 8-year-old children born with NE, Tonks and colleagues (2019) reported attention difficulties, slower reaction

times and poorer visuo-spatial processing abilities when compared to a healthy control group, matched on age and gender.

These specific cognitive functions interact with other domains of development. For example, verbal working memory tasks involve language abilities and tasks testing reaction times require motor abilities. It is possible that deficits in other developmental domains such as language and motor abilities impact the application of cognitive skills in day-to-day life. Deficits in other domains of development mean that children may have a reduced capacity to practice and refine these associated cognitive skills which may explain why memory and processing speed abilities appear to be preserved in infancy (Zorn et al., 2018) but difficulties in these domains become more apparent in later life.

Cognition in Later Life

When children born with NE are assessed in early childhood, outcomes on cognitive assessments are variable, particularly in children diagnosed with moderate NE. The majority of studies have found children without disabilities to score within the normal, but lower, range on assessments of cognition (Pappas et al., 2015; Schreglmann et al., 2020). To date, only a handful of studies have followed-up children into adolescence. However, these studies found that as children born with NE get older, delays in domains of cognition become more apparent. Lindström and colleagues (2006) measured the cognitive abilities of teenagers aged 15-19 years of age who were born with NE. Seventy-one percent of the sample without disability exhibited cognitive dysfunction that impacted their daily living and academic achievement. Further research has established that by late childhood and adolescence, those born with moderate NE are more likely to be at least one grade level behind their peers at school (Lee & Glass, 2021). Studies should continue to follow-up children born with NE into later life in order to better define the trajectory of cognitive development of those in this clinical cohort.

Language Development of Children Born with NE

Research suggests that NE impacts language outcomes in the developing child, although findings are mixed. Standardised assessments of development are the most common tool used to assess language skills in children born with NE. Some studies identify high levels of impairment (Herrera et al., 2018; Mitra et al., 2018; Murray et al., 2009) while others report language abilities within the normal range (Finder et al., 2019; Marlow et al., 2005; Rollins et al., 2014; Rutherford et al., 1998), albeit children born with NE tend to score in the lower normal range. One consistent finding across studies is that the severity of NE correlates with increasingly adverse language outcomes, suggesting a dose-response relationship between the severity of neural damage and subsequent language abilities (Finder et al., 2019; Martinez-Biarge et al., 2012; Steinman et al., 2009; Trivedi et al., 2017).

Biomarkers of NE and their Association with Language Outcomes

Biomarkers identified using MRI techniques are found to be the best predictor of language outcomes in children born with NE (Barta et al., 2018). The grade of neural damage sustained at birth is typically correlated with language outcomes in later life. Studies have found increasingly extensive neural damage caused by NE to be associated with poorer language outcomes in later life (Chalak et al., 2018; Gorgen et al., 2014; Mitra et al., 2018; Rollins et al., 2014).

Damage to specific neural regions is associated with language deficits including the perisylvian area, watershed infarctions and the subcortical regions of the brain. The perisylvian area is a neural region responsible for language (Catani et al., 2005). The grey and white matter within this region have been found to be altered in those born with NE (Shapiro et al., 2017). Shapiro et al. (2017) investigated the relationship between alterations in brain structure caused by NE in infants aged 6 months and its association

with developmental outcomes in children when they turned 30 months of age. Decreased volume in perisylvian grey and white matter at 6 months was associated with lower language scores on the Bayley-III at 30 months but not with cognitive and motor outcomes. Similarly, Trivedi et al. (2017) found that a higher neural injury grade was significantly associated with poorer language outcomes at 18 to 24 months, as measured using the Bayley-III. The impact on language development tends to be most severe when damage occurs in the subcortical regions of the brain such as the BGT. The effect of neural damage sustained by NE on language development also persists into later life. Steinman et al. (2009) found watershed brain injury sustained at birth caused by NE correlated with verbal intelligence at 4 years of age. The more severe the brain injury at birth, the lower the verbal intelligence when the children were 4 years old.

One item on the Gross Motor Function Classification System (GMFCS; Palisano et al., 2008) is associated with the presence of communication impairments. The GMFCS was conducted on a group of 126 children at age 24 months of age with sustained injury to the brainstem or the BGT as a result of NE. Children were rated as having either age-appropriate expressive speech and language; a mild/moderate communication impairment, i.e., able to speak with the use of formal assistive communication methods; or severe communication impairment, i.e., non-verbal using assistive methods or unable to communicate at all. Using this system, 82% of the children were classified as having communication impairments, with 48% of the sample classified as non-verbal at 24 months (Martinez-Biarge et al., 2012).

Language Outcomes of Children Born with NE

Standardised assessments are commonly used to measure language development in children born with NE, in particular the Bayley Scales (Bayley, 2006), and the Griffiths Scales (Luiz et al., 2006). The two scales measure language according to two further

subdomains including expressive and receptive language. Expressive language assesses an individual's ability to use language in order to speak and communicate using words, phrases and sentences. It also assesses the ability to use grammar accurately. Receptive language is the ability to understand words and spoken language, it involves processing information being conveyed to an individual such as questions or instructions.

Varying rates of language delay are identified among those born with moderate to severe NE ranging from 1.4% (Rutherford et al., 1998) to 40% (Mitra et al., 2018). Some studies report language deficits and academic difficulties (Herrera et al., 2018; Murray et al., 2009; Robertson et al., 1985), whilst others report language assessment scores within the normal range (Marlow et al., 2005; Massaro et al., 2015; Miller et al., 2004; Twomey et al., 2010). A large proportion of studies that use standardised assessments tend to report composite language scores and do not differentiate between performance on subscales such as receptive and expressive language (Dixon et al., 2002; Herrera et al., 2018; Massaro et al., 2015; Mitra et al., 2018; Murray et al., 2009; Rollins et al., 2014). Thus, the trajectory of language development in children born with NE is still poorly defined.

The majority of studies investigating language outcomes of children born with NE fail to include a comparison sample of healthy children born without NE. Much of the medical literature uses the cut-off points determined by measures such as the Bayley Scales to indicate above or below average scoring. However, studies have found tools such as the Bayley Scales to inflate scores and underestimate developmental delay (Anderson et al., 2010; Green et al., 2023), including in those born with NE (Jary et al., 2013). A subset of studies has therefore directly compared scores of children born with NE to those of typically developing children born without complication. Most children born with NE with no concomitant or comorbid disability score in the normal range on

standardised assessments of language. For example, Marlow and colleagues (2005) found 77% of children born with NE to receive developmental assessment scores within the normal range. However, when compared to a typically developing comparative group, the NE children's scores are significantly lower, albeit still within the normal range (Finder et al., 2019; Marlow et al., 2005) suggesting that NE negatively impacts language development. Finder et al. (2019) compared the language scores of 24-month-old children who had either experienced no complications at birth, complications but no NE, mild NE, moderate NE or severe NE. There was a mean difference of 2.7 points between the control group and the mild NE group, and a difference of 14.5 between the control group and those born with severe NE. This suggests that as the severity of NE increases, mean composite scores decrease, suggesting a dosage-response effect.

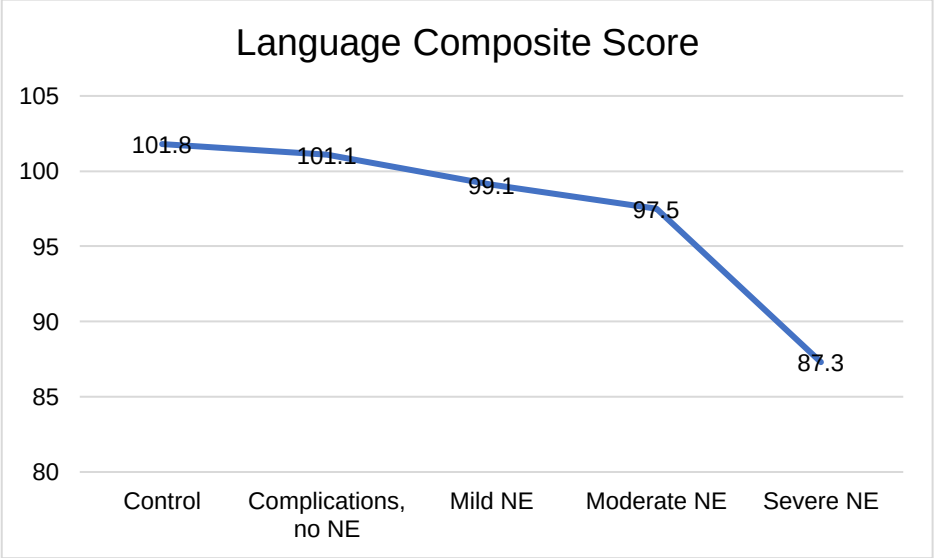

Figure 1. Mean language scores for children born with NE and control groups obtained using the Bayley-III assessment (Finder et al, 2019).

When the language abilities of children born with NE are assessed using both global and language specific measures, different profiles of language ability are observed. Only one study to date has measured the language abilities of children born with NE

using language specific tools. Chin and colleagues (2019) assessed the language abilities of 2-year-old children born with NE using both a global measure of development, the Mullen Scales of Early Learning (MSEL; Mullen, 1995) and a language-specific tool, the MacArthur-Bates Communicative Development Inventories (MB-CDI; Fenson & Marchman, 2007). The MSEL measures general language skills such as receptive and expressive language alongside non-verbal problem solving, termed as visual reception. The MC-CDI reports on specific language skills such as volume of word production and average length of the longest three sentences produced.

Chin et al. (2019) reported mean receptive and expressive language scores within the normal range using the MSEL. However, when children born with NE were assessed using a language specific scale, an uneven profile of impaired expressive language (characterised by poorer vocabulary and shorter utterances as measured by the MC-CDI) and visual-reception was established, alongside relatively intact receptive language. Whilst differences in the language profile of children born with NE were identified using the MC-CDI the implications of using parent report questionnaires must also be considered. Parents are keen observers of their child's development. However, certain biases may impact the accuracy and reliability of parent reports particularly those from parents of children in clinical groups. Parents have been found to emphasise challenging behaviours (Zapolski & Smith, 2013). Alternatively, parents may overestimate the abilities of their children as they may be reluctant to acknowledge that their child is experiencing any difficulties (Ozonoff et al., 2011).

This review of the literature indicates that there is very little detailed information on the acquisition and development of language in children born with NE. Standardised assessments of language ability are helpful in observing the relationship between specific biomarkers and general language outcomes in later life (Shapiro et al., 2017; Steinman et

al., 2009; Trivedi et al., 2017). However, they do not help characterise the trajectory of language development in children born with NE nor do they identify the impact of NE on specific language skills. Studies that continue to conduct assessments of global development should employ appropriate control groups. More detailed analysis of language outcomes must be reported, including performance on expressive and receptive language domains. In order to better delineate the language outcomes of children born with NE, language-specific assessments and observational studies of language abilities such as spontaneous speech sampling and fine-grained analysis should be used to provide an in-depth insight into the functional and communicative language skills among those born with NE.

Motor Development of Children Born with NE

Motor development describes the physical growth and development of a child. It is associated with the strengthening of a child's bones and muscles that in turn facilitate the ability to move, touch, and manipulate their surroundings (Goodway et al., 2019). There are two categories of motor skills: gross motor skills and fine motor skills. Fine motor skills involve intricate actions initiated with the hands and fingers that allow the manipulation of small objects. Gross motor skills involve the coordination of the child's whole body and includes skills such as weight distribution, and the ability to sit, crawl, walk, run and jump (Albers et al., 2007).

Biomarkers of NE and their Association with Motor Outcomes

MRI techniques are used to reliably predict motor outcomes of children born with NE with up to 82% sensitivity (Mitra et al., 2018). Neural damage is significantly associated with the development of movement disabilities and poor motor outcomes (Goregen et al., 2014; Polat et al., 2013; Van Kooij et al., 2010). Specific neural biomarkers associated with NE include damage to the PLIC, the BGT, and grey and white

matter regions (Ferrari et al., 2011). Damage to these regions is also linked to the onset of movement disabilities.

Rutherford and colleagues (1996) found damage to the PLIC was almost always associated with motor deficits in children born with NE. The primary motor cortex sends signals via the PLIC; therefore, damage to this region often results in the onset of movement disabilities in the developing child (Noback et al., 2005). The severity of motor delay associated with damage to the PLIC is related to the extent of additional damage to the BGT region. While the function of the BGT is heterogeneous, one of its core functions is to facilitate voluntary motor movements (Lanciego et al., 2012). Miller et al. (2005) found 25% of children born with NE had endured injury to the BGT. Those who sustained damage to the BGT were likely to experience motor delay identified using the Bayley Scales at 30 months of age.

The function of white matter is associated with motor skill learning (Sampaio-Baptista et al., 2013) and irregular white matter has been associated with poor motor skills in the developing child (Mostofsky et al., 2007). Weeke and colleagues (2018) found damage to deep grey matter and white matter caused by NE to be associated with below average motor composite scores on the Bayley-III at 24 months, indicating developmental delay in this domain.

Movement Disabilities in Children Born with NE

The onset of motor specific disabilities is a commonly reported outcome in those born with moderate to severe NE (Jary et al., 2019; Lindström et al., 2006; Perez et al., 2013; Van Schie et al., 2015). Ferrari et al. (2011) assessed general movement in children born with moderate to severe NE at 24 months and found a number of movement disabilities to be associated with site-specific neural damage such as dystonic cerebral palsy, spastic quadriplegia, spastic diplegia and spastic hemiplegia. Cerebral Palsy (CP) is

one of the most common disabilities to occur as a result of severe NE. Approximately one in eight children born with moderate or severe NE will go on to develop CP (Badawi et al., 2005). CP is a movement disorder, characterised by poor coordination, stiff or weak muscles, and tremors. Additional symptoms may include difficulties with speech, sensation, vision, hearing and swallowing (American Psychiatric Association [APA], 2013). One of the leading causes of CP in those diagnosed with NE is significant neural damage, particularly to the BGT region.

The GMFCS (Palisano et al., 2008) is a clinical classification system used to evaluate the gross motor skills of children diagnosed with CP. It is a common assessment conducted in those who have developed CP as a result of NE (Ambalavan et al., 2006; Charon et al., 2016; Herrera et al., 2018; Rollins et al., 2014; Shellhaas et al., 2015; Skranes et al., 2017; Van Schie et al., 2015). Gross motor abilities are classified on a scale of 1 to 5 through the assessment of movements such as climbing stairs, walking, jumping, speed, balance and coordination. A GMFCS classification of level 1 describes motor deficits that have a low impact on daily living, whereas levels 3 to 5 indicate significant impact on daily functioning. A correlation between the severity of NE endured and GMFCS level has been observed, where increasingly severe NE is associated with higher scores on the GMFCS (Ambalavan et al., 2006; Skranes et al., 2017).

Motor Outcomes of Children Born with NE without a Movement Disability

Delay in motor development has been observed in children born with NE even in those without a diagnosis of a movement disability. The Movement-ABC (Movement Assessment Battery for Children; Henderson et al., 2007) is a motor specific behavioural assessment used to identify motor impairment in children from 3 to 16 years of age. The tool assesses fine motor and gross motor skills including manual dexterity, balance, aiming and catching. Several studies have used the Movement-ABC as an endpoint

assessment to identify the effect of NE on motor development (Barnett et al., 2004; Mercuri & Barnett, 2003; Van Kooij et al., 2010; Van Schie et al., 2015). Approximately 24% of children born with NE score below the 15th percentile on the Movement-ABC assessment, indicating borderline motor impairment. A further 25% score at or below the 5th percentile indicating abnormal motor functioning (Barnett et al., 2004; Jary et al., 2019; Van Kooij et al., 2010; Van Schie et al., 2015). When compared to a control group born without NE, those born with NE were found to exhibit significantly poorer motor skills when assessed using the Movement-ABC (Van Kooij et al., 2008).

Global assessments of development are also used to measure the impact of NE on motor outcomes in children aged 3 years and younger. Global assessments include the Bayley Scales (Bayley, 2006), and the Griffiths Scales (Luiz et al., 2006). Both tools include an assessment of gross motor and fine motor abilities and are used to identify motor delay in children born with NE (Barnett et al, 2004; Chalak et al., 2018; Finder et al, 2019; Herrera et al., 2018; Murray et al., 2009; Rollins et al., 2014; Shellhaas et al., 2015; Skranes et al., 2017). Whilst some studies have reported motor delay using global assessments of development (Murray et al., 2009; Van Schie et al., 2015), other studies have reported mean scores in the normal range (Barnett et al., 2004; Chalak et al., 2018; Finder et al., 2019; Murray et al., 2009; Shellhaas et al., 2015). In one study, no significant difference in motor composite scores was found between children born with NE and a control group born without NE when assessed using the Bayley-III. A mean difference of only 1.4 points was observed between groups, with the NE children scoring slightly lower than the control group (Finder et al., 2019). None of the studies report performance on sub-scale domains such as fine motor and gross motor scores. Reporting performance on sub-domains is necessary in order to establish whether fine motor and gross motor skills are differentially impacted by NE.

Global assessments of development are typically used in younger cohorts aged 1 to 2 years. Motor specific assessments such as the Movement-ABC can only be used from the age of 3 years. The predictive ability of global assessments has been investigated by observing the relationship between motor composite scores obtained by global assessments and scores on motor specific assessments obtained in later life (Barnett et al., 2004; Jary et al., 2019; Mercuri & Barnett, 2003; Van Schie et al., 2015). Comparative studies found motor composite scores obtained by the Bayley-III to have poor predictive abilities of motor outcomes (Hayes et al., 2016; Jary et al., 2019). The Movement-ABC identifies more cases of motor delay compared to global assessments of development (Barnett et al., 2004; Jary et al., 2019; Van Schie et al., 2015). Jary and colleagues (2019) assessed motor development in children born with NE using the Bayley-III when they were 18 months of age. Motor skills were assessed again when children were 6 to 8 years of age using the Movement-ABC. At 6 to 8 years of age, one-third of children received scores indicating motor impairment on the Movement-ABC assessment that had not been detected at age 18 months using the Bayley-III. The disparity between motor deficits observed using global assessments and motor specific assessment might be explained in one of two ways: the global assessments are not sensitive enough to identify motor delay in young children, or the issues with motor development did not exist at the time of the first assessment but developed as the children got older. Further research is required to explain the trajectory of motor development in children born with NE.

The Relationship Between Motor and Language Development

The relationship between motor and language development in typically developing children has been extensively researched (Iverson, 2010; Valla et al., 2020). Motor milestones are an indicative developmental marker of motor development, but also for cognitive, language and social skills. Underlying components of motor skills are

found to facilitate the development of language and communication skills. For example, in infancy rhythmic arm movements such as banging objects on a table is associated with the onset of babbling (Eilers et al., 1993), independent sitting occurs alongside distinct change in vocalisations (Yingling, 1981), and walking occurs around the same time as the emergence of first words (Lenneberg, 1967). Babbling is a rhythmically organised motor act produced from the mouth (MacNeilage & Davis, 2000). As infants carry out rhythmic arm movements, it allows them to practice rhythmically timed actions used in babbling (Iverson et al., 2007). When an infant learns to sit upright the position of the body changes. Pressure is eased from the ribcage and lungs allowing deeper breaths that facilitate the production of longer utterances (Lee et al., 2010). An upright head position alters the position of the spine and vocal tract, and the tongue falls forward in the mouth. These new positions give the infant increased control over utterances and new opportunities for vocal production (Young, 2004). Practicing vocalisations in an upright position leads to more skilled and controlled vocalisations (Yingling, 1981). Walking offers an upright position that facilitates an infants' ability to explore their surroundings. Infants are able to locate and navigate towards distant objects of interest and to share these objects with their caregivers, thus facilitating the development of skills such as joint attention. When an infant shares an object with their caregiver, the caregiver is likely to vocally respond to the object (Karasik et al., 2009), and infants tend to learn words when attention is focused on a referent object (Tomasello & Farrar, 1986). It is clear that motor development has a direct influence on the development of language and communication skills in children.

Only one study to date has investigated the relationship between motor functioning and communication skills in 2-year-old children born with NE (Martinez-Biarge et al., 2012). The study identified communication impairments in all children

diagnosed with CP where CP was caused by significant injury to the BGT. Injury to the BGT resulting from NE was associated with oromotor dysfunction. Oromotor dysfunction causes deficits in controlling the lips, tongue and jaw. This consequently leads to feeding difficulties, difficulties with speech and language development, facial expression and use of gestures (Parkes et al., 2010). However, this study considered only one aspect of motor dysfunction and its impact on communication development in children born with NE diagnosed with CP.

Motor delay in children born with NE is common and the grade of NE tends to correlate with the motor abilities of the developing child (Jary et al., 2019; Lindstrom et al., 2006; Perez et al., 2013; Van Schie et al., 2015). Up to 50% of those born with moderate to severe NE develop motor specific disabilities such as CP as a result of neural damage (Dilenge et al., 2001; Pin et al., 2009). However, motor delay has also been observed in children without brain injury (Barnett et al., 2004; Jary et al., 2019; Van Schie et al., 2015). Research on typically developing children has found that motor and language skills are closely interrelated (Iverson, 2010; Leonard & Hill, 2014; Valla et al., 2020). Due to the risk of motor and language deficits in children born with NE future research should further investigate the relationship between these two developmental domains in this clinical group.

Conclusion

Standardised assessments of development are the most common tool used to assess cognitive, language and motor functioning in children born with NE. It has been established that the grade of NE, the extent of neural injury and the onset of disability are reliable predictors of poor developmental outcome scores in children born with NE. Those born with mild to moderate NE who do not meet these criteria tend to receive scores in the normal, but lower range on standardised assessments at an early age (Finder

et al., 2019; Marlow et al., 2005). However, reports of impairment seem to become more apparent as children get older (Lindström et al., 2006). The developmental profile of children born with NE is still relatively undefined, but research suggests that children in this clinical cohort are at an increased risk of developmental delay regardless of NE grade, extent of neural injury or disability.

Chapter Two

Parent-Child Interaction and Child Development

Introduction

As outlined in chapter one, NE has been found to have a lasting impact on child development. Whilst standardised assessments of development allow us to measure the current developmental abilities of children, they do not tell us about the underlying processes that may be influencing child development. Children predominantly acquire knowledge and skills through interaction with their primary caregivers. The direct observation of parents and their children at play is a common method used to study characteristic behaviours that might be associated with the developmental progress of children. Observing parent-child interactions allows us to monitor the impact of clinical conditions on children's behaviour such as temperament, attention skills and social interest. This method is also used to examine behaviours of the parent and whether certain parenting behaviours promote the development of children. Extensive research has been conducted on the interactions between parents and children, both in typically developing and clinical groups. Despite the impact of NE on children and their parents, the parent-child interactions of this clinical group have yet to be studied. This review of the literature aims to outline current parent-child interaction research in relation to child development and to examine the parent-child interactions of those in clinical groups that may share similarities with those affected by NE.

Parent-Child Interaction

During the early years of life, children primarily learn through the interactions they have with their primary caregivers. From birth into early childhood most children spend the majority of their time with their parents, particularly in western societies. Parents exhibit particular behaviours and create environments for their children that

enable nurturing, care and facilitate development (Bornstein, 2002). From birth, children also show a propensity for social interaction and are typically found to respond to social stimuli such as vocalisations and facial expressions (Bowlby, 1969). Therefore, it is proposed that parent-child interactions have an important influence on child development.

The relationship between a parent and their child is bidirectional in nature. The collaborative partnership between a child and their parent is therefore considered a 'dyad'. The parent and the child each have an influence on the other during interaction in the way that a parent responds to the child and the child responds to their parent (Sameroff, 2009). Parents play a crucial role in child learning as they provide an environment that fosters learning though guidance and support during interaction. Children are able to influence the care they receive from their parent by how they respond and parents in turn adjust and scaffold their behaviours according to characteristics of the child (O'Reilly & Bornstein, 1993; Vallotton, 2009). The Zone of Proximal development is a theoretical construct that describes child learning and development (Vygotsky, 1978). During back-and-forth interactions between a parent and a child, parents gauge the developmental capabilities of their child and adjust their learning environment by exposing them to challenging situations that are developmentally appropriate, thus maximising their child's learning potential.

Parenting styles are another theoretical approach used to characterise parent-child relationships. Such theories demonstrate the influence of parenting on child behaviours beyond the period of infancy. Baumrind's typology of parenting (1966; 1967; 1971) identifies three distinct parenting styles: permissive, authoritarian and authoritative. Parents categorised as permissive tend to be warm and nurturing towards their children. However, these parents also employ low levels of control and discipline, allowing their child to behave freely without punishment. Children of permissive parents tend to be

characterised as immature and have difficulties controlling their impulses. These children tend to do less well at school due to the lack of demandingness from their parents (Maccoby & Martin 1983). Authoritarian parents show low levels of warmth and responsiveness. They are demanding of their children and display high levels of control through strict rules with little room for negotiation or flexibility. Communication tends to be one-sided, with limited input from the child. Children of authoritarian parents are usually highly dependent on their parents. These children tend to be less sociable, more withdrawn and vulnerable to depression or stress (Maccoby & Martin 1983).

Authoritative parents are relatively strict and establish clear and consistent rules and expectations, but this is coupled with high levels of warmth and emotional support. Parents who use this parenting style encourage individuality and independence whilst providing guidance and discipline. These parents are open to communication with their child and the child actively participates in the interaction. Children of authoritative parents tend to be independent and self-motivated. They exhibit excellent prosocial skills, low levels of behavioural problems and usually perform well at school (Maccoby & Martin 1983). A study on preterm children and their parents found parenting styles to be associated with children's sensory and behavioural outcomes (Neel et al., 2019). Overall, the study found permissive parenting to be associated with poorer sensory and behavioural outcomes. Parents who used authoritative parenting had children with fewer behavioural problems. This suggests that parenting styles have an impact on the sensory and behavioural outcomes of children in clinical groups.

Parent-Child Interaction and Child Development

The behaviours of the parent and child during interaction have been found to have a meaningful influence on children's development and have therefore become an important focus of research. Childhood is a critical time for the development of cognitive,

language and social-emotional skills, and a time where parents are largely responsible for the care of their child. Thus, characteristics of parent-child interactions have been found to impact children's cognitive, language and social-emotional outcomes (Lugo-Gil & Tamis-LeMonda, 2008; Saint-Georges et al., 2013; Topping et al., 2013). Atypical patterns of behaviour observed during parent-child interactions have been linked with new-born medical status (Divitto & Goldberg, 1979) and developmental disorders such as autism (Wan et al., 2013), developmental delay and cerebral palsy (Festante et al., 2019).

Early theories of child development suggest that social interaction is crucial for child cognitive development (Bruner & Bornstein, 1989). Parents have a general idea of their child's developmental level and therefore have been found to expose them to situations that align with their cognitive level and interests accordingly (NICHD Early Child Research Network, 2005). Parents often exhibit behaviours that explicitly encourage cognitive stimulation and learning such as labelling objects by name or colour, asking questions that require problem solving or asking the child to follow a series of steps to complete an activity (Brady-Smith et al., 1999; Nandy et al., 2021). A number of parent qualities observed during parent-child interaction have been found to support children's cognitive development such as parental sensitivity, warmth and promoting child autonomy (Landry et al., 2000; Lugo-Gil & Tamis-LeMonda, 2008; Mermelshtine & Barnes, 2016).

Early developmental research has also highlighted the significance of social interaction on child language development (Bruner, 1983; Vygotsky, 1962). Social environments are thought to serve as a motivator for children to acquire language (Kuhl, 2007). Parent-child interactions therefore play a pivotal role in children's language development as they present situations where children are exposed to parental speech and engage in interactive behaviours that aid language development. Child-directed speech

(CDS) is the speech register that parents use to communicate with their children. Research has found that the quantity and quality of CDS has an impact on child language development (Rowe, 2012; Rowe & Snow, 2020). In typically developing samples of children, research has demonstrated that parents who speak more to their children acquire larger vocabularies (Hart & Risley, 1995). Similarly, children's language abilities are found to be superior when parents use lexically diverse and grammatically complex language (Rowe, 2012; Song et al., 2014). Parental behaviours aside from their CDS are also found to influence child language abilities. It has been found that parents who show higher levels of sensitivity and responsiveness have children with superior language abilities when compared to children who have parents that display directive and controlling behaviours (Bornstein et al., 2020; Neuhauser et al., 2018a; Song et al., 2014).

Social interaction is vital for the development of children's social-emotional abilities. Social-emotional development is defined as a child's ability to effectively communicate, form meaningful relationships and understand and regulate their emotions. Therefore, this domain of development has been found to have an influence on children's behaviour during parent-child interactions (Zeman et al., 2006). Parent-child interactions form the basis for social and emotional bonding as the parent responds to their child's needs. During interaction, parents also facilitate children's social-emotional development by demonstrating how to express and regulate their emotions appropriately (Bornstein & Manian, 2013; Grusec et al., 2000). The way in which parents display emotion and support children's emotions from infancy into childhood has been found to have long-term implications on children's social competence. Research has demonstrated that responsive and supportive parenting displayed during parent-child interactions at infancy has been found to predict children's social competence when they reach preschool age (Rispoli et al., 2013).

Influences on Parent-Child Interaction

The process of parenting model (Belsky, 1984) suggests that parenting behaviours are influenced by characteristics of the parent and the child. Parent characteristics include age, personality, psychological functioning, sense of parenting competency and attitudes towards parenting. For example, older mothers in their thirties have been found to display higher levels of supportiveness and closeness with their children compared to mothers in their teens and early twenties (Barnes et al., 2014). Characteristics of children that may impact parent-child interaction include age, gender, temperament and their developmental competence (Belsky, 1984). For example, when analysing parental speech input longitudinally the quality of parental speech changes as their child's language abilities develop over time. Parents are found to support their children's vocabulary development by exposing them to new types of language at various stages of development (Rowe, 2012).

Whilst characteristics of the parent and child dictate the course of parent-child interactions, it is important to consider that various external factors may also influence the content and quality of these interactions. Bronfenbrenner and Morris (2006) emphasise the importance of the bioecological model of development. The model describes the contribution of environmental factors to parenting behaviours and how present surroundings can impact child development. For example, converging evidence suggests that socioeconomic status (SES) has an impact on parent-child interactions. In contrast to parents with lower-SES, parents with higher-SES tend to display more cognitively stimulating behaviours, higher levels of positive affect and sensitivity towards their children (Tamis-LeMonda et al., 2009). Higher-SES parents are more financially able to provide resources that facilitate learning such as age-appropriate toys and books (Chen et al., 2018). Lower-SES households are generally more crowded compared to

higher-SES households (Lecheile et al., 2020) and parents may have to work longer hours leading to a reduction in quality time between the parents and their children.

Parental stress is an additional factor that has been found to negatively influence parent-child interactions. Stress in parents can be caused by a number of reasons such as lower-SES (Hoff et al., 2002) or caring for a child with a disability (Esdaile, 2006). Parental stress might be exacerbated in those with children with clinical conditions as they are more likely to encounter uncertainty in their child's development. Parents might find it difficult adapting to accommodate a child with a disability or require extra resources to support their child. It has been observed that parents who show higher levels of stress are more likely to use harsher parenting practices, show less sensitive behaviours and lower levels of supportive behaviours (Guajardo et al., 2009; Jackson & Choi, 2018; Neuhauser, 2018). The negative impact of stress on parent-child interaction has been linked with poorer developmental outcomes in children (Magill-Evans & Harrison, 2001).

Analysing Parent-Child Interactions

The direct observation of parents and their children at play is the gold standard method used to study parent-child interactions. Parent-child interactions are typically video recorded and observational coding schemes are used to characterise behaviours of interest during the interaction (Bakeman, 2005). Both global and microanalytic observational coding schemes can be used to categorise qualities of parent-child interactions (Morawska et al., 2015). Global rating tools are used to assess general constructs of interest during instances of parent-child interaction. The behavioural quality is typically scored on a scale, rating the behaviour level from high to low (i.e. high frequency of the behaviour examined during the interaction to infrequent occurrence of the behaviour observed). One rating is given to each parent-child interaction recording for each construct of interest. In contrast, microanalytic coding systems are used to code all

occurrences of clearly defined behaviours during parent-child interactions. Fine-grained characteristics of the interaction are captured by coding the occurrence of behaviours on a second-by-second basis.

General Constructs of Parent-Child Interaction

A broad range of constructs have been investigated relating to behaviours of the parent, the child and the dyad. Some common constructs measured during parent-child interaction have been found to facilitate child development such as caregiver sensitivity. Caregiver sensitivity refers to a parent's ability to appropriately interpret and respond to their child's cues in a warm and engaging manner (Claussen & Crittenden, 2000). Children who are exposed to higher levels of caregiver sensitivity are found to have more advanced cognitive (Feldman et al., 2004), language (Bornstein et al., 2020; Tamis-LaMonda et al., 2001) and attention skills (Gartstein et al., 2008; Graziano et al., 2011).

Other parental behaviours may interfere with developmental outcomes such as directiveness. Caregivers who show high levels of directive behaviour tend to be demanding or controlling over their children. They exhibit more instructional language and are more dominant during the play session (Egeland et al., 1993; Ispa et al., 2013). Parents who show lower levels of directiveness tend to follow the child's lead during play and provide feedback that relates to the child's focus of attention. Typically developing children with highly directive parents are less likely to develop advanced cognitive skills when compared to children exposed to lower levels of directive parenting (Bernstein et al., 1987). However, more recent research suggests that directive behaviours may be beneficial for children, particularly in those with clinical conditions (Bodner et al., 2020; Freeman & Kasari, 2013). Directive behaviours have been found to promote engagement, responsiveness and joint attention in clinical groups of children who are more likely to

demonstrate higher levels of passivity and lower social interest (Bodner et al., 2020; Bottema-Beutel et al., 2018).

Child behaviours that are commonly measured in the context of parent-child interaction include child attention, affect or responsiveness (Brady-Smith et al., 1999; Doussard-Roosevelt et al., 2003; Kochanska & Askan 2004; Wan et al., 2013). Coding schemes that assess child qualities during parent-child interaction observe behaviours such as initiating or maintaining interaction with their parent, the degree of positivity used when interacting with their parent and the degree of interest the child pays to the parent, such as the use of eye-contact, gaze following or exploring an object to which the parent directs attention (Brady-Smith et al., 1999; Wan et al., 2017). Studies have found that behaviours of the child may be predictive of future clinical outcomes (Wan et al., 2013). For example, Wan and colleagues (2013) found children's higher levels of negative affect and lower attentiveness to the parent measured at 12 months of age predicted autism outcome at 3 years of age.

The parent and the child must interact and respond to each other in order for the interaction to be mutual and engaging. Responsiveness is a construct that is often studied in the context of parent-child interactions. When a parent is responsive to their child they may vocalise to the child in a prompt and appropriate manner, provide tactile arousal, or provide positive encouragement of attention towards an object or situation of interest. They might respond to the child's social prompts by imitating the child's vocalisations, physically orient towards the object explored by the child, and display warmth and positive affect (Bornstein & Tamis-LeMonda, 1989). Features of child responsiveness include eagerness to engage with their parent, promptness, sincerity of the response, and whether the child's reaction will please the parent (Kochanska & Aksan, 2004).

Research suggests that the variation in responsiveness may be dependent on the child's cognitive skill and whether the parent matches their responses according to the child's current interests. Children are more likely to respond to their parent's bids if the bid is related to the activity the child is currently engaged in, and if it is within the child's developmental range of understanding (Mahoney & Neville-Smith, 1996). Thus, it has been established that responses from the parent must be contingent in order to increase the child's level of engagement during an interaction.

Contingency involves prompt and meaningful responses between a parent and their child and promotes fluent and mutual communication within the dyad (Bornstein et al., 2008; Masek et al., 2021; Tamis-LeMonda et al., 2001). Contingent responsiveness is typically examined in terms of semantic and/or temporal contingence. Semantic contingency considers how meaningful a verbal response is in relation to the current action or topic of interest of the social partner (Harkness, 1988). Temporal contingency evaluates the promptness of the response to a social bid, the optimal window of response is typically one to five seconds after the behavioural or verbal bid (Bornstein et al., 2008). For example, during play the child picks up a ball and the parent says, "That's a bouncy ball. What colour is the ball?" The response is temporally contingent if the parent's verbal response was prompt, occurring during or shortly after the child's action. It is also semantically contingent as the verbal response relates to the child's behaviour (i.e. describing the object the child is holding).

From the second year of life children's expressive language abilities tend to rapidly develop (Vihman, 2018). Thus, the ability to take part in back and forth conversations with their parents typically increases from this period. It is suggested that parental contingent responsiveness encourages children to use more complex vocalisations, leading to increasingly advanced responses from their parents (Goldstein &

Schwade, 2008; Gros-Louis et al., 2014; Masur & Olson, 2008). When parents and children practice contingent back and forth communication the complexity of these conversations continuously advances (Edmunds et al., 2019).

Research consistently demonstrates that when parents respond to their children in a contingent manner, children acquire language rapidly and more easily compared to children whose parents do not demonstrate contingent responses (Roseberry et al., 2014; Tamis-LeMonda et al., 1998). Contingency has been found to be a significant predictor of vocabulary diversity in typically developing children (Gilkerson et al., 2018; Tamis-LeMonda et al., 2014) and of children in clinical groups such as slow-to-talk children (Conway et al., 2018b) and children with developmental delay or a diagnosis of autism (Adamson et al., 2019).

Global features of the dyad are also invoked to characterise the quality of a parent-child interaction. A dyadic interaction brings the parent and the child into a sphere where each person's behaviours, verbalisations and emotional states influences the other and takes into account the cooperation between a pair of individuals. Dyadic mutuality is a broad construct and many overlapping features are used to describe the term in the literature. Coding schemes have been devised to capture the coordination of behaviour between the parent and child during interaction. Features such as mutuality, engagement and synchrony have been studied (Funamoto & Rinaldi, 2015). A common quality of the dyad that has been investigated in the context of parent-child interaction is dyadic mutuality. Dyadic mutuality describes the degree of attention, reciprocity and synchrony shared in the parent-child dyad. It is expressed through the child's acceptance of the caregiver's involvement and is represented as a smooth back and forth interaction characterised by mutual enjoyment, cooperation and responsiveness (Deater-Deckard &

Pentrill, 2004; Wan et al., 2017). The parent's response to the child and the child's response to the parent are the defining features of dyadic mutuality.

Four operationalised indicators have been used to describe parent-child mutuality (Deater-Deckard & Petrill, 2004). First, the parent's response to the child must be immediate and contingent, and second, the child must respond to their parent's communicative bids. The third consideration is the reciprocity of behaviour and emotion within the dyad, which is characterised by the use of eye contact and shared positive or neutral affect during the interaction. Lastly, turn-taking of verbal and non-verbal behaviours between the parent and child must be carried out.

Dyadic mutuality contributes to healthy social development and is associated with fewer behavioural problems in children (Deater-Deckard & Petrill, 2004). Higher levels of dyadic mutuality observed during parent-child interactions is also associated with superior cognitive outcomes in children five years of age (Kirsh et al., 1995). Poehlmann and Fiese (2001) found that reciprocal and engaging dyadic interactions mediate the relationship between neonatal risk status and child cognitive outcomes in later life. Lower levels of dyadic mutuality have been linked to symptom severity in children with clinical conditions such as autism (Wan et al., 2013). The majority of studies that investigate dyadic mutuality use global analytic coding schemes to rate the extent of dyadic mutuality during parent-child interaction (Aksan et al., 2006; Deater-Deckard et al., 1997). In order to examine dyadic mutuality on a microanalytic level, constructs that underpin dyadic mutuality have been outlined including joint attention and child directed speech.

Joint Attention

Joint Attention (JA) is the act of engaging in a shared activity or mutually focusing on an object with a social partner (Bakeman & Adamson, 1984). Successfully

participating in JA therefore reflects high dyadic mutuality. JA is initiated when an individual directs the attention of another using verbal or behavioural indications, such as eye-gazing or pointing to an object or event (Schertz et al., 2018). When responding to JA, one member of the dyad follows the gaze, gesture or verbal cue of their communicative partner (Adamson et al., 2010).

In typically developing children, JA skills begin to develop from infancy and advance through early childhood as children develop their language skills and social cognition (Charman, 2003). From birth infants develop the ability to orientate towards human faces and follow moving objects of interest (Scaife & Bruner, 1975). At around 3 to 6 months of age infants learn how to attend to both their mother and objects simultaneously which allows opportunities for shared learning (De Barbaro et al., 2016). Infants begin to direct the attention of others towards objects of interest using vocalisations and gestures such as pointing from 6 to 12 months of age (Bakeman & Adamson, 1984; Carpenter et al., 1998; Moore et al., 2014). This allows the infant to initiate JA with others. From 18 months of age toddlers start to use language (Gilkerson et al., 2017), allowing them to coordinate attention with others verbally and to request objects or activities that are not present in the immediate environment. From the second year of life toddlers frequently engage in JA with a social partner (Eggebrecht et al., 2017; Mundy et al., 2007) and the skills required for JA continue to be refined through early childhood (Adamson et al., 2004).

Child development is facilitated through shared knowledge and experience with the support of their caregivers (MacPherson & Moore, 2017). Thus, JA has been found to optimise a child's capacity to gain knowledge from the social interactive environment. JA has been positively linked to abilities in children such as language skills (Mundy & Gomes, 1998; Salo et al., 2018; Tomasello & Todd, 1983), social cognition (Mundy &

Gomes, 1998), visual attention (Striano & Stahl, 2005) and academic success (Mundy & Newell, 2007). Partaking in episodes of JA with their parent is a prerequisite for children's learning and has been shown to facilitate language development. Children are observed to have larger expressive language vocabularies if they spend longer periods of time engaged in JA (Carpenter et al., 1998; Tomasello & Todd, 1983). Similarly, JA can help support cognitive development. As children begin to understand the influence they have on the behaviour of others, they can initiate instances of JA that allow them to explore novel circumstances in the world around them with support and guidance from their parent.

JA in Clinical Populations

Research demonstrates that children in clinical populations display differences in JA as their condition may impact skills such as gaze following and social referencing (Dean et al., 2020; Spiker et al., 2002). Investigating JA in clinical groups helps us understand how particular deficits might impede children's development, but it can also demonstrate how JA can help facilitate children's outcomes, particularly in children at risk of developmental delay. The ability to take part in instances of JA is compromised in children with disabilities such as deafness (Paparella & Kasari, 2004), blindness (Grumi et al., 2021) and selective mutism (Nowakowski et al., 2011) as the skills required to initiate or respond to JA cues are directly affected in these individuals. Consequently, children in these clinical groups take part in significantly fewer episodes of JA compared to typically developing children. When directing the attention of a child who is hard of hearing, the child must look away from the object of interest to receive communication about the stimuli through sign language (Paparella & Kasari, 2004). Children who are blind cannot see visual stimuli and must therefore find alternative routes to co-reference with their parents (Grumi et al., 2021). Children with selective mutism are less likely to

initiate or respond to instances of JA using language, particularly if they find a situation overwhelming or stressful (Nowakowski et al., 2011).

JA in other clinical groups has been widely studied. Children diagnosed with autism experience difficulties with social communication and social interaction and display restrictive or repetitive behaviours (APA, 2013). They often experience language delay (Tager-Flusberg, 2016), and display lower levels of social engagement, prosocial behaviours and communicative gestures (Osterling et al., 2002). Due to these symptoms, JA in dyads where the child has autism may differ when compared to dyads with typically developing children (Mundy, 1995).

Eye-gaze is an important function of JA as it is used to orient the social partner to a mutual point of focus. However, differences in eye-gaze behaviour have been observed in children with autism which may, in turn, impact JA. Thorup and colleagues (2016) used a live eye-tracking paradigm to test gaze-following abilities in children with autism and typically developing children. The experimenter had two toys in front of them to their left and to their right. The experimenter looked to one of the toys, either by shifting their eyes to the object, or turning their whole head and eyes. Typically developing children followed the gaze of the examiner using both types of cues equally well. Children with autism were more likely to follow the gaze of the experimenter if the whole head and eyes shifted to the object, compared to gazing with just the eyes meaning they may miss a number of parental cues for JA.

Children with autism demonstrate “sticky fixation” (Elsabbagh et al., 2013) or difficulty shifting their attention away from a target on which they are fixated. Sacrey and colleagues (2013) filmed children aged 6 to 36 months playing with small graspable toys. Videos were analysed to measure the time the child spent looking at the toy before they moved their hand toward it, and the duration of time they spent looking at the toy after

grasping it. Children with autism showed prolonged latency to disengage their attention from the toy. Sticky fixation patterns and lower levels of gaze following in children with autism may impede the child's ability to shift their attention to parental bids of JA, leading to lower levels of JA in this clinical group.

Repetitive symptoms in children with autism have also been found to disrupt play during instances of parent-child interaction. Hudry and colleagues (2013) found repetitive behaviours in children with autism predicted reduced JA during parent-child interaction in children 2 to 5 years of age. Children with autism are less able to sustain JA as repetitive behaviours disrupt the child's attentional focus, and parents are less able to read the child's behavioural prompts and interests.

Children with Attention deficit hyperactivity disorder (ADHD), characterised by inattention, impulsivity and/or hyperactivity (APA, 2013), also display deficits in attentional shift (Sokolova et al., 2017). In one study, children were administered an eye-tracking task measuring sustained attention at 3, 6, 12 and 24 months of age (Miller et al., 2018). ADHD diagnosis was established when the children were 8 to 10 years of age. The children who went on to receive a diagnosis of ADHD demonstrated atypical patterns of sustained visual attention compared to the non-diagnosed children. The children diagnosed with ADHD had higher overall looking times in infancy. Longer looking times has been linked to deficits in attention shifting, slower cognitive processing and poorer developmental outcomes (Cuevas & Bell, 2014), which in turn might have a negative impact on the child's ability to respond to initiations of JA.

Symptoms of hyperactivity have also been shown to compromise instances of JA between the developing child and their parent. Levels of attentiveness were measured in children aged 1- and 2-years during observations of free play during parent-child interaction. When the children turned 3.5 years of age levels of hyperactivity were

measured (Lawson & Ruff, 2004). Overall, it was found that attentiveness during free play was negatively associated with their hyperactivity score indicating that symptoms of hyperactivity compromised the child's attention during play with their parents. Children who exhibit symptoms of hyperactivity might find it harder to engage in periods of JA with their parents due to differences in orientating and fixation times related to hyperactivity. Similarly, parents of children who show symptoms of hyperactivity might find it difficult to recognise their child's behavioural initiations for JA.

Child-Directed Speech During Parent-Child Interactions

As noted, qualities of dyadic mutuality such as joint attention observed during parent-child interaction have been found to play an important role in supporting children's development. However, it is also important to consider individual behaviours that shape the dyadic interaction. Research has identified that the language parents use during interaction is also an important factor that influences child outcomes, particularly language development (Rowe, 2012; Tamis-LeMonda et al., 2001). Language is the primary means by which a parent conveys information to a child, and through language, children are able to interact with the world around them. Analysing the language parents use during periods of parent-child interaction gives us an insight into how parental language influences children's development.

When parents communicate with their children they systematically alter features of their speech to get the attention of their child, and to ensure the child understands what the parent is trying to convey. Child-directed speech (CDS) is the speech register used when communicating with children. When adults talk to children, they use a different pattern of speech compared to when they are talking to other adults. When comparing CDS to adult directed speech, in addition to signature prosodic modifications, adults tend to use fewer words and shorter utterances (Fisher & Tokura, 1995; Soderstrom et al.,

2008), ask more questions and use more isolated words when talking to children (Soderstrom et al., 2008). The use of CDS is critically important for children's language development and social competence (Tamis-LeMonda et al., 2001). It is also useful for conveying affect (Trainor et al., 2000), and for gaining and maintaining child attention (Dunst et al., 2012). Several features of CDS are considered to be important to children's linguistic functioning, including parental speech quantity and quality.

Quantity of CDS

The quantity of parental speech input has been found to be crucial to child language development. Parental speech quantity can be measured by counting the total number of words, word tokens or utterances spoken during an interaction (Rowe, 2012). In 1995, a study by Hart and Risley identified a 30-million-word gap in words heard by age four when comparing children raised in lower-SES homes to children raised in higher-SES homes. A higher number of words heard corresponded to better language outcomes in children. Children raised in high-resource homes were more likely to display superior language skills compared to those raised in low-resource homes. This finding established that parental speech quantity plays a significant role in child outcomes and that children who hear more words know more words. Since this finding was established, many studies have investigated the impact of parental speech quantity on child language outcomes in both typically developing and clinical populations (Kuchirko, 2019).

Parents are prone to adjusting their language input according to the developmental level of their child (Snow, 1995). Therefore, if a parent of a child with a clinical condition perceives their child to be developmentally delayed they may simplify their language to ensure the child understands what they are saying. Children born preterm and those with a diagnosis of autism are at a higher risk of speech and language delay (Delehanty et al., 2018; Foster-Cohen et al., 2010) and are therefore more likely to demonstrate lower

levels of verbal productivity and communicative exchanges (Brosch-Fohraheim et al., 2019; Salerni et al., 2007). It is anticipated that parents of children in these clinical groups might decrease their quantity of speech input to match the verbal productivity of their children. However, Salerni and colleagues (2007) measured the quantity of tokens and frequency of utterances spoken per minute by mothers of children born preterm and by mothers of typically developing children. The study failed to reveal any significant differences in maternal speech quantity between the two groups indicating that mothers of children born preterm spoke to their children just as often as mothers of typically developing children.

Similarly, when comparing the speech quantity of parents with autistic children to the speech of parents of typically developing children, the majority of studies did not find significant differences in the number of utterances spoken from the parent to the child during periods of parent-child interaction (Bang & Nadig, 2015; Watson, 1998, Wolchik & Harris, 1982). However, research has established a link between language input quantity and child language outcomes in children with autism. A significant positive association between the number of word tokens used by adults, and children's later vocabulary production has been demonstrated during adult-child interactions where the child has a diagnosis of autism (Warren et al., 2010). A similar relationship has been observed in children born preterm: the more parents spoke to their child when they were in the NICU, the higher the child's cognitive and language scores were at 18-months of age (Caskey et al., 2014).

These findings suggest that language delay commonly observed in children in clinical groups is not a consequence of experiencing less parental speech than their typically developing peers. Children born preterm tend to be more passive and display lower levels of social attention (Anderson et al., 2011; Field, 1983). Similarly, children

with autism have been observed to frequently ignore their mothers and shift their attention elsewhere (Doussard-Roosevelt et al., 2003). Despite being exposed to similar amounts of speech as typically developing children, children in these clinical groups might not be paying attention to a significant proportion of words being spoken to them leading to a mismatch between parental expectations and the child's capacity to process language. Although parents have the intention of supporting their children's development, children in these groups might find typical quantities of speech to be overstimulating or intrusive. Indeed, over stimulating and intrusive behaviours displayed by parents have been associated with deficits in children's later expressive language ability (Bruce et al., 2022; Crnic et al., 1983). It is proposed that the right balance of directive and supportive behaviour must be established, and parents must adjust their level of language to suit their child.

Quality of CDS

Whilst the quantity of parental speech input has been demonstrated to have an important influence on children's language development, the influence of parental language is complex and nuanced and goes beyond the number of words a parent says to their child (Masek et al., 2021). Rowe (2012) established that the quantity of parental speech is important for children's language development at around two years of age, whereas the quality of parental speech plays a significant role in children's linguistic ability in those aged three years and older. It is suggested that the quality of parental language might have an overall greater influence on child language outcomes at older ages (Anderson et al., 2021). A review by Rowe and Snow (2020) categorised parental language input quality along three dimensions including conceptual features (for example, decontextualized talk), linguistic features (such as syntactic complexity and vocabulary diversity) and interactive features (for example, conversation and turn-taking).

The association between parent's linguistic features and child outcomes of those in clinical groups has been widely studied. Therefore, this section of the review will primarily focus on the literature outlining parental linguistic features, namely syntactic complexity and vocabulary diversity.

Syntactic complexity describes the sophistication of language used by parents and is often measured by calculating the Mean Length of Utterance (MLU) (Brown 1973). A higher MLU indicates the use of more sophisticated language, whereas shorter utterances suggest the use of simple grammatical structures (Soderstrom, 2007). An early study by Frank and colleagues (1976) reported a positive correlation between maternal and child MLU. When mothers used more sophisticated language with their children, children's syntactic language skills are observed to be more advanced. Vocabulary diversity refers to the number of different word types a parent uses when speaking to their children (Anderson et al., 2021). In general, research has found a positive association between parent language quality and child language abilities and it has been observed that parent who use more diverse language tend to have children with bigger vocabularies (Pan et al., 2005).

CDS in Clinical Groups

When observing the quality of speech input of parents with children with disabilities, it is commonly assumed that parents will use lower quality speech input in an attempt to match their speech to the language ability of their children (Conti-Ramsden, 1994). When comparing the quality of parental language input of children at-risk of delay to parents of typically developing children, the findings are in fact mixed. In a study of children born preterm, lexical diversity and syntactic complexity of maternal speech directed to children was analysed (Suttora & Salerni, 2011). It was found that the overall quality of verbal input addressed to the preterm children did not differ from the language

used by mothers of full-term children. Similarly, the lexical diversity of parental input of mothers with children with autism was compared to mothers of typically developing children (Bang & Nadig, 2015; Swensen, 2007). Overall, no significant differences in maternal lexical diversity were observed between the two groups including word types, noun types and the percentage of different nouns used. Despite displaying similar levels of lexical diversity, mothers of children who were later diagnosed with autism have been reported to use shorter utterances during parent-child interaction compared to mothers of typically developing children (Brisson et al., 2014). Similarly, Choi et al. (2020) found that parents of children with an elevated risk of autism produced utterances with a lower MLU compared to parents of children with typical risk of autism. However, other studies have observed no significant differences in respect to parental MLU between dyads with a child with autism and those typically developing (Bang & Nadig, 2015; Swensen et al., 2007; Wolchik & Harris, 1982).

Rowe et al. (2009) investigated the quality of linguistic input of parents with children who endured early brain injury. In this study the early brain injury group were compared to dyads with children born without complications. When the children were 30 months of age, the parents and children were videotaped for 90 minutes engaging in day-to-day activities. MLU and the number of word types used by the parent was compared between each group. It was hypothesised that parents of children with early brain injury would use more simplified speech with their children, but the study found no significant differences in MLU or number of word types used by each group of parents. Although the quality of parental language was similar between the two groups, the study found differences in the predictive strength of parent's language quality on children's language outcomes. The study found that the quality of parental language input predicted children's vocabulary growth equally well for both groups. However, parental language quality was

a more potent predictor of syntactic growth for children with early brain injury than for children born without complication. This suggests that the quality of parental linguistic input plays a significant role in syntactic growth in children with early brain injury.

Parent-Child Interactions in the Context of NE

The adverse outcomes associated with NE means that mothers and children impacted by the condition might show behaviours that are characteristically different when compared to those who do not experience complications during birth. The impact of clinical outcomes associated with NE on the child combined with the experience of a traumatic birth could affect the transactional relationship between the child and their mother, subsequently modifying characteristics of parent-child interactions. Any variation in behaviours observed during parent-child interactions may mediate the effect of NE on developmental outcomes.

The clinical profile of NE means that children who endure the condition are at significant risk of developmental delay compared to children who are born without complication (Volpe, 2018). Similar to other clinical groups, NE has been associated with language delay (Shapiro et al., 2017), attention issues (Tonks et al., 2019) and problems with motor development (Jary et al., 2019). Children born with NE are 5.9 times more likely to receive a diagnosis of autism (Badawi et al., 2006). These disadvantages that arise as a result of NE might compromise the behaviour of the child during periods of parent-child interaction.

In a qualitative study by Herringhaus and colleagues (2013), parents reported their experience of caring for a child born with NE. Mothers felt that the prolonged separation and lack of contact during treatment in the NICU hampered the initial parent-infant bond. TH, the primary treatment method administered to treat infants born with NE, involves 72 hours of restricted parent holding and skin- to-skin contact of the infant as it is required

that the whole body is cooled. During treatment, parents reported feeling reluctant to touch their infant because of the high-tech equipment attached to them during their time in the NICU. These experiences may therefore impact the behaviour of the mother during parent-child interaction. Parent mental health and the social and language outcomes of children born with NE are found to be linked (Danguecan et al., 2020). However, the mechanisms of the relationship between these factors have not yet been investigated.

To date, no studies have researched the impact of NE on parent-child interactions. Taking account of the known outcomes of children and parents impacted by NE we can infer how the condition might alter behaviours during parent-child interaction, and whether dyads in this group might display characteristic differences during interaction when compared to children born without complication and their parents. As qualities of parent-child interactions have been widely researched in other clinical conditions, we can also use these findings to understand the potential impact of NE on dyadic interactions.

Conclusion

Parent-child interactions are crucial to healthy child development. When a child and parent take part in mutual activity, they influence each other's behaviour thus shaping the course of the interaction (Sameroff, 2009). Features of dyadic interactions such as joint attention, and parental speech input is linked to a number of child outcomes such as cognitive and language functioning. The investigation of parent-child interactions in clinical groups and in typically developing children has allowed us to establish qualities of interactions that influence children's development.

There is very little research on the influence of NE on children and parents outside the medical literature. Current studies tend to focus on the impact of clinical outcomes on standardised assessment scores (Sharpiro et al., 2017; Weeke et al., 2018) and qualitative studies on parental experiences of NE (Lemmon et al., 2017; Herringhaus et al., 2013).

The developmental outcomes of children born with NE are mixed and a significant proportion of children impacted by the condition are at-risk of developmental delay (Pappas et al., 2015; Perez et al., 2013; Schreglmann et al., 2019). Despite being widely studied in typically developing samples and other clinical groups, no studies to date have investigated parent-child interactions in the context of children and parents affected by NE. Studying individual differences in parent-child interactions in this clinical group will allow us to explore the variability of outcomes observed in children born with NE and to examine factors that support or impede their developmental outcomes.

Chapter Three

Methods

Current Study

The current study is part of a larger prospective longitudinal research project conducted by the NIMBUS (Neonatal Inflammation and Multiorgan dysfunction and Brain injury research group) and NEPTuNE (Neonatal Encephalopathy PhD Training NEtwork) groups. The research is funded by the Health Research Board (HRB) and involved the recruitment of children born with NE at birth from three maternity hospitals in Dublin, Ireland including the Coombe Women and Infants Hospital, the National Maternity Hospital and the Rotunda Hospital. When an infant was admitted to the neonatal unit for NE, our collaborating medical doctors (M.O and T.H[1]) were informed and they approached the family for consent to take part in the research project. This study involved the follow-up of this sample using a standardised assessment of development when the children turned 2 years of age or older.

The control group included in this study (the No-NE group) consists of typically developing children born without complications. When children were 2 to 4 years of age, parents and their children were invited to the Infant and Child Research Lab based in Trinity College Dublin to take part in a longitudinal study of parent-child interaction and a standardised assessment of development was administered. Participants were recruited by L.K[1] through social media and flyers distributed in local creches and supermarkets.

Ethical Considerations

This project received ethical approval from the research ethics committees at the three participating maternity hospitals. Further approval was granted by the School of

[1]See Appendix 1 for contributions to work.

Psychology Research Ethics Committee at Trinity College Dublin (SPREC092014-01). This study complies with the Declaration of Helsinki's ethical standards. Written informed consent was obtained from parents on behalf of themselves and their child at each stage of recruitment, prior to the commencement of testing.

Participants

One hundred and five infants born with NE were recruited at birth between 2016 and 2021. From this sample 10 infants died due to NE. A proportion of those were lost to follow up as they did not consent to being re-contacted, were uncontactable, moved away or chose to drop out of the study. A number of infants were outborn patients transferred to one of the three Dublin maternity hospital to receive specialist treatment such as TH during the neonatal period. These families live out of county and many did not consent to follow-up as they lived too far from the laboratory to participate in the study. A total of 53 children born with NE and their parents re-enrolled in the study and returned for follow-up when the child was 2 years of age or older. Eighty-one children were recruited in the No-NE group. Using a standardised effect size (Cohen's d) of 0.5, this sample size was calculated to detect a medium effect size with 80% power at the 5% level of significance. Many of the behavioural and biological functions tested have not been previously studied in this population, therefore this estimate should be considered as an approximation.

Inclusion and Exclusion Criteria

When recruiting infants born with NE at birth, the inclusion criteria defined by Huang et al. (1999) were used to identify infants with the condition. In the immediate postnatal period infants exhibited abnormal neurological signs such as seizures or hypotonia, organ dysfunction and at least two or more of the following markers including: the need for resuscitation after birth, cord blood with a pH less than 7.2 and

presented evidence or suspicion of HIE based on foetal distress, loss of heart beat and/or abnormal foetal scalp pH. Infants were also recruited if they sustained early brain injury diagnosed using cranial ultrasound in the first 48 hours of life. Infants born with NE were excluded from the study if they were born with congenital abnormalities or had been affected by maternal substance misuse.

In the No-NE group, full-term born typically developing children were eligible to take part in the study if the child was 2 to 4 years of age at the time of assessment. Children were excluded from the group if they were born pre-term, endured any significant health problems at birth or in early life, or had any known developmental risk factors. None of the children in the no-NE group were affected by neonatal risk factors such as maternal infection, difficulty feeding, delivery complications or spent significant time in the NICU.

Propensity Score Matching

When comparing the NE and No-NE group, unequal group sizes and statistically significant differences in participant age were observed at each stage of assessment. Therefore, propensity score matching (PSM; Austin, 2011) was used to match participants in the NE and No NE group by age. PSM is a statistical technique that is used to select control participants by accounting for covariates. Differences in age between the NE and No NE group increases the possibility of bias as age may influence outcomes rather than the condition itself. PSM reduces selection bias, improves effect estimates and strengthens causal inferences by matching participants with similar characteristics. PSM uses a regression model to calculate a propensity score that predicts the probability of belonging to the NE group or the comparison group based on chosen covariates.

In the current study, children were assessed at one of two time points using an age appropriate developmental assessment. Children aged 2 to 3 years of age were assessed

using the Bayley-III (Bayley, 2006) and children 4 years and older were assessed using the WPPSI-IV (Weschler, 2013). PSM was calculated for each group to establish age appropriate matches. Age matched comparisons for 31 children born with NE were selected from a database of 81 typically developing children in the group assessed using the Bayley-III. Matches for 18 children born with NE were selected from a group of 35 typically developing children assessed using the WPPSI-IV.

Descriptive statistics comparing the sample before and after matching is outlined for the group who completed the Bayley-III (see Table 3.1) and for those who completed the WPPSI-IV (see Table 3.2). A standardised difference in means (standardised bias) was calculated to compare the propensity scores for each sample before and after PSM is conducted. In the cohort assessed using the Bayley-III, the standardised bias for mean propensity scores before matches were established was 0.75, this value decreased by 58.70% to 0.31 after matching. The propensity score variance was 0.001 for the No NE group and 0.000 for the NE group, thus establishing a variance ratio of 0.

Table 3.1. *Descriptive statistics for the age of participants at Bayley-III assessment.*

	Before (N=125)					**After** (N=62)				
	M	*SD*	*Min*	*Max*	*p*	*M*	*SD*	*Min*	*Max*	*p*
No NE	24.05	1.36	21.17	27.13		24.37	1.36	21.17	27.03	
NE	29.57	7.63	23.10	46.37	.000	24.86	1.20	23.10	28.73	.137

In the cohort assessed using the WPPSI-IV, the standardised bias for mean propensity scores before matching was 0.53, this decreased by 24.1% to 0.41 after matching. A variance ratio of 0.3 was established.

Table 3.2. *Descriptive statistics for the age of participants at WPPSI-IV assessment.*

	Before (N=54)					**After** (N=36)				
	M	*SD*	*Min*	*Max*	*p*	*M*	*SD*	*Min*	*Max*	*p*
No NE	53.24	3.30	47.90	59.77		53.64	3.04	50.23	59.77	
NE	56.84	6.71	45.10	69.40	.011	56.14	6.15	45.10	66.60	.130

Sample Characteristics

Clinical Characteristics of NE Group. During their stay in the NICU, infants born with NE underwent daily clinical neurological examination and the grade of NE was assigned using the Sarnat grading system (Sarnat & Sarnat, 1976). The Sarnat grade equates to the most severe grade of NE designated to the infant whilst in the NICU. Clinical demographic information was collected for each infant born with NE including gestational age, birth weight, and whether the child was treated using TH. Additional factors that may contribute to neonatal risk are also reported including: if the infant had an abnormal neural MRI scan, whether the infant had seizures, a 5 minute Apgar score less than 7, a cord blood pH less than 7.2, whether the infant was intubated, lack of suck and feeding problems, the presence of maternal infection, if the infant was hospitalised for more than ten days and if additional delivery complications arose such as fetal bradycardia, meconium aspiration, maternal haemorrhage, placental abruption or nuchal cord complications. Clinical information is presented according to NE grade in Table 3.3. A majority of the sample was diagnosed with moderate NE (grade II). Due to the small number of participants diagnosed with mild (grade I) and severe NE (grade III) group comparisons were not established.

Table 3.3. *Clinical demographic information of the NE group.*

Variables	**NE I** (*N=4*)	**NE II** (*N=42*)	**NE III** (*N=7*)	**Total** *(N=53)*
Gestational Age (*Weeks, SD*)	40.83 (1.28)	39.39 (1.90)	38.96 (2.95)	39.45 (1.98)
Birth Weight (*KG, SD*)	3.96 (6.59)	3.38 (6.50)	3.28 (8.22)	3.41 (6.85)
Gender (*Male, %)*	3 (75.00%)	26 (68.42%)	2 (28.57%)	31 (63.27%)
TH (*Yes, %)*	1 (25.00%)	35 (100.00%)	7 (100.00%)	42 (95.45%)
MRI (*Abnormal, %)*	0 (0%)	14 (31.11%)	4 (57.14%)	18 (40.00%)
Seizures (*Yes, %)*	1 (25.00%)	15 (39.47%)	4 (57.14%)	20 (50.00%)
5 Min Apgar <7 *(Yes, %)*	1 (25.00%)	21 (50.00%)	7 (100%)	29 (54.72%)
Cord Blood <7.2 *(Yes, %)*	0 (0%)	22 (52.38%)	4 (57.14%)	26 (49.06%)
Intubated *(Yes, %)*	2 (50.00%)	19 (45.24%)	4 (57.14%)	25 (47.17%)
Suck/Feeding Problems *(Yes, %)*	0 (0%)	3 (7.14%)	1 (14.29%)	4 (7.55%)
Maternal Infection *(Yes, %)*	0 (0%)	5 (11.91%)	0 (0.00%)	5 (9.43%)
Hospitalised >10 days *(Yes, %)*	2 (50.00%)	16 (38.10%)	4 (57.14%)	22 (41.51%)
Delivery Complication *(Yes, %)*	0 (0%)	17 (40.48%)	2 (28.57%)	19 (35.85%)

Demographic Information. When children were 24 months of age or older, demographic information was collected from the full sample including the child's gestational age, gender and the number of siblings the child had. The average gestational age of birth was 39.66 weeks (*SD* = 1.75) in the NE group and 39.74 weeks (*SD* = 1.03) in the No-NE group; no difference in gestational age was observed between groups (t = 0.21, p = 0.84). Of the sample of 62 children followed up at 24 months of age, 34 were male (54.84%) with 18 (58.06%) in the NE group and 16 (51.61%) in the No-NE group. Twenty-eight participants in the cohort were female (45.16%), 13 (41.94%) were in the NE group and 15 females (48.39%) in the No-NE group. There was no statistical difference in the ratio between male and females taking part in the study (X^2 = 0.26, p = 0.61). The majority of the sample had no siblings (N = 23) and 16 children had one sibling; there was no difference in the number of siblings between groups (X^2 = 0.52, p = 0.82).

When the children were assessed at 48 months of age, demographic information collected from the sample included the child's gestational age, gender, ethnicity, the age of the mother and the highest level of education completed by the mother. The average gestational age of birth was 39.98 weeks (*SD* = 2.01) in the NE group and 39.32 weeks (*SD* = 2.10) in the No-NE group; no difference in gestational age was observed between groups (t = -0.95, p = 0.35). Of the sample of 36 children followed up at 48 months of age, 20 were male (55.56%) with 11 (61.11%) in the NE group and 9 (50 %) in the No-NE group. Sixteen participants in the cohort were female (44.44%), 7 (38.89%) were in the NE group and 9 females (50 %) in the No-NE group took part. There was no statistical difference in the ratio between male and females assessed at 48 months (X^2 = 0.45, p = 0.50). All participating families were White apart from one family in the NE group who was of African decent. The mothers taking part in this study were aged

between 24 and 45 years (M = 35.96, SD = 5.79) and all had completed second-level education. There were no statistical differences in the highest level of education the mother received (X^2 = 8.98, p = 0.11), half of the mothers in the sample had completed an undergraduate degree (N = 18, 50%).

Procedure

When the children were 2 years of age or older, the child and their parent were invited to the Infant and Child Research Laboratory at Trinity College Dublin, Ireland for an observation of parent-child interaction and a developmental assessment. Prior to attending the appointment parents were given a parent information leaflet outlining details of the research. If they agreed to take part in the study parents were asked to complete an online questionnaire. The survey pack included a series of questionnaires used to measure demographics and behaviours of the child. During their visit to the laboratory, the parent was given a form to confirm their consent to participate in the study. The children and their parents then took part in an episode of parent-child interaction which was observed and recorded. Mothers and their children aged 2 to 3 years took part in two 5-minute episodes of parent-child interaction where they engaged in free play and structured play, respectively. Mothers and children who were 4 years of age or older took part in 5-minutes of structured play. Upon completing the parent-child interaction, age appropriate developmental assessments were conducted with the children. If the child was 2 to 3 years of age the child completed the third edition of the Bayley (Bayley, 2006) assessment. If the child was 4 years of age or older the WPPSI-IV assessment (Wechsler, 2013) was administered to assess cognitive development and the Movement-ABC was used to assess motor functioning.

Measures

Bayley Scales of Infant and Toddler Development – Third Edition (Bayley-III)

The third edition of the Bayley Scales of Infant and Toddler Development (Bayley, 2006) was used to assess the developmental outcomes of children in this study. The Bayley-III is a gold standard assessment used to assess the developmental functioning of children aged 1 to 42 months. It takes approximately 90 minutes to administer the assessment. The Bayley-III uses a series of play tasks to assess children's cognitive, language and motor development. The language and motor scales are assessed by two further subdomains. The language scale assesses receptive and expressive language abilities, and the motor scale assesses fine motor and gross motor skills.

The cognitive scale includes items that assess sensorimotor development, object exploration and manipulation, concept formation, memory and other aspects of cognitive processing. Tasks include matching shapes onto a puzzle board, sorting toys by size and colour and providing toys to facilitate imaginative and representative play.

The receptive language scale tests how well the child understands directions, words and grammatical concepts. Tasks used to test receptive language include asking the child to follow two-part directions, asking the child to identify objects or actions from a series of pictures, and identifying items of clothing and body parts on a doll. The expressive language scale assesses how well the child communicates using sounds, gestures and words. Sample items used to test expressive language include asking the child to name a series of pictures, colours and actions from a picture book.

The fine motor scale looks at how well the child can use their fingers and hands to manipulate and use objects. Test items include building a tower with small blocks, stringing three blocks with holes onto a string and using scissors. The gross motor scale

assesses how well the child can use their body to move. Test items used to measure gross motor abilities include asking the child to walk up and down stairs, kick a ball and jump off a step.

A composite score is calculated for the cognitive, language and motor scales of the Bayley-III. Scaled scores are available for the subdomains of the language and motor scales. The scores are based on a comparison of the child to a normative age-matched sample and the composite scores for each domain of the Bayley-III are interpreted as follows:

- 100 (SD = 15) Standardized mean score 50th percentile: mid average functioning.
- <85(1 SD below the mean) 16th percentile: indicates mild impairment/ at 'risk' of developmental delay – needs to be monitored/advice to parents on techniques to improve development/referral to therapist depending on degree of impairment.
- <70 = (2 SD below the mean) 2nd percentile: indicates moderate/severe impairment requiring early intervention team input.

Figure 2. Interpretation of Bayley-III Scores (Del Rosario et al., 2020).

Validation of the Bayley-III scale is based on test scores from 1,700 children from the US and 221 children from the UK and Ireland, taking into account age, gender, ethnicity, parental education and geographic region. Concurrent validity of the tool has been established by comparing each domain of the Bayley-III to domain specific assessments. The Bayley-III motor scale and Peabody Developmental Motor Scales (Folio, 2000) were positively correlated with 79.17% total agreement of scores. The language and cognitive scales correlate positively with verbal IQ (0.71-0.83) and full-scale IQ (0.72-0.79) on the WPPSI-III (Weschler, 2002).

Weschler Preschool and Primary Scale of Intelligence – Fourth Edition (WPPSI-IV)

The fourth edition of the WPPSI (Weschler, 2012) was used to assess the developmental functioning of older children in this study. The WPPSI-IV is an intelligence test developed for children aged 2.2 to 7.7 years of age. In this study the WPPSI-IV was administered when the children were 4 years of age and older. The assessment takes approximately 60 to 90 minutes to administer, depending on the age and ability of the child. A series of tasks from the WPPSI-IV was administered to assess domains of children's cognitive and language development including verbal comprehension, visual spatial skills, fluid reasoning, working memory, processing speed and full-scale IQ.

The verbal comprehension scale assesses the child's verbal reasoning skills, concept formation and general knowledge. Sample items used to assess verbal comprehension include asking the child general knowledge questions such as "What do people use to stay dry in the rain?" and asking the child to identify similarities between common objects and concepts such as "Juice and milk are both?". The visual spatial index measures the child's processing skills by organising visual information, attending to visual detail and integrating visual and motor functions. Test items include asking the child to assemble puzzle pieces and recreate designs using coloured blocks. The fluid reasoning scale assesses the child's inductive reasoning skills, visual intelligence, simultaneous thinking, conceptual thinking, and classification ability. Fluid reasoning is assessed by asking the child to select the correct picture missing from incomplete patterns, and to group pictures by a particular common trait. The working memory scale tests the child's concentration skills and their ability to resist interference from previously memorised items. Sample items include asking the child to recall a series of pictures and locations of animal's homes. The processing speed index scale assesses the child's ability

to quickly and correctly scan and discriminate simple visual information. The child is asked to mark pictures in a search group that match the target picture and to mark pictures which belong to a particular category.

A full-scale IQ is calculated from scores on six of the ten subtests administered. The full-scale IQ score represents the child's intellectual ability and provides a broad representation of cognitive functioning. Composite and IQ scores have a mean of 100 and a standard deviation of 15. Scores are interpreted as follows:

Score	Interpretation
Below 70	Extremely Low
70-79	Borderline
80-89	Low Average
90-109	Average
110-119	High Average
120-129	Superior
130+	Very Superior

Figure 3. Interpretation of WPPSI scores (Weschler, 2012).

Normative data were collected from 1,700 children in the US and 805 children in the UK and Ireland to validate the psychometric properties of the scale. The test-retest stability of the WPPSI-IV has been examined by administering the WPPSI-IV twice, with a mean test-retest interval of 23 days. Good test-retest reliabilities have been demonstrated for the composite scores with Pearson correlation coefficients ranging from $r = 0.84$ to 0.89. Excellent correlation is reported for the full-scale IQ scores ($r = 0.93$). Inter-rater reliability of the WPPSI-IV has been established whereby two independent researchers scored subtests of the WPPSI-IV. High inter-rater agreement was demonstrated with intra-class correlations ranging from $r = 0.98$ to 0.99 (Syeda & Clime, 2014).

Movement Assessment Battery for Children – Second Edition (Movement-ABC)

The second edition of the Movement-ABC (Henderson et al., 2007) was used to assess the motor development of older children born with NE taking part in this study. The Movement-ABC is an assessment that is used to identify delay or impairment in motor development in children aged 3 to 16 years. It takes approximately 20 to 40 minutes to administer the assessment. The Movement-ABC comprises a series of tasks that assess domains of fine motor and gross motor functioning such as manual dexterity, aiming and catching and balance.

The manual dexterity scale includes items that assess how well the child uses their hands. The Movement-ABC tests the speed and sureness of movement of each hand, the coordination of the two hands for the performance of a single task and the hand-eye coordination used when in control of a pen. Sample items used to assess manual dexterity include posting coins into a coin box using their preferred and non-preferred hands, threading blocks with holes onto a lace and completing a drawing trail where the child must draw a continuous line without drawing over a boundary.

The aiming and catching scale tests how well the child can aim and catch objects. The scale assesses the accuracy of retrieving a beanbag when thrown towards them, and the accuracy of aiming the beanbag at a target. Tasks in the aiming and catching scale include catching a beanbag being thrown from a distance and throwing the beanbag onto a mat at a distance of 1.8m. The balance scale measures the child's static and dynamic balance. Static balance refers to the ability to maintain the body's centre of gravity whilst standing.

Dynamic balance involves maintaining balance when the centre of gravity and base of the support are moving, it involves transporting the body from one place to another. Sample items include asking to child to balance on one leg and the balance on each leg is

observed. The child is asked to walk along a line on their tip toes, and to jump onto five mats each laid next to one another. A total test score is calculated from the eight subtests administered. A traffic light system is used to interpret the child's total test score, as follows:

Child's Score	Total Test Score	Percentile Range	Description
Red Zone	Up to and including 56	At or below 5th percentile	Significant movement difficulty
Amber Zone	Between 57 and 67	Between the 5th and 15th percentile	'At-risk' of having a movement difficulty
Green Zone	Any score above 67	Above the 15th percentile	No movement difficulty

Figure 4. Interpretation of Movement-ABC scores (Henderson et al., 2007).

The Movement-ABC is used to identify movement difficulties in children by comparing the child's score to the normative data. Normative data were collected from 1172 children from the UK. Inter-rater reliability of the Movement-ABC has been established. Videotaped performances of the Movement-ABC were scored by 131 paediatric physiotherapists; very high inter-rater agreement was demonstrated with kappa coefficients ranging from 0.95 to 1.00 (Smits-Engelsman et al., 2008).

Parent-Child Interaction

Two types of play interaction were initiated and dyadic mother-child interactions were observed and recorded including structured play and free play. Structured play interactions were used to compare the quantity and quality of JA, CDS and child speech between the NE and no-NE groups. Characterisitcs of JA were also analysed during the

free play interactions and their association with the developmental outcomes of children born with NE was observed.

To initiate free play interactions a box of toys was provided to the child and parent. The box included a number of toys such as a ball, building blocks, a Mr Potato Head set, toy vehicles and animals. The investigator told the parent to play with the toys as they would at home and the investigator left the room and recorded the interaction for 5-minutes. Once the free play interaction had taken place, the investigator took the box of toys away and provided a bag that included the stimuli for the structured play interaction. The bag contained a magnetic construction set and the investigator left the room to allow the mother and child to play. The investigator recorded the interaction for 5-minutes.

Each interaction was video recorded using two Axis Q6035 PTZ Dome Network Cameras which were mounted to the walls in the testing room of the Infant and Child Research Laboratory. The cameras recorded the parent-child interactions with 1080 pixel resolution, 30 fps frame rate and 20x optical zoom. The cameras were connected to a desktop computer in an adjacent observation room and Mangold VideoSync Pro 1.5 was used to record the interactions. High quality audio recordings of each parent-child interaction were captured using a Zoom H2n Handy Recorder. The recording device was concealed in the corner of the room out of the sight of the parent and child.

Chapter Four

The Neurodevelopmental Profile of a group of Children following NE

Introduction

Advances in neonatal intensive care and the administration of TH has improved survival rates of children born with NE (Natarajan et al., 2017; Shankaran et al., 2012). However, infants diagnosed with NE are still at an increased risk for adverse clinical outcomes. Similarly, the developmental outcomes of children born with NE are variable with some exhibiting developmental delay (Pappas et al., 2015; Perez et al., 2013; Robertson & Finer, 1993) and others performing as well as typically developing children (Azzopardi et al., 2014). NE is a heterogeneous disorder and a number of problems that occur during the antenatal and intrapartum periods have been linked to long-term neurodevelopmental delay (Ahearne et al., 2017; Glass et al., 2018; Jenster et al., 2014; Polat et al., 2013).

Neonatal Risk

Clinical problems that are linked with adverse developmental outcomes include an abnormal MRI result (Bhroin et al., 2022; Van Kooij et al., 2010), abnormal EEG readings (Glass et al., 2018; Murray et al., 2016), low Apgar scores (Razaz et al., 2016), metabolic acidosis (measured using cord blood; Ahearne et al., 2017), feeding problems (Martinez-Biarge et al., 2012) and maternal infection (Jenster et al., 2014). Previous studies have created neonatal risk indices in other clinical groups such as those born preterm (Egotubov et al., 2020), those with a low birthweight (Poehlmann & Fiese, 2001) and in those diagnosed with autism (Stein et al., 2006). Neonatal risk indices take into account adverse health outcomes and complications that may occur as a symptom of the clinical disorder and have been used to predict future child development. The level of neonatal risk has been found to be associated with the extent of developmental delay,

with studies generally finding negative relations between neonatal risk scores and child developmental outcomes (Shuster et al., 2023; Stein et al., 2006). A number of clinical complications caused by NE have been linked to neurodevelopmental delay.

However, in a majority of cases the occurrence of a singular complication does not fully account for developmental difficulties. A combination of risk factors is more likely to explain the extent of neurodevelopmental delay in this clinical group. To date, no studies have applied a neonatal risk index for those born with NE. Using a neonatal risk index will allow us to further investigate the developmental profile of children born with NE and the link between neonatal risk factors and child developmental outcomes.

Autism and NE

Autism spectrum disorder is a neurodevelopmental condition that has been found to impact children's social-emotional abilities, communication skills and behaviour (APA, 2013; Lord et al., 2018). Approximately 1% of children globally are diagnosed with autism with prevalence rising in specific clinical and sociodemographic groups (Agrawal et al., 2018; Zeidan et al., 2022). Certain clinical risk factors that can occur during the neonatal period such as fetal distress, birth injury or trauma, low Apgar score, feeding problems and umbilical-cord complications have been found to be associated with autism (Gardner et al., 2011). These neonatal risk factors also commonly occur in those born with NE. A population based study identified a strong association between NE and a diagnosis of autism (Badawi et al., 2006). The rates of autism in a group of children born with NE was compared to a typically developing group born without complication. It was found that children born with NE were 5.9 times more likely to receive a diagnosis of autism compared to the control group. Similar to those born with NE, higher rates of developmental delay are observed in those with a diagnosis of autism (Maenner et al., 2021). Delays in language (Reindal et al., 2021), motor (Liu & Breslin, 2013) and

cognition (Maenner et al., 2021) are commonly reported. Despite the elevated risk of autism and developmental delay in those born with NE, the link between autism symptoms and children's developmental abilities in this group has yet to be investigated.

Executive Functions

Executive functioning (EF) is an umbrella term that describes a set of cognitive processes that enables children to plan, reason and problem solve. EF skills help children regulate their actions to facilitate the attainment of self-directed goals. Abilities involved in EF include attentional control, cognitive flexibility, cognitive inhibition, working memory and information processing. These higher order cognitive skills play an important role in behavioural control, regulating emotions and making decisions (Diamond, 2013). A number of studies have found children born with NE to exhibit EF impairments (Azzopardi et al., 2014; Hayes et al., 2018; Lindström et al., 2006; Marlow et al., 2005). Children born with NE have been found to experience a range of cognitive and behavioural challenges that are potentially related to EF abilities. Poorer memory abilities (Annick et al., 2018; Van Handel et al., 2012), attention (Tonks et al., 2019) and behavioural problems (Lindström et al., 2006) are commonly reported in those born with NE and may impact children's EF. Research on typically developing children suggests that EF is critical for children's cognitive and emotional development (Bernier et al., 2010). Although impaired EF has been identified in those born with NE, research has not yet determined the impact of EF on children's developmental abilities in this clinical group.

Sleep Difficulties

Epidemiological studies have found the prevalence of sleep difficulties in school aged children to vary between 10 to 40% (Blunden et al., 2004; Williamson et al., 2019). NE has been found to have a negative impact on cognitive, motor, sensory and

behavioural outcomes (De Vries et al., 2010) and may further exacerbate the presence of sleep disturbance in this clinical cohort. Zareen and colleagues (2021) found school aged children born with NE to have higher rates of sleep pathology when compared to a control group born without complications. Over half of a sample of children born with NE were classified as having a sleep disorder. Sleep anxiety, bedtime resistance and sleep onset delay were the most common sleep problems observed in the NE sample. Sleep problems in children have been linked to a number of negative outcomes such as reduced quality of life (Zareen et al., 2021), adverse physical and psychological health (Chaput et al., 2016), and behavioural and social-emotional problems (Gregory & O'Connor, 2002). A study on typically developing children found disrupted sleep to have a negative impact on children's cognitive abilities (Schlieber & Han, 2017). Despite the presence of sleep disorders and developmental delay in children born with NE, the association between sleep difficulties and children's developmental abilities in this clinical cohort has yet to be investigated.

Language and Motor Development

Language and motor development in children is closely linked; it has been found that motor abilities facilitate the acquisition of language skills. A clear relationship between language and motor domains has already been established in the child development literature (Iverson, 2010; Valla et al., 2020; see literature review in chapter one). The development of motor skills allows children to explore their surroundings and encounter new opportunities for learning (Hofsten, 2009). In the case of children born with NE, those with better motor abilities may have more opportunities for social interaction and involvement, which in turn provides them with the opportunity to practice their language skills. Due to the impact of NE on children's motor development (Jary et

al., 2019; Van Kooij et al., 2008) and language (Chin et al., 2019) investigating the relationship between the two developmental domains in those born with NE is warranted.

Current Study

The aim of this study is to characterise the developmental profile of children born with NE using standardised assessments of development. This cross-sectional study measures and compares the cognitive, language and motor abilities of children born with NE with the abilities of typically developing children born without complications using the Bayley-III or the WPPSI-IV assessment. Factors that may further impact the developmental functioning of children born with NE such as neonatal risk, autism symptoms, executive functioning skills and sleep problems are also measured. Due to the influence of motor abilities on the development of language skills, the relationship between language and motor outcomes in children born with NE is also analysed. The following hypotheses were investigated:

Hypothesis 1. Children born with NE will score significantly lower than a typically developing comparison group born without complications on standardised measures of cognitive and language ability.

Hypothesis 2. Neonatal risk scores will be negatively associated with children's developmental assessment scores. Children born with NE who have higher neonatal risk scores are expected to have lower developmental outcome scores. It is also expected that children born with NE who display a higher number of autism symptoms, executive functioning deficits and sleep problems will receive below average scores on developmental assessments of cognitive, language and motor skills.

Hypothesis 3. As is the case for typically developing children, it is expected that children's language abilities will be positively associated with motor ability scores in children born with NE.

Method

Participants

Ninety-eight children aged 24 to 66 months participated in the study: Forty-nine children born with NE and 49 typically developing children born without complications were matched by age using propensity score matching; a detailed description of this study sample is outlined in chapter three. The developmental assessment completed by the children depended on the age of the child at the time of assessment. Overall, 62 children aged 24 to 42 months completed the Bayley-III assessment (31 children born with NE) and 36 children aged 42 to 66 months of age completed the WPSSI-IV assessment (18 children born with NE). When assessing executive functioning abilities of children, 86 parents completed a parent report questionnaire, of whom 33 were parents of children born with NE and 53 were parents of typically developing children.

Procedure

Infants born with NE were originally recruited at the time of the child's birth from participating maternity hospitals in Dublin, Ireland. Mothers of infants diagnosed with NE were asked to take part in the research study. The medical records of those who consented to participate were made available and were used to calculate neonatal risk scores. When children in the NE group were 24 months or older, families were re-contacted for follow-up. Prior to attending a follow-up appointment, parents of children born with NE were given a questionnaire pack and were asked to complete a series of parent report questionnaires that measured autism symptoms, executive functioning skills and sleep difficulties related to their children. The children and their parents were then invited to the Infant and Child Research Lab to complete a developmental assessment. Children aged 24 to 42 months were administered the Bayley-III, and children aged 42 months and older completed the WPPSI-IV. A typically developing group of children

born without complications was also assessed using standardised measures of development and a parent report questionnaire on child executive functioning.

Measures

Bayley Scales of Infant and Toddler Development – Third Edition (Bayley-III; Bayley, 2006). To assess children's developmental abilities, children aged 24 to 42 months of age were administered the Bayley-III (Bayley, 2006) by a trained researcher. The cognitive and language scales were completed by children born with NE and typically developing children born without complications. Those born with NE also completed the motor scale of the assessment. All methods and procedures used to assess development using the Bayley-III are detailed in chapter three.

Wechsler Preschool and Primary Scale of Intelligence – Fourth Edition (WPPSI-IV; Wechsler, 2013). Children born with NE and typically developing children born without complications were administered the WPPSI-IV (Wechsler, 2013) if the child was aged 42 months or older. Ten tasks from the WPPSI-IV were administered to assess six domains of cognitive and language functioning including: verbal comprehension, visual spatial skills, fluid reasoning, working memory, processing speed and full-scale IQ. All procedures for the administration of the WPPSI-IV are detailed in chapter three.

Movement Assessment Battery for Children – Second Edition (Movement-ABC; Henderson et al., 2007). The motor abilities of children born with NE were assessed using the Movement-ABC (Henderson et al., 2007) by a trained researcher when children were aged 42 months or older. Three subdomains of motor functioning were assessed including manual dexterity, aiming and catching and balance. An overall score of motor abilities was also calculated. All methods and procedures for the administration of the Movement-ABC are outlined in chapter three.

Neonatal Risk Index. The medical records of children born with NE were examined and a neonatal risk index score was calculated for each participant born with NE. Neonatal risk is measured on a scale ranging from 0 to 9 points, with higher scores indicating higher levels of neonatal risk. One point is given based on the presence of each of the following risk factors: an abnormal MRI result, the presence of seizures, a 5 minute Apgar score less than 7, a cord blood pH less than 7.2, whether the infant was intubated, lack of suck and feeding problems, the presence of maternal infection and if the infant was hospitalised for more than ten days. Another point was given if delivery complications arose such as fetal bradycardia, meconium aspiration, maternal haemorrhage, placental abruption or nuchal cord complications. Descriptive statistics of the neonatal risk index is outlined in chapter three.

Behaviour Rating Inventory of Executive Function – Preschool Version (BRIEF-P; Gioia et al., 2003). Executive functioning of children was measured using the BRIEF-P parent-report questionnaire (Gioia et al., 2003). The questionnaire consists of 63 statements with a reference period of the past 6 months. The statements relate to five domains of executive functioning including inhibitory control, shifting, emotional control, working memory, and planning and organising. Each scale is combined to form index scores which measure control, flexibility, emergent metacognition and a global executive composite (GEC). Examples of statements include “Gets caught up in the small details of a task or situation and misses the main idea” and “Has trouble getting started on activities or tasks even after instructed”. The parent is asked to rate each statement using a three-point scale ranging from “Never”, “Sometimes” or “Often”. T-scores are calculated for each domain of the BRIEF-P, a T-score of 65 represents a score that is 1.5 standard deviations from the mean and is interpreted as ‘abnormally elevated’.

In order to calculate the T-scores, the scores of the participant are compared to those in the normative sample established by the authors of the scale. Normative data were collected from 460 parents of children aged 2 to 5 years and took into account gender, socioeconomic status, ethnicity and geographical location. Overall, the BRIEF-P has been found to demonstrate acceptable to excellent reliability and yielded test-retest correlations between 0.78 to 0.90 (Gioia et al., 2003).

Autism Screening Questionnaires. Due to the elevated risk of autism in children born with NE, autism symptoms were measured in the NE group using one of two autism screening questionnaires:

The Modified Checklist for Autism in Toddlers, Revised – (M-CHAT-R; Robins et al., 2009). The M-CHAT-R (Robins et al., 2009) was used to measure traits of autism in children born with NE at 24 months of age. The M-CHAT-R is a parent-report screening tool devised for children aged 16-30 months. The questionnaire comprises 20 yes-no questions that relate to traits of autism and is completed by the parent about their child's behaviour. Examples of questions include "When you smile at your child, does he or she smile back?" and "If you turn your head to look at something, does your child look around to see what you are looking at?". A total score of 0 to 2 indicates that the child is at low-risk of having autism. A score of 3 to 7 signifies that the child has a medium-risk of having autism. A score of 8 to 20 implies that the child is at high-risk of having autism. The M-CHAT-R is a widely used autism screening instrument that demonstrates adequate sensitivity: 47% of children classified at medium and high-risk of autism on the M-CHAT-R went on to receive a formal diagnosis of autism (Robins et al., 2014).

Autism Behaviour Checklist – (ABC; (Krug et al., 2008). The ABC (Krug et al., 2008) was used to measure symptoms of autism in children at 36 months of age or older. The ABC is a parent-report questionnaire devised for children aged 3 to 14 years. The

questionnaire consists of 57 statements that relate to five traits of autism including sensory behaviours, relating, body and object use, language and social and self-help behaviours. Sample statements include “Echoes questions or statements made by other people” and “Gets involved in complicated rituals such as lining things up”. The parent is asked to select the items that most accurately describe the child; each statement includes a point score ranging from 1 to 4 if the parent identifies the statement to be true. Scores are calculated for each domain of the ABC and are combined to produce a total score. It is proposed that children with overall scores equal to or greater than 68 are considered at high risk for autism. Scores between 54 and 67 represent a moderate probability of autism and scores between 47 and 53 are considered as inconclusive. If the child receives a score below 47 the child is at low-risk of having autism. Normative data were collected from 1049 families to validate the psychometric properties of the scale. The ABC total score has demonstrated adequate reliability to be used as a screening instrument for autism, reporting a coefficient alpha of 0.87 in a test of internal consistency (Eaves & Williams, 2006).

Tayside Sleep Questionnaire – (McGreavey et al., 2005). High rates of sleep disturbance have been identified in school-aged children born with mild to moderate NE (Zareen et al., 2021). Therefore, the presence of sleep problems in the sample of children born with NE was measured using the Tayside Sleep Questionnaire (McGreavey et al., 2005). The Tayside Sleep Questionnaire is a parent-report questionnaire that was developed to detect sleep disturbance in children aged 1 to 5 years. The questionnaire consists of 10 statements concerning the child’s sleep pattern over the previous three-month period. Sample statements include “the child does not fall asleep in his or her own bed” and “the child wakes up two or more times in the night”. Each statement is ranked on a 5-point rating scale ranging from 0 to 4 (0 = the sleep behaviour never occurs; 1 =

the problem occurs once or twice a month; 2 = occurs one or two times a week; 3 = occurs between three and five nights a week and 4 = the sleep problem happens every night). Children with a score of 8 or above are classified as having mild to moderate sleep problems. The validation of the Tayside Sleep Questionnaire is based upon normative data collected from 628 children ranging from 12 to 66 months of age (mean age of 41.7 months). The validity of the scale was measured using qualitative methods, with all but one parent agreeing with the outcomes of the scale. Reliability was established for the scale and good internal consistency has been reported, yielding a Cronbach alpha coefficient of 0.85 (McGreavey et al., 2005).

Statistical Analysis

Statistical analyses for this study were conducted using SPSS (Version 26, IBM). Data were checked for normality to meet the assumptions of parametric statistics using QQ plots and the Shapiro Wilk's test. Homogeneity of variance was checked using Levene's test. The developmental scores of children born with NE were compared to those of typically developing children using independent sample t-tests. A Bonferroni correction was applied to analyses with multiple comparisons. Pearson's correlation coefficients were used to observe the relationship between neonatal risk and developmental assessment scores. The relationship between language and motor outcomes in children born with NE was also calculated using correlation coefficients. The proportion of children born with NE in this study who displayed autism symptoms, executive functioning difficulties, sleep problems and below average scores on assessments is also reported. Although multiple comparisons were conducted, it was considered that a Bonferroni correction would result in an overly stringent alpha level leading to a risk of Type II error (Armstrong, 2014). In the interest of this exploratory

analysis, a Bonferroni correction was not applied to the correlation analyses in order to identify relevant associations between the variables.

Results

Comparison of Developmental Outcomes in NE and No-NE group

Independent sample t-tests were used to analyse the scores of a group of children born with NE and typically developing children born without complications (no-NE group) on developmental outcomes. Children born with NE scored significantly lower than the No-NE group on each domain of language including receptive language, expressive language and total language composite score. There were no significant group differences on the cognitive domain (see table 4.1).

Table 4.1. *Developmental scores assessed using the Bayley-III in the NE and No-NE groups.*

Variable	**NE** (*N= 31*)	**No-NE** (*N= 31*)		
	M (SD)	*M* (SD)	***Cohen's d***	**t, *p-value***
Cog Comp	100.97 (10.36)	104.68 (15.05)	0.29	t = 1.13, *p* = 0.26
Lang Comp	95.39 (18.50)	112.55 (16.16)	0.99	t = 3.89, *p* = 0.00**
Rec Lang Scale	9.35 (3.19)	12.45 (3.09)	0.99	t = 3.89, p = 0.00**
Exp Lang Scaled	9.03 (3.39)	11.71 (3.11)	0.83	t = 3.24, p = 0.00**

Note. Cog Comp = Cognitive Composite Score; Lang Comp = Language Composite Score; Rec Lang Scale = Receptive Language Scaled Score; Exp Lang Scaled = Expressive Language Scaled Score.

**$p < 0.01$; significance level adjusted to ≤ 0.01 to correct for multiple analyses.

An independent samples t-test was used to compare the performance on each subscale of the WPPSI-IV between the two groups. Children born with NE performed

significantly worse on the verbal comprehension scale. Differences were observed on processing speed index scores; however, this significant difference diminished following a Bonferroni correction to account for multiple comparisons. There were no significant group differences on any other domain of the WPPSI-IV including visual spatial skills, fluid reasoning, working memory or full-scale IQ (see table 4.2).

Table 4.2. *Developmental outcome scores assessed using the WPPSI-IV in the NE and No-NE groups.*

Variable	**NE** (*N= 18*) *M* (SD)	**No-NE** (*N= 18*) *M* (SD)	***Cohen's d***	**t, *p-value***
VCI	94.72 (23.54)	117.44 (12.94)	1.20	t = 3.59, *p* = 0.00**
VSI	107.72 (24.35)	102.39 (16.46)	0.26	t = -0.77, *p* = 0.45
FRI	106.83 (23.26)	109.56 (14.73)	0.14	t = 0.42, *p* = 0.68
WMI	101.83 (23.81)	107.33 (13.32)	0.29	t = 0.86, *p* = 0.40
PSI	92.83 (20.92)	107.00 (12.38)	0.82	t = 2.47, *p* = 0.02
Full Scale IQ	104.50 (25.89)	109.38 (14.34)	0.23	t = 0.70, *p* = 0.49

Note. VCI = Verbal Comprehension Index; VSI = Visual Spatial Index; FRI = Fluid Reasoning Index; WMI = Working Memory Index; PSI = Processing Speed Index.

**$p < 0.01$; significance level adjusted to ≤ 0.01 to correct for multiple analyses.

An independent samples t-test was conducted to examine group differences in domains of executive functioning in the NE and No-NE group. The NE group scored significantly higher on the Emotional control scale of the BRIEF-P questionnaire, with higher scores indicating greater dysfunction. Differences were also observed on the Inhibit, Working Memory, Plan/Organise and General Executive Composite scales but they were no longer significant following a Bonferroni correction for multiple

comparisons. No difference was observed between the two groups on the Shift scale (see table 4.3).

Table 4.3. *Comparison of executive functioning scores using the BRIEF-P in the NE and No-NE groups.*

Variable	**NE** (N=33)	**No-NE** (N=53)		
	M (SD)	*M* (SD)	**Cohen's d**	**t, *p-value***
Inhibit	52.48 (12.12)	47.11 (9.07)	0.50	t = -2.36, *p* = 0.02
Shift	50.06 (12.34)	46.53 (9.05)	0.33	t = -1.53, *p* = 0.13
Emotional Control	52.15 (12.56)	45.81 (8.14)	0.60	t = -2.86, *p* = 0.01**
Working Memory	55.69 (14.72)	49.81 (11.47)	0.45	t = -2.08, *p* = 0.04
Plan/Organise	53.21 (14.25)	47.00 (10.77)	0.49	t = -2.31, *p* = 0.02
GEC	53.58 (14.42)	46.85 (10.29)	0.54	t = -2.53, *p* = 0.04

Note. GEC = General Executive Composite.

***p* ≤ 0.01; significance level adjusted to ≤ 0.01 to correct for multiple analyses.

Relationship Between Neonatal Risk and Developmental Outcome Scores

Correlation coefficients were conducted to analyse the association between neonatal risk scores and developmental outcomes scores of 2 and 3-year-old children born with NE. A significant relationship between expressive language and neonatal risk was found, indicating that children with higher neonatal risk scores performed worse on the assessment of expressive language abilities. No other significant associations were found between neonatal risk and the other domains of development (Table 4.4).

Table 4.4. *Relationship between neonatal risk and Bayley-III scores in NE group (N=25).*

	Neonatal Risk Score
Cognitive Composite Score	-.30
Receptive Language Scaled Score	-.11
Expressive Language Scaled Score	-.49*
Language Composite Score	-.30
Fine Motor Scaled Score	.03
Gross Motor Scaled Score	-.01
Motor Composite Score	.00

*$p \leq 0.05$

Pearson correlation coefficients were computed to analyse the relationship between neonatal risk and developmental outcome scores in 4-year-old children born with NE. Neonatal risk scores were negatively associated with fluid reasoning, working memory and processing speed scores on the WPPSI-IV. No associations were observed between neonatal risk and verbal comprehension, visual spatial abilities and full-scale IQ (Table 4.5).

Table 4.5. *Relationship between neonatal risk and WPPSI-IV scores in NE group (N=15).*

	Neonatal Risk Score
Verbal Comprehension Index	-.34
Visual Spatial Index	-.24
Fluid Reasoning Index	-.54*
Working Memory Index	-.65**
Processing Speed Index	-.60*
Full Scale IQ	-.44

*$p \leq 0.05$; **$p \leq 0.01$.

The Developmental Profile of Children Born with NE

Within group analyses were conducted to characterise the developmental profile of children born with NE. The proportion of children receiving below average scores on each domain of the Bayley-III (receiving a score <85 indicates 'risk' of developmental delay, a score <70 indicates moderate to severe impairment) was computed. Of the 44 children with NE assessed using the Bayley-III, two children received scores indicating a risk of cognitive delay, 11 children were at-risk of language delay, while one child exhibited moderate to severely delayed language functioning. Four children were at-risk of motor delay and one child had moderate to severe motor problems (see table 4.6). When observing developmental outcome scores on each subdomain of the Bayley-III, 9 children on the receptive language scale, 11 children on the expressive language scale, 3 children on the fine motor scale and 6 children on the gross motor scale received a below average score <7 (see table 4.7).

Table 4.6. *Proportion of children in the NE group with below average composite scores on the Bayley-III.*

N=44	Mean Composite Score	Below Average Composite Scores	
	Mean (SD)	N (%) <85	N (%) <70
Cognitive Scale	100.11 (9.24)	2 (4.55%)	0 (0.00%)
Language Scale	97.59 (17.66)	11 (25.00%)	1 (2.27%)
Motor Scale	98.47 (12.16)	4 (9.09%)	1 (2.27%)

Table 4.7*. Proportion of children in the NE group with below average scaled scores on the Bayley-III.*

N=44	Mean Scaled Score	Below Average Subscale Scores	
	Mean (SD)	N (%) <7	N (%) <4
Receptive Language	9.78 (3.33)	9 (20.45%)	0 (0.00%)
Expressive Language	9.39 (3.03)	11 (25.00%)	0 (0.00%)
Fine Motor	10.34 (2.09)	3 (6.82%)	0 (0.00%)
Gross Motor	9.00 (2.56)	6 (13.64%)	0 (0.00%)

Of the 19 children in the NE group who were assessed using the WPPSI-IV, 2 children received scores <70 on all domains indicating developmental delay. An additional participant received a composite score <70 on the processing speed scale. Three participants received a borderline score on the verbal comprehension scale and two on the processing speed scale by scoring <80 on each domain. One participant had a full-scale IQ <80 indicating a risk of developmental delay (see table 4.8). The proportion of

children receiving below average scores on the Movement-ABC was also calculated. Of the 18 children assessed using the Movement-ABC, 5 children (27.78%) received a score less than 67, indicating risk of having a movement difficulty. The mean total test score on the assessment was 74.50 (SD = 23.17).

Table 4.8. *Proportion of children in the NE group with below average scores on the WPPSI-IV.*

N=19	Mean Composite Score	Below Average Composite Scores	
	Mean (SD)	N (%) <80	N (%) <70
Verbal Comprehension	93.68 (23.32)	3 (15.79%)	2 (10.53%)
Visual Spatial	107.95 (23.69)	0 (0.00%)	2 (10.53%)
Fluid Reasoning	106.42 (22.67)	0 (0.00%)	2 (10.53%)
Working Memory	101.58 (23.16)	0 (0.00%)	2 (10.53%)
Processing Speed	92.12 (20.57)	2 (10.53%)	3 (15.79%)
Full Scale IQ	103.74 (25.38)	1 (5.26%)	2 (10.53%)

The proportion of children born with NE receiving below average scores on each developmental domain and in each age range is reported in table 4.9. Language difficulties were the most commonly reported with 32.08% of the NE group receiving a below average score on a standardised assessment of language. Autism symptoms, EF deficits and sleep problems in the cohort were also measured. In the sample of 53 children born with NE, 11.32% reported a high number of autism symptoms, 15.09% of children had EF deficits and 28.3% had sleep problems.

Table 4.9. *Number of children in NE group at each assessment wave receiving below average scores across various developmental domains.*

	2yrs (*N=23*)	3yrs (*N=10*)	4yrs+ (*N=20*)	Total (*N=53*)
	N	*N*	*N*	*N* (%)
Cognitive Scale	2	0	3	5 (9.43%)
Language Scale	11	1	5	17 (32.08%)
Motor Scale	3	2	5	10 (18.87%)
Autism Symptoms	2	2	2	6 (11.32%)
EF Deficits	1	2	5	8 (15.09%)
Sleep Problems	3	6	6	15 (28.30%)

Note. Cognition, language and motor scores in 2 and 3 year olds was measured using the Bayley-III; cognition and language scores in 4 year olds was measured using the WPPSI-IV; motor scores in 4 year olds was measured using the Movement-ABC; EF = Executive functioning measured based on parent report using the BRIEF-P; Autism symptoms were measured using the M-CHAT-R or ABC; sleep problems were assessed using the Tayside Sleep Questionnaire.

Overall, six participants born with NE exhibited a high number of autism symptoms as measured by parent report questionnaires. Table 4.10 reports the proportion of below average developmental outcome scores received by the six children with a high number of autism symptoms. Of the children who displayed a high number of autism symptoms, one participant received a below average score on the language domain, two participants had below average scores on the language and motor domains and one participant had below average scores on all three domains of development. Two

participants displaying autism symptoms had developmental assessment scores within the average range.

Table 4.10. *Autism symptoms and below average developmental assessment scores in children born with NE (N=6).*

Participant	Cognitive Delay	Language Delay	Motor Delay
1	No	Yes	Yes
2	Yes	Yes	Yes
3	No	Yes	No
4	No	No	No
5	No	Yes	Yes
6	No	No	No
Total %	16.67%	66.67%	50%

In our sample of children born with NE, 8 participants exhibited EF deficits by receiving a score of 65 or below on the BRIEF-P questionnaire. Table 4.11 reports the proportion of below average developmental outcome scores received by these 8 children with EF deficits. Overall, two participants received below average scores only on the language domain, two participants had below average scores on the language and motor domains and a further two participants received below average scores on all three domains of development. Two participants exhibiting EF deficits received developmental assessment scores within the average range.

Table 4.11. *Executive functioning deficits and below average developmental assessment scores in children born with NE (N=8).*

Participant	Cognitive Delay	Language Delay	Motor Delay
1	No	Yes	Yes
2	No	Yes	No
3	Yes	Yes	Yes
4	Yes	Yes	Yes
5	No	No	No
6	No	Yes	No
7	No	Yes	Yes
8	No	No	No
Total %	25 %	75%	50%

Fifteen participants born with NE had sleep difficulties as reported using the Tayside Sleep questionnaire. Despite a higher proportion of NE children exhibiting sleep difficulties fewer participants received below average scores on assessments of development when compared to those with autism symptoms and EF deficits. This suggests that sleep problems may not be associated with poorer assessment scores in this group. Of the 15 participants with sleep problems, one participant received a below average score only on the language domain, another participant had below average scores on the language and motor domains and one participant had below average scores on all three domains of development. Twelve participants with reported sleep difficulties received developmental assessment scores within the average range.

The Association between Language and Motor Outcomes in Children Born with NE

Pearson correlation coefficients were computed to analyse the relationship between language and motor outcomes in children born with NE. Multiple positive significant associations were observed between language and motor composite and scaled scores on the Bayley-III (see table 4.12). Language composite scores correlated highly with motor composite scores, fine motor skills and gross motor skills. Receptive language scores correlated highly with motor outcomes including fine motor and gross motor skills. Expressive language scores correlated with motor composite scores and fine motor abilities but not with gross motor skills. These relationships suggest that children with better language skills have higher scores on the motor scale of the Bayley-III.

Table 4.12. *Relationship between Language and Motor Bayley-III scores in NE Group at 2 years (N=43).*

Bayley-III Domain	Language Composite	Receptive Language	Expressive Language
Motor Composite	.45**	.48**	.37*
Fine Motor	.42**	.48**	.30*
Gross Motor	.32*	.34*	.27

*$p \leq 0.05$; **$p \leq 0.01$

Pearson correlation coefficients were used to assess the relationship between language and motor outcomes in children born with NE at age 4 years. A significant positive association was observed between verbal comprehension index subscale scores of the WPPSI-IV and Movement-ABC scores, indicating that higher scores on the Movement-ABC are associated with higher verbal comprehension ($r = 0.69$, $p = <0.01$).

Discussion

Children born with NE endure a number of risk factors that potentially compromise their development. The current study examined the neurodevelopmental profile of a group of children following NE. The first hypothesis examined the cognitive and language outcomes of children born with NE compared to typically developing children born without complications. When assessed using standardised developmental instruments, it was found that children born with NE had poorer language abilities when compared to typically developing children born without NE. At 2 to 3 years of age, children born with NE scored significantly lower on each language subscale on the Bayley-III as predicted. However, counter to the study's hypothesis, no differences were observed in scores on the cognitive scale. This pattern of findings was replicated in the cohort of 4-year olds assessed using the WPPSI. Children born with NE scored significantly lower on the verbal comprehension and processing speed scale compared to the No NE group, however, no differences were observed on any other subscale relating to cognitive development. Despite cognitive scores similar to those of typically developing children, children born with NE are reported to exhibit a higher degree of executive functioning deficits.

Previous studies have found the developmental outcome scores of children born with NE to be within the normal range when assessed in early childhood, particularly in those born with mild NE and in those without a disability (Azzopardi et al., 2014; Handley-Derry, 1997; Rao et al., 2019; Robertson & Finer, 1985). However, evidence demonstrates that children born with NE are not following the same developmental trajectory as typically developing children (Finder et al., 2019). Despite receiving scores in the normal range, as children born with NE get older, deficits become more apparent. Studies have found that some children born with NE who receive cognitive scores in the

normal range in early childhood go on to exhibit memory, cognitive and EF problems in later life (Annick et al., 2018; Lindström et al., 2006). Research has found that a developmental assessment score from a single time point is not an accurate predictor of children's outcomes at school age (Anderson et al., 2010; Chalak et al., 2014; Hack et al., 2005). It is recommended that children born with NE are followed up into later childhood at multiple time points (Neel et al., 2023). Standardised assessments such as the Bayley-III focus on global development and assess basic cognitive abilities. Such global assessments may not be sensitive enough to identify cognitive difficulties in children born with NE. Some studies have found the Bayley-III to underestimate levels of developmental delay and the proportion of children classified with a disability (Anderson et al., 2010; Green et al., 2023; Jary et al., 2013). Alternatively, issues with cognitive development may not exist at the time of assessment. It might be the case that cognitive delay is not present in NE children at an early age but difficulties develop or become more apparent as they get older. Despite mean scores in the average range on tests of cognition, in the current study children born with NE exhibited more deficits in executive functioning compared to the No-NE group. Standardised tests of development measure cognitive functioning on a global level but they do not consider how cognitive abilities are applied in daily life. The BRIEF-P contains statements that describe how the child's developmental skills are utilised in day-to-day life for example: "Has trouble thinking of a different way to solve a problem or complete an activity when stuck" and "Needs to be told to begin a task even when willing to do it". Although the child is capable of navigating a task, EF skills such as behavioural inhibition, emotional control and planning and organising may impact their ability to complete it. Children who score low on the BRIEF-P may need extra support to complete tasks, and if support is not provided

it may compromise the child's learning, leading to a decline in cognitive abilities in later life.

Parenting style may have also influenced the results of this study. Research has demonstrated specific parenting styles to impact the development of children in clinical groups (Neel et al., 2019). A review of the literature conducted by Neel and colleagues (2018) established parent responsiveness and demandingness to be positively associated with improved cognition in children born preterm. It is possible that parents of the children in this study used higher levels of responsivity and appropriate levels of demandingness, contributing to cognitive scores in the typical range. However, parenting style was not taken into consideration in this analysis. Further research should look to investigate the link between parenting style and the cognitive outcomes of children impacted by NE.

Processing speed is the only other domain of cognition where a difference in performance was observed between groups. Processing speed tasks measure the child's mental speed and cognitive flexibility. These tasks require the recruitment of other cognitive functions such as attention, short-term visual memory, visual discrimination, visuo-motor coordination and concentration. When carrying out tasks that measure processing speed, previous studies have found children born with NE to have slower response times, show inconsistency in their response speed, and reactions times decline during assessment compared to typically developing children (O'Connor et al., 2017; Spencer et al., 2021). Poor performance on processing speed tasks has been attributed to attention problems that are commonly reported in children born with NE (Tonks et al., 2019). Attention problems in children born with NE have been measured using behavioural assessments (Hayes et al., 2018) and this cohort tends to perform worse than typically developing controls on attention tasks (Marlow et al., 2005). Children born with

NE are also more likely to receive a diagnosis of ADHD in later childhood. Symptoms of inattention, impulsiveness and hyperactivity may negatively impact their performance on processing speed tasks (Smith et al., 2016).

As expected, children born with NE in the present study have poorer language outcomes compared with typically developing children. These findings are consistent with the previous literature which found that children born with NE perform in the low to average range on language measures (Chin et al., 2019; Shapiro et al., 2017). To date only one study has reported the use of a language specific assessment. Chin and colleagues (2019) used the parent report MacArthur-Bates Communicative Development Inventories (MB-CDI; Fenson et al., 2007) and found children born with NE to have a smaller vocabulary and use shorter utterances. Future studies could conduct fine grained analyses of the characteristics of child language using natural speech samples to further define the language profile of this clinical group.

Hypothesis two sought to examine the association between neonatal risk caused by NE and children's developmental assessment scores. As hypothesised, the study observed a number of negative associations between neonatal risk scores and developmental assessment scores. It was observed that children born with NE with higher neonatal risk scores scored lower on assessments of expressive language in the cohort of 2 and 3-year-old children, and domains of cognition in the sample of 4-year olds. These findings are in line with the current literature: a previous study found school aged children with specific language impairment to experience two or more neonatal risk factors compared to typically developing children without a diagnosis of language impairment (Whitehouse et al., 2014). In the current study no differences were observed in the number of children with below average cognitive scores in the NE and no-NE groups; however, those who did receive below average scores were more likely to

experience higher levels of neonatal risk. The relationship between cognitive scores and neonatal risk in this sample of 4-year olds born with NE further supports the finding that difficulties in cognitive development might become more apparent in later life. Working memory had the strongest association with neonatal risk in 4-year-olds born with NE. This finding aligns with previous studies which reported that memory problems in particular tend to become more evident as children born with NE get older (Annick et al., 2018; Van Handel et al., 2012; Zorn et al., 2018).

Additionally, this study looked to identify the proportion of children born with NE displaying autism symptoms, EF deficits and sleep problems. The proportion of NE children with below average scores on developmental assessments in these categories is also observed. Overall, 11.32% of this sample of children born with NE (six children) exhibited a high number of autism symptoms (indicating an elevated risk of having autism), with two of these participants receiving a formal diagnosis. In line with the autism literature, four of these children also received below average scores on language assessments. Three of these children also received below average scores on motor assessments. Children diagnosed with autism commonly experience language delay (APA, 2013; Gernsbacher et al., 2016; Reindal et al., 2021). Studies have also found children with autism to be at-risk of motor delay (Liu & Breslin, 2013). Therefore, it is likely that there is also a relationship between high levels of autism symptoms and developmental delay in children born with NE. Due to the elevated risk of autism in those born with NE, it is advised that children in this clinical group should be screened for neurodevelopmental disorders including autism as part of clinical practice. Screening and parent report questionnaires are inexpensive and easy to administer and can bring to attention children who may require extra support. This ensures that children who are at

risk of developmental difficulties receive support as early as possible as many children born with NE do not receive support from services until later in life.

In the current study, 15.09% of children born with NE were reported to exhibit EF difficulties by their parents. The highest proportion of EF deficits were observed among the cohort of 4-year-old children, suggesting that EF difficulties may become more apparent beyond toddlerhood into the preschool years. Of the eight children who were found to display EF deficits, the majority of children received below average scores on assessments of language (six children) and four of the children received below average scores on assessments of motor abilities. Although EF is a skill that largely requires the use of cognitive functions, only two children in the sample with EF deficits received a below average score on assessments of cognition. This suggests that developmental assessments capture particular aspects of cognition and may not necessarily reflect how children are able to apply their knowledge in day-to-day life. A study on teenagers born with NE found that cognitive and executive functioning problems in particular interfered with daily life situations (Lindström et al., 2006). It is possible that children born with NE perform well on tasks that require simple cognitive abilities, such as puzzles and following instructions, but find it difficult to apply these skills to everyday life. This suggests that although developmental delay may not be immediately evident in children born with NE, problems might arise or become more apparent at older ages. Thus, the long-term monitoring of the developmental outcomes of children born with NE is recommended.

In this study the association between sleep problems and below average developmental scores in children born with NE was weak. Only 3 out of 15 children whose parents had reported sleep problems had below average scores on language assessments and 2 out of 15 had below average scores on an assessment of cognition. In a

sample of 1023 typically developing children, the mean score on the Tayside Sleep Questionnaire was 7.4 (SD = 7.2) with 35% of children receiving a score above 8 indicating sleep difficulties (McGreavey et al., 2005). A smaller proportion of children were identified as having sleep problems in this cohort of children born with NE (28.30%); this proportion is consistent with the prevalence rate for sleep difficulties among typically developing children reported in the literature (Blunden et al., 2004; Williamson et al., 2019). It is possible that lower levels of sleep disturbance in this sample of children born with NE is due to the small sample size. Additionally, the no-NE group did not complete the sleep questionnaires, therefore a direct comparison of the presence of sleep difficulties could not be established in each group. Future studies would benefit from the measurement of sleep difficulties in a larger cohort of children born with NE and the inclusion of a control group for comparison.

The third hypothesis concerned the relationship between language and motor abilities in children born with NE. As hypothesised, language and motor outcomes in children born with NE were positively related. Development is not domain specific and language skills develop alongside skills from other developmental domains including fine and gross motor skills. An infant's ability to navigate and interact with the environment allows them to practice skills that facilitate language and communicative functions (Iverson, 2010; Valla et al., 2020). For example, the use of early gestures has been linked with typical receptive and expressive vocabulary in children with perinatal brain lesions. In contrast, children who did not gesture during infancy were likely to experience language delay (Sauer et al., 2010).

Limitations

The current study has some limitations. From birth to follow up, the study was impacted by high levels of attrition leading to a limited sample size. TH is administered

in only four hospitals in Ireland (three of which are hospitals where participants were recruited for this study), therefore many patients recruited to this study live outside of Dublin county and were transported to these hospitals for specialist treatment as outborn patients. When invited for follow-up, some families were unable to attend the assessment due to the distance of the laboratory from their homes. Due to the costs and resources required to travel long distances, well-educated, high resource households were more likely to attend the developmental follow-up which compromises the generalisability of the results. Due to the small sample size of this study, the results must be interpreted with caution. It is possible that the low levels of cognitive delay observed in this sample of children born with NE may be attributed to the study's limited sample size and demographic.

The grade of NE sustained by the child is one of the most important predictors of child developmental outcomes (Robertson et al., 1985; Van Handel et al., 2007). Due to the small sample size of this study and the uneven group sizes of children with each grade of NE, the grade of NE could not be taken into consideration. In this study the majority of children were diagnosed with moderate NE. Thus, differences in developmental outcomes for each grade of NE could not be established. Recent research focuses on the importance of investigating the developmental outcomes of children born with mild NE (Conway et al., 2018; Finder et al., 2019). TH is not typically used to treat infants with mild NE making recruitment of those diagnosed with mild NE more difficult. Future studies should consider the inclusion of children born with mild NE to help better define the developmental outcomes of this group.

Despite these limitations this study has many strengths including the inclusion of a control group and the observation of developmental outcomes by subdomain. The majority of studies that investigate the developmental outcomes of children born with NE

focus on global development and tend to report only composite scores. Although normative data is available for standardised measures of development, many studies have found such tools inflate scores in typically developing children and underestimate developmental delay (Anderson et al., 2010; Green et al., 2023; Jary et al., 2013). The inclusion of a control group in the current study allows us to directly compare outcome scores with those born with NE and counteracts the possibility of inflated scores.

TH has only been the standard treatment for children born with NE in the past ten years. Accordingly, many of the studies of developmental outcomes relate to children who have not undergone TH. This study adds to the body of literature that investigates the development of children born with NE treated with TH (Azzopardi et al., 2014; Natarajan et al., 2017; Shankaran et al., 2012).

The association between childhood risk factors (such as autism, EF deficits and sleep problems) and children's developmental abilities has been established in other clinical groups and in typically developing children (Bernier et al., 2010; Maenner et al., 2021; Schlieber & Han, 2017). Despite these risk factors being common in those born with NE, the relationship with children's developmental abilities had yet to be investigated in this clinical group. This is the first study to analyse the association between factors such as autism symptoms, EF deficits and sleep problems, and developmental outcomes in children born with NE.

Previous studies have established a link between motor and cognitive abilties in children in clinical groups including those born with NE (Houwen et al., 2016; Jary et al., 2019). Despite the vast literature outlining the link between motor and language development in typically children (Iverson et al., 2010; Valla et al., 2020) this is the first study to report a relationship between motor and language abilites in children born with NE. Future research should investigate fine grained behaviours characteristic to children

born with NE and how these features might promote or hinder specific domains of development.

Chapter Five

Study 1: Parent-Child Interaction and child outcomes in the context of atypical development – Insights from a study of children with an elevated likelihood of autism.

Background

Parent-child interactions play a critical role in facilitating the development of children (see chapter two for a review of the literature on parent-child interactions and child development). The bidirectional nature of parent-child interactions means that parents and their children have a dynamic and reciprocal influence on each others' behaviour when they interact with one another (Sameroff, 2009). A parent's behaviour, communicative style and receptiveness can facilitate an environment that supports child learning. Similarly, the way the child behaves and responds to these cues can alter the way the parent interacts with them. Clinical conditions have been found to alter the behaviour of the child and the parent. Thus, the parent-child interactions of dyads that include children in clinical groups have been found to differ when compared to typically developing dyads (Festante et al., 2019; Rozenbaltt-Perkel & Zaidman-Zait, 2020; Wan et al., 2013).

The parent-child interactions of those with a diagnosis or an elevated likelihood of autism has been extensively researched. However, not many studies have examined the relationship between global qualities of parent-child interaction and the developmental outcomes of children in this clinical cohort. Children with autism experience symptoms that influence their ability to process and respond to social information (Kojovic et al., 2019). In particular, children with autism experience challenges in interpreting social cues and expressing themselves appropriately (Pijl et al., 2021; Russell et al., 2012). In turn, parents may misunderstand their child or have expectations in relation to their child's

developmental level. As a result parents may alter the way they communicate and respond to their child with autism (Meirsschaut et al., 2011; Vivanti & Nuske, 2017). Additionally, parents of children with autism may experience higher levels of stress due to the demands of caregiving (Enea & Rusu, 2020). Stress has been found to influence parent-child interactions and can impact the way a parent interacts with their child (Magill-Evans & Harrison, 2001).

Children born with NE are 5.9 times more likely to be diagnosed with autism compared to typically developing children (Badawi et al., 2006). Similar to those with autism, children born with NE experience problems with social functioning (Lindström et al., 2006), information processing (Tonks et al., 2019) and are at-risk of developmental delay (Robertson et al., 1985; Schreglmann et al., 2019). Parents of children born with NE experience higher levels of stress and are uncertain of their child's long-term outcomes (Lemmon et al., 2016). Mothers also reported that the administration of TH hampered the parent-infant bond due to restricted skin-to-skin contact whilst their child was being treated (Lemmon et al., 2017). Despite these findings, the parent-child interactions of this clinical group have yet to be investigated.

Due to the overlapping characteristics between those impacted by autism and NE, and the lack of research on parent-child interaction following NE, the current study presented in this chapter has been conducted as a preliminary investigation of the longitudinal associations between parent-child interactions and developmental outcomes in a closely related clinical group, namely children with an elevated likelihood of autism. This exploratory study has been conducted as a precursor to investigating the parent-child interactions of dyads following NE. Conducting this study has allowed us to refine behavioural coding techniques, to test a macroanalytic coding framework for its

usefulness in this context and to select appropriate constructs of parent-child interaction to be studied in the context of those impacted by NE.

Abstract

Background. Early parent-child interactions have a critical impact on the developmental outcomes of the child. It has been reported that infants with a family history of autism and their parents may engage in different patterns of behaviours during interaction compared to those without a family history of autism. Thus, this study investigated the association of parent-child interactions with child developmental outcomes of those with typical and elevated likelihood of autism.

Method. This longitudinal study investigated the relationship between global attributes of parent-child interaction and the developmental outcomes of infant siblings with elevated likelihood (EL: n= 29) or typical likelihood (TL: n = 39) of developing autism. Parent-child interactions were video recorded during a session of free-play when the infants were six months of age. The quality of parent-child interactions was rated using the Manchester Assessment of Caregiver-Infant Interaction. Developmental assessments were carried out when the children were 12 and 24 months of age.

Results. The intensity of mutuality was significantly higher in the TL group than in the EL group, and developmental outcomes were poorer in the EL group when compared to the TL group. Positive associations between parent-child interaction scores at six months and developmental outcomes at 12 months were observed only in the TL group. However, in the EL group, higher levels of infant positive affect and attentiveness paid to the caregiver is associated with lower autism symptoms. Due to the sample size and design of the study, the findings must be viewed as indicative.

Conclusion. This preliminary investigation demonstrated differences in the association between parent-child interaction quality and developmental outcomes for children with typical and elevated likelihood for autism. Future studies should combine

micro-analytic and macro-analytic approaches to parent-child interaction to further examine the nature of this relationship.

Introduction

Autism is a neurodevelopmental condition characterised by delays in communication and social interaction, alongside restrictive interests and repetitive behaviours (APA, 2013). Children with autism often experience developmental delay, predominantly affecting language and social-emotional development (Bellman et al., 2013; Delehanty et al., 2018; Raza et al., 2020). Autism is a highly heritable condition (Rosenberg et al., 2009), therefore an increased likelihood of developing autism could be determined by assessing family history of psychiatric and neurodevelopmental problems. Those with a first-degree relative, such as a biological parent or sibling diagnosed with autism, have an Elevated Likelihood (EL) of developing the disorder. Individuals with no family history of neurodevelopmental disorders have Typical Likelihood (TL) of developing autism (Ozonoff et al., 2011). The use of "high-risk" and "low-risk" is often used when investigating early signs of autism in infants. Advocates have suggested that the use of the term "risk" has negative connotations. The preferred terminology is now "increased or elevated likelihood" and "low or typical likelihood" (Fletcher-Watson et al., 2017).

The Transactional Model of Child Development (Sameroff, 2009) describes parent-child interaction as a reciprocal process that involves the parent's response to the child and in turn the child's response to the parent. The observation of parent-child interactions allows us to understand a child's social, relational, and communicative abilities in a dynamic transaction shaped by the social opportunities and environments provided by their caregivers. The quality of parent-child interactions is a significant mediator in the relationship between neurobiological risk factors and child development

(Beaudoin et al., 2019). Higher quality parent-child interactions, indicated by reciprocal and engaging dyadic interactions, are associated with improved developmental outcomes in domains such as cognitive (Evans & Porter, 2009; Feldman, 2007), social-emotional (Cerezo et al., 2008) and language development (Topping et al., 2013). The heritable nature of autism, combined with its impact on communication and social development, means children with an EL of developing autism and their parents are susceptible to lower quality interactions when compared to parent-child dyads with TL for autism.

Global and microanalytic observational tools have been used to measure qualities of parent-child interactions in parent-infant dyads where the infant is at EL of autism (Wan et al., 2018). Global rating tools are used to assess general constructs of interest. The behavioural quality is typically scored on a scale and indicate whether high levels of the behaviour are present during the interaction or low levels of the behaviour are observed. Microanalytic tools are used to code clearly defined behaviours during parent-child interactions by coding behavioural constructs on a second-by-second basis (Morawska et al., 2015).

Elevated Likelihood for Autism and Infant Behaviours

The investigation of parent-child interaction in infants with an 'EL' of autism is of particular interest as it allows an exploration of the emergence of early characteristics that relate to the autism phenotype. Children on the autism spectrum exhibit behaviours that may impact the quality of parent-child interactions, for example, they have been shown to display fewer social interests and reduced social engagement (Campbell et al., 2015; Wimpory et al., 2000) compared to children who are typically developing. Studies utilising microanalytic coding schemes have identified that EL infants display lower levels of prosocial behaviours (Russell et al., 2012) and communicative gestures (Delehanty & Wetherby, 2021), less positive facial expressions (Cassel et al., 2007) and

limited joint attention (Adamson et al., 2019). When observing parent-child interactions on a global level, infants with EL show fewer and less clear initiations of interaction (Pijl et al., 2021). They are also found to display lower levels of positive affect and attention towards their parent when compared to infants with TL (Wan et al., 2013).

Infants primarily learn through the interactions they have with their caregivers. During typical development, learning is facilitated through communicative and affective cues provided by the parent. The infant takes notice, learns and responds to such cues, but this process is observed to be disrupted in those with autism (Vivanti & Nuske, 2017). Instances of learning that occur during parent-child interactions may be interrupted if infants display lower levels of positive affect or pay less attention their caregivers. This in turn may have a negative impact on child developmental outcomes in those with an EL for autism.

Autism symptoms are being identified in children at an increasingly young age. Behavioural differences between those with EL and TL of developing autism have been reported in infants as young as six months of age (Cassel et al., 2007; Gangi et al., 2020), although these differences are not clearly established until they are 12 months of age (Ozonoff et al., 2010; Rozga et al., 2011). Additional studies investigating the early emergence of autism traits during naturalistic parent-child interactions in infants six months and younger are required.

Elevated Likelihood for Autism and Maternal Behaviours

It is important to consider parental behaviours during instances of interaction. As parents are aware of the heritability of autism, mothers of infants with an EL for autism may adjust their behaviours if they believe their infant is at an increased risk of developing autism. Additionally, the parent may have adapted their parenting in response to their older children with autism. Studies investigating parent behaviours using global

measures of parent-child interaction demonstrate that mothers of infants with EL of autism are more likely to display different levels of sensitivity (Wan et al., 2013) and higher levels of directive behaviour during parent-child interaction, perhaps as a response to their infant's behaviours (Meirsschaut et al., 2011; Quigley & McNally, 2013; Wan et al., 2012). Parental directive behaviour coded during interaction using microanalytic coding schemes, is characterised by increased gesture use (Talbott et al., 2015), closer physical proximity (Srinivasan & Bhat, 2020) and a greater amount of verbal instruction directed to the infant (Harker et al., 2016; Patterson et al., 2014).

Different patterns of associations have been observed between directive parental behaviours and child outcomes. Some studies have found directiveness to be associated with reduced infant engagement and slower development in both typically developing children and those with an EL for autism (Saint-Georges et al., 2011). When mothers exhibit directive behaviours higher levels of negative affect are observed in infants with an EL for autism (Steiner et al., 2018). This impacts the frequency and duration of the interaction that may otherwise promote learning in children. However, other studies consider directiveness to be adaptive and contribute functional value to child development. Doussard-Roosevelt and colleagues (2003) found children with autism to be responsive towards maternal directive behaviours. Although intrusive behaviours often disrupt children's play, directives are often preceded with high levels of joint engagement in the activity prompted by the caregiver assuming that it is sensitive to the infant's state and interests (Bottema-Beautel et al., 2018; Masur et al., 2013).

Caregiver sensitivity describes the ability to detect and interpret the infant's behavioural signals and to respond to them with attentiveness, warmth, appropriate engagement and support. Whilst Wan et al. (2012, 2013) did report lower levels of sensitivity in mothers of EL infants, many studies have provided evidence of similar

levels of sensitivity for caregivers of EL and TL cohorts (Baker et al., 2010; Harker et al., 2016).

Elevated Likelihood for Autism and Dyadic Behaviours

Dyadic mutuality describes the general amount of engagement shared between the mother and infant. It considers the degree of reciprocity, attunement and synchrony shared in the dyad. Mutuality is expressed through the infant's acceptance of the caregiver's involvement and is represented in togetherness through cooperative play, flow and body orientation (Deater-Deckard & Pentrill, 2004; Wan et al., 2017). Dyad engagement intensity refers to the quality of the mutual activity between the mother and infant. It is characterised by the level of interest and positivity the dyad expresses towards one another or through mutual focus on an object. Dyadic mutuality and engagement intensity are global constructs that have been directly linked to favourable outcomes in children such as positive, reciprocal relationships (Ensor et al., 2011) and a reduction in child behaviour problems (Pasiak & Menna, 2015). Features of parent-child mutuality and engagement intensity coded microanalytically such as synchrony and joint attention have been linked to improved cognitive and language outcomes in children (Provenzi et al., 2018; Salo et al., 2018). Mutual interactions allow the parent and infant to take part in shared opportunities that facilitate infant learning. The parent can provide experiences that are informative, the infant can learn from these cues, respond to them and increase their proficiency through back and forth interaction (Edmunds et al., 2019). Therefore, it is expected that higher levels of dyadic mutuality and engagement intensity will contribute to improved developmental outcomes in children.

When compared to dyads of infants with TL of developing autism, mother-infant dyads where the infant is at EL of autism have been observed to exhibit lower interactive reciprocity, dyadic mutuality and engagement during parent-child interactions (Campbell

et al., 2015; Wan et al., 2013). Parent-child dyads of typically developing children spend more time in mutual engagement compared to dyads where the child has a diagnosis of autism (Rozenbaltt-Perkel & Zaidman-Zait, 2020). Infants with EL of developing autism tend to display lower levels of synchrony (Kellerman et al., 2020), joint attention (Adamson et al., 2019) and social interest (Killmeyer et al., 2019) when compared to infants with TL. Such behaviours are important when eliciting mutual engagement with their parent and lower levels of these behaviours may therefore contribute to lower levels of engagement intensity.

A study on children with a developmental language disorder found attuned dyadic behaviour to be associated with improved receptive language ability, while parent behaviours alone were not associated with child language abilities (Jokihaka et al., 2022). This suggests that it is important not only to consider the behaviours of the parent and child, but also of the dyad. Other scales measuring global qualities of parent-child interactions (Biringen, 2000) have been used to measure parent-child interactions where the child has a diagnosis of autism (Dolev et al., 2009). But such scales only consider qualities of the parent and the child separately and do not take into account the cooperation between the pair of individuals.

Investigating the Quality of Parent-Child Interaction and Child Outcomes in Autism

The majority of studies that investigate play behaviours in relation to child developmental outcomes in those with an EL of developing autism have observed associations with behaviours coded microanalytically (Northrup & Iverson, 2015; Salo et al., 2018; Talbott et al., 2015) but research has yet to establish the relationship between global constructs of parent-child interaction and developmental outcomes. Morawska and colleagues (2015) found global measures of parent-child interactions to be more sensitive when coding behaviours during play. In a study of infants with EL or TL of developing

autism, Wan et al., (2013) examined whether the quality of parent-child interactions at 12 to 15 months predicted autism classification at three years of age. Using the Manchester Assessment of Caregiver and Infant Interaction (MACI; Wan et al., 2017) to rate the quality of parent-child interactions, Wan et al. (2013) found that lower levels of dyadic mutuality, infant positive affect and attentiveness were associated with autism symptom severity at three years.

The current study is a preliminary investigation examining the relationship between global qualities of parent-child interaction and developmental outcomes in infants at EL for autism. While previous studies (e.g. Wan et al., 2013) focussed on predicting autism outcomes, the current study investigated the relationship between parent-child interactions and domain-specific developmental outcomes using standardised assessments.

The Current Study

The current study investigated the longitudinal relationship between emerging parent-child interaction qualities at six months of age, and the child's developmental functioning at 12 and 24 months. Repeated measures were carried out on an independent sample of children with EL or TL of developing autism. Parent-child interactions at six months were rated using the MACI (Wan et al., 2017) and developmental outcomes were assessed using the Mullen Scales of Early Learning (MSEL; Mullen 1995). Symptoms of autism were also assessed at 24 months of age using the Autism Diagnostic Observation Schedule-Generic (ADOS-G; Lord et al., 2000). The study examined whether the association between parent-child interaction qualities and developmental outcomes was differentially impacted by autism likelihood status. For the purpose of this study, high quality parent-child interactions will be characterised by higher levels of sensitive responding from the mother, attentiveness to their caregiver, liveliness and positive affect

from the child, and higher levels of mutuality and engagement intensity observed between the parent and child.

The following hypotheses were investigated.

Hypothesis 1. Compared with the TL group, infants at EL of developing autism would display distinct interactions characterised by lower levels of attentiveness towards their mother, reduced liveliness and less positive affect. Mothers of EL infants would display similar levels of sensitivity but more directive behaviours. EL dyads are predicted to display lower levels of mutuality and the intensity of the engagement will be lower.

Hypothesis 2. For the relationship between parent-child interaction variables and developmental outcomes tested within each group, it was expected that higher quality parent-child interactions would be positively associated with developmental outcome scores in the EL and TL groups. High quality parent-child interactions will be negatively associated with child autism symptoms in those in the EL group.

Method

The study is based on longitudinal data collected as part of the PRegnancy Investigation of Siblings and Mothers of children with autism (PRISM) study, conducted in Perth, Western Australia (Unwin et al., 2016). Expectant mothers of TL infants were recruited through advertisements in local newspapers. Those expecting EL infants were recruited through referrals from obstetricians, gynaecologists, and via advertisements in local newspapers. Mothers and their children were invited for participation at three separate time points, specifically, when the child was 6, 12 and 24 months of age.

Participants

Sixty-eight mothers and their infants enrolled in the study. The sample was categorised into two groups depending on the infant's autism likelihood status. Twenty-nine infants (42.65%) were considered at elevated likelihood (EL) of developing autism,

and 39 infants (57.35%) were considered as having typical likelihood (TL) for autism. Infants who had an older biological sibling with a clinical diagnosis of Autism or Pervasive Developmental Disorder – Not Otherwise Specified (PDDNOS) were defined as "EL". The standard diagnostic procedure for autism in Western Australia during the time of this study followed the DSM-IV guidelines (APA, 1994). Clinical protocols at the time required a multidisciplinary healthcare team to assess each case and comprised a paediatrician, clinical psychologist and a speech and language pathologist. For each EL family, the mother participating in the study was the biological mother of both the EL child and the older sibling with autism. Infants were defined as "TL" if they had no known developmental risk factors and also had one or more biological siblings of at least three years of age without a diagnosis of a neurodevelopmental disorder. The appropriate local ethics committees granted ethical approval for this study. Participating caregivers gave informed consent on behalf of themselves and their infant to participate in the study. Before completing each stage of the study, all participants were reminded that they could withdraw from the study if they wished and were debriefed at the end of the study.

Sample Characteristics. Demographic information collected from participating mothers included the child's age, gender, gestational age and number of siblings, maternal age, household income and maternal autism traits. Demographic characteristics are displayed according to group in Table 5.1.1. The sample of children participating in the study did not differ significantly in gestational age or gender when comparing the two groups. The number of siblings however was higher in the EL group compared to the TL group. Household income was classified into 12 categories starting from $1 to $8,000 per year up to $104,001 or more per year. The TL group was found to have a significantly higher household income than the EL group. The median household income of the TL group was $104,001 or more per year (income category 12), whereas the median income

of the EL group was $78,001 to $104,000 per year (income category 11). Autistic traits were measured in mothers using the Autism Spectrum Quotient (AQ) questionnaire (Baron-Cohen et al., 2001). Neither group exhibited significant levels of autistic traits (i.e., no mother obtained a score of 32 or above) and there was no statistical difference in maternal AQ scores between the EL group and TL group (see Table 5.1.1).

Table 5.1.1. *Sample characteristics of Typical and Elevated Likelihood groups.*

Characteristics	**TL (N = 39)**	**EL (N = 29)**	***df***	**Test Statistic**
Gestational Age (Weeks; *M, SD*)	38.54 (1.19)	38.59 (1.18)	66	t = 0.16, p = .870
Gender			1	X^2 = 0.10, p = .751
Male (*%*)	20 (51.28)	16 (55.17)		
Female (*%*)	19 (48.72)	13 (44.83)		
Number of Siblings (*Mdn, Range*)	1.00 (4.00)	2.00 (5.00)		U= 177.00, p = .000**
Maternal Age (Years; *M, SD*)	33.29 (3.25)	35.71 (4.54)	65	t = 2.55, p = .013**
Household Income (*Mdn, Range*)	12.00 (3.00)	11.00 (8.00)		U = 309.00, p = .002*
Maternal AQ Score (*M, SD*)	11.42 (4.64)	13.18 (6.18)	64	t = 1.45, p = .191

Note. Maternal AQ Score measured using Autism-Spectrum Quotient Questionnaire

* $p < 0.05$; ** $p < 0.01$

Procedure

When the infants were six months of age, both mother and infant were invited to the laboratory at the Telethon Kids Institute (The University of Western Australia) and a session of parent-child interaction was video recorded. A selection of developmentally appropriate toys was provided for this interaction. The mother was instructed to play with their infant, just as they do at home. The interaction was video-recorded for six minutes commencing as soon as the research assistant left the room. At 12 months of age, children and their parents were invited back to the lab and the child's developmental functioning was assessed using the MSEL (Mullen, 1995). At 24-months, the child's developmental functioning was once again assessed using the MSEL and the children were assessed for symptoms of autism using the ADOS-G (Lord et al., 2000).

Within the EL sample, 20.7% was lost to follow-up by the time of the ADOS-G assessment at 24 months, compared with 12.8% of TL participants. A chi-square test demonstrated a significantly greater loss of participants from the EL group in comparison to the TL group ($X^2(1) = 4.22$, $p = 0.04$). Table 5.1.2 indicates the number of participants who returned for assessment at each time point according to group.

Table 5.1.2*. Group attrition rates.*

Group	N at 6m	N at 12m	N at 24m	N at ADOS-G
TL	39	39 (0.00%)	36 (7.69%)	34 (12.82%)
EL	29	26 (10.34%)	24 (17.24%)	23 (20.70%)
Full Sample	68	65 (4.41%)	60 (11.76%)	57 (16.18%)

Note. % = Attrition Rate

Analyses were conducted to investigate whether there were any characteristic differences between the completers (participants who returned and took part in every

phase of the study) and non-completers (participants who did not take part in every phase of the study) in the TL and EL groups. The quality of parent-child interactions was compared across the completers and non-completers. Where the infant had a TL of developing autism, a Mann-Whitney *U* test indicated that there were no differences in the quality of parent-child interactions on any domain of the MACI. In the EL group, there was a significant difference in infant affect observed between the two groups. The infants who were in the non-completer group (*Mdn* = 1.00) had significantly lower scores on the positive affect domain of the MACI compared to those in the completer group (*Mdn* = 3.00) $U = 14.00$, $Z = -3.33$, $p = .001$.

Materials

The Manchester Assessment of Caregiver-Infant Interaction (MACI). The parent-child interaction videos were rated by a trained research assistant using the global analytic rating scheme, MACI (Wan et al., 2017). The MACI is designed to assess the quality of seven domains of parent-child interaction: caregiver sensitivity, caregiver non-directiveness, the infant's attentiveness to caregiver, infant positive affect, infant liveliness, dyad mutuality and dyad engagement intensity. Each domain is rated on a seven-point Likert scale and each parent-child interaction video is given one rating for each domain on the scale, giving a total of seven scores for each video. A score on the higher end of the scale (i.e. scores closer to 7) indicates high levels of the quality specified.

Caregiver sensitivity describes the parent's response to infant behaviour. Caregivers demonstrating high sensitivity respond to their infants with an attentive attitude, warmth, appropriate engagement and support. A score of 1 indicates minimal sensitive responsiveness and a score of 7 indicates high sensitive responsiveness. When a caregiver exhibits non-directiveness they follow the lead of the infant during play and

provide feedback that relates to the infant's focus of attention. A caregiver demonstrating high levels of directiveness will exhibit demanding and intrusive behaviours and use negative comments during the interaction with their infant. A score of 1 indicates highly directive caregiver behaviour and a score of 7 indicates a highly nondirective caregiver. The infant's attentiveness to their caregiver refers to the infant's use of direct eye contact, body orientation, and joint activity in relation to their caregiver. The infant may reference caregiver activity through acts such as imitation. A score of 1 indicates that the infant was minimally attentive and a score of 7 indicates that the infant was highly attentive to their caregiver. Infant positive affect measures the infant's use of positive behaviours, vocalisations and expression during the parent-child interaction. Negative affect is demonstrated through the use of negative expressions, vocalisations and gestures. A score of 1 indicates that the infant exhibited high negative affect and a score of 7 indicates high positive affect. Infant liveliness measures the amount and level of physical activity the infant partakes in during the interaction and considers behaviours that are spontaneously initiated by the infant. A score of 1 indicates that the infant was not at all lively during the interaction and a score of 7 indicates that the infant was extremely lively. Dyadic mutuality measures the general amount of engagement shared between the parent and infant. It considers the level of reciprocity, attunement and togetherness exhibited during the interaction. The domain considers the level of shared attention, mutual play, flow, and body orientation of the dyad. A score of 1 indicates that the dyad exhibited very low mutuality, a score of 7 indicates consistently high mutuality. Engagement intensity measures the quality of the mutual interaction during periods of engagement such as mutual object focus. The domain considers the degree of interest, arousal and positivity/excitement exhibited by both parties during the period of mutuality. A score of 1 indicates almost no engagement and a score of 7 indicates a very intense engagement.

The psychometric utility, validity and reliability of the MACI have been established in a normative sample of children aged 3 to 14 months (Wan et al., 2017). In the current study, inter-rater reliability (IRR) of the MACI coding was established through blinded independent double coding of 15 (22.06%) randomly selected parent-child interaction recordings. Good to high inter-rater agreement was demonstrated on each single measure of the MACI with intra-class correlations ranging from $r = 0.63$ to 0.95.

Mullen Scales of Early Learning (MSEL). The MSEL (Mullen, 1995) is a standardised assessment tool used to measure development in children aged 0 to 68 months. In this study, the MSEL was administered when the children were 12 and 24 months of age, it took 15 to 35 minutes to administer depending on the child's age and ability. The MSEL was used to assess children's visual reception, fine motor skills, and receptive and expressive language. Visual reception is a measurement of cognitive functioning that incorporates visual discrimination, memory, organisation and spatial awareness. Tasks include placing block set shapes onto a puzzle board, producing object associations and looking for missing objects. The MSEL assesses fine motor skills such as motor planning and control, object manipulation and writing readiness. The use of pincer grip, grasping, and object transference is assessed by instructing the child to play with toy blocks. Tasks such as turning pages in a book and the imitation of crayon lines are also observed.

Receptive language skills, such as auditory comprehension, auditory memory and auditory sequencing, are assessed. Children are tested to see if they understand familiar names, words and instructions and can identify their own body parts and pictures in a book. Expressive language abilities assessed include speaking, language formation and verbal conceptualisation. The production of babbling, consonant sounds and two-syllable

sounds is observed in younger children. The naming of objects and use of two-word phrases is assessed during the 24-month assessment.

Visual reception, fine motor and language scores are combined to calculate the Early Learning Composite Score (ELC). The ELC provides a measure of general intellectual functioning and is used to avoid truncation in the case of extreme scores on singular domains. The raw score of each subdomain is converted into a T-score, where 20 is the minimum score and represents a score that is three standard deviations below the mean, a score of 50 is considered average, and the standard deviation is 10. The MSEL has an internal validity between 0.75 and 0.80 dependent on the domain, and a test re-test coefficient of 0.70 in children aged one to twenty-four months (Mullen, 1995).

Autism Diagnostic Observation Schedule-Generic (ADOS-G). Module one of the ADOS-G (Lord et al., 2000) was administered to measure autism symptoms in the children when they reached 24 months of age. A trained administrator carried out the play-based assessment designed to elicit social interaction, communication skills, imagination and creativity (flexible thinking), and stereotyped behaviours. Upon assessment, individuals are rated with an ADOS-G classification that includes non-spectrum, autism or autism-spectrum, dependant on their scoring on the four assessed domains. A higher score on the ADOS-G indicates increased autism symptom severity. A score of at least 14 on the social and communication scales indicates an autism classification. A score of at least 7 on these domains is required for an autism-spectrum classification. A score below 7 yields a non-spectrum classification. The ADOS-G demonstrates very good inter-rater reliability coefficients (kappas exceeding 0.60) and test re-test coefficients ranging from 0.59 to 0.82. Overall, seven children across the cohort exhibited high levels of autism symptoms by receiving a score of 7 or more, six from the EL group and one child from the TL group. Due to the small number of those

displaying autism symptoms, any group differences that might emerge as early signs of autism may not be detected.

Autism-Spectrum Quotient (AQ). Due to the heritable nature of autism, the AQ (Baron-Cohen et al., 2001) was used to measure traits of autism in the mothers in this sample. The AQ is a self-report questionnaire that comprises fifty statements that positively or negatively relate to traits of autism in adults. It measures five traits of autism including social skills, imagination, communication, attention switching and attention to detail. Examples of statements include "I notice patterns in things all the time" and "I enjoy social chitchat". The mothers were asked to rate each statement using a four-point scale ranging from "definitely agree" to "definitely disagree". A score of 32 or above on the AQ is indicative of significant autism traits; 80% of individuals with a score above this threshold received a clinical diagnosis of high functioning autism (Baron-Cohen et al., 2001). The AQ demonstrates good test re-test and interrater reliability. When administered twice within a two-week time period, scores from the first and second administration of the AQ did not differ at the group level and were strongly correlated. The AQ yielded Cronbach alpha coefficients between 0.62 to 0.77, indicating moderate to high internal consistency (Baron-Cohen et al., 2001).

Statistical Analysis

Statistical analyses for this study were conducted using SPSS (Version 26), with alpha criterion set to $p < 0.05$ unless stated otherwise. Data were checked for normality to meet the assumptions of parametric statistics using Shapiro Wilk's test and homogeneity of variance was checked using Levene's test. Log transformations were applied to non-normal data and non-parametric tests were used when such transformations did not produce normality. Group differences in demographic characteristics, autism symptoms (using the ADOS-G) and the quality of parent-child interaction (using the MACI) were

analysed using independent samples t-tests, Mann-Whitney *U* or Chi-square tests as appropriate. A 2x2x4 ANOVA was conducted to test for group differences in developmental outcomes using the four MSEL domain scores from assessments at 12 and 24 months. Kendall's Tau correlation coefficients were calculated to analyse the relationship between individual domains of the MACI, the MSEL and ADOS-G.

Results

Developmental Functioning and Autism Symptoms

MSEL scores of children collected at 12 and 24 months were compared by group. Table 5.1.3 presents descriptive statistics and group differences for these developmental outcome scores. A 2x2x4 analysis of variance (ANOVA) was used to test differences in MSEL scores for the four domains (visual reception, fine motor skills, and receptive and expressive language) assessed at two time points (12 and 24 months) according to group (TL and EL). Due to heterogeneity of variance within the data, the significance level for this analysis was adjusted to $p < 0.01$ to ensure the robustness of the findings using this parametric test. The sphericity assumption was met for the effect of each domain as indicated by Mauchly's test, $X^2(5) = 8.02$, $p = 0.155$. Interaction effects from the ANOVA were not significant, the EL and TL groups did not differ significantly in MSEL scores when factoring in age of assessment. There was a significant effect of autism group, with the EL group scoring significantly lower than the TL group on each domain of the MSEL, $F(3,174) = 4.91$, $p = 0.003$. There was also a significant main effect of age of assessment, with MSEL scores lower for assessment at 12 months compared to assessment at 24 months for both groups $F(3,174) = 5.53$, $p = 0.001$.

Table 5.1.3. *Descriptive statistics for MSEL scores at 12 and 24 months for each group.*

		Group					
		Typical Likelihood			*Elevated Likelihood*		
Child Age	**Domain**	**Mean**	**SD**	**Range**	**Mean**	**SD**	**Range**
12 Months	Visual Reception	51.15	7.25	28.00	47.31	8.85	36.00
	Fine Motor	57.33	9.50	38.00	52.77	8.07	30.00
	Receptive Language	52.95	4.81	21.00	49.23	6.94	30.00
	Expressive Language	52.26	5.59	20.00	50.00	8.28	32.00
	ELC	106.46	10.24	44.00	99.92	11.32	43.00
24 Months	Visual Reception	63.06	8.45	37.00	56.13	15.97	60.00
	Fine Motor	52.89	8.45	36.00	46.79	10.09	45.00
	Receptive Language	59.89	6.22	23.00	52.08	13.20	51.00
	Expressive Language	62.36	9.72	41.00	51.58	11.31	40.00
	ELC	119.17	12.09	47.00	103.54	21.16	77.00

Note. ELC = Early Learning Composite.

Symptoms of autism in the children were measured when they were 24 months of age using module one of the ADOS-G (Lord et al., 2000). Mann-Whitney *U* tests were conducted to compare ADOS-G domain scores between the EL and TL groups. Children in the EL group received higher scores (indicative of more pronounced autism symptoms) on the communication, flexible behaviour and stereotyped thinking domains of the ADOS-G when compared to the TL group (see Table 5.1.4 for descriptive statistics for the two groups and outcomes of the Mann-Whitney *U* tests).

Table 5.1.4. *ADOS-G scores at 24 months by group.*

	TL (*N = 34*)		EL (*N = 23*)		Group Difference			
ADOS-G Domain	**Median**	**IQR**	**Median**	**IQR**	***U***	***Z***	**Cohen's d**	***p***
Communication	0.50	1.25	1.00	3.00	328.50	-2.10	0.52	0.04*
Social	0.00	1.00	1.00	3.75	338.50	-1.72	0.48	0.09
Flexible Thinking	0.00	1.00	0.50	2.00	344.50	-2.04	0.46	0.04*
Stereotyped Behaviours	0.00	0.00	0.00	0.25	371.00	-2.52	0.36	0.01*

Note. Higher scores on the ADOS-G reflect higher levels of autism symptoms

*$p < 0.05$.

Comparing Parent-Child Interaction Quality at 6 Months in TL and EL groups

In order to test hypothesis 1, the quality of parent-child interaction was compared between the EL and TL groups. Shapiro Wilk's tests and histograms indicated that the MACI data were not normally distributed. Therefore, the groups were compared for each of the seven domain scores using a Mann-Whitney *U* test. Descriptive statistics are presented in Table 5.1.5 and tests of group differences in MACI scores are presented in Table 5.1.6. A Mann-Whitney *U* test indicated that the level of engagement intensity in the EL group was significantly lower compared to the TL group. There were no significant group differences on any of the other MACI variables (see Table 5.1.6) but a number of domains demonstrated a pattern of non-significant trends where the EL group displayed lower levels of caregiver sensitivity, infant attentiveness to caregiver and dyadic mutuality compared to the TL group.

Table 5.1.5. *Descriptive statistics for MACI scores at 6 months for each group.*

	Group					
	Typical Likelihood			*Elevated Likelihood*		
	Mean	**SD**	**Range**	**Mean**	**SD**	**Range**
Infant Age (in months)	6.13	0.24	1.27	6.10	0.27	1.10
Interaction Domain						
Caregiver Sensitivity	3.79	1.24	5.00	3.28	1.00	3.00
Caregiver Non-directiveness	4.18	1.34	4.00	3.62	1.45	5.00
Infant Attentiveness to Caregiver	3.97	1.22	5.00	3.41	1.21	5.00
Infant Positive Affect	2.82	1.59	6.00	2.52	1.60	5.00
Infant Liveliness	3.95	1.05	4.00	4.14	0.99	3.00
Dyadic Mutuality	3.33	1.32	5.00	2.83	1.00	4.00
Engagement Intensity	3.79	0.77	4.00	3.41	0.95	4.00

Note. Interaction domains are scored on a scale from 1–7; low scores indicate poor interactions (for example, lower levels of sensitivity).

Table 5.1.6. *Comparison of MACI scores at 6 months by group.*

	Typical Likelihood (*N = 39*)		*Elevated Likelihood* (*N = 29*)		*Group Difference*			
Interaction Domain	**Median**	**IQR**	**Median**	**IQR**	***U***	***Z***	**Cohen's d**	***p***
Caregiver Sensitivity	4.00	2.00	3.00	2.00	433.50	-1.70	0.41	0.09
Caregiver Non-Directiveness	4.00	2.00	4.00	3.00	445.50	-1.52	0.37	0.13
Infant Attentiveness to Caregiver	4.00	2.00	4.00	2.00	429.50	-1.76	0.42	0.08
Infant Positive Affect	3.00	3.00	2.00	2.50	498.50	-0.86	0.20	0.39
Infant Liveliness	4.00	2.00	4.00	2.00	515.00	-0.65	0.15	0.51
Dyadic Mutuality	3.00	2.00	3.00	1.50	434.50	-1.68	0.40	0.09
Engagement Intensity	4.00	1.00	3.00	1.00	415.00	-2.01	0.47	0.05*

Note. Interaction domains are scored on a scale from 1–7; low scores indicate poor interactions (for example, lower levels of sensitivity*).*

*$p \leq 0.05$.

Associations between Parent-Child Interaction Variables at 6 Months and Child Outcome Variables at 12 & 24 Months

In order to test hypothesis 2, Kendall's Tau correlations were computed to analyse the relationship between the quality of parent-child interactions and developmental outcomes in children with TL and EL of developing autism. Parent-child interactions were observed when the infants were 6 months of age, MSEL scores were acquired across the 12- and 24-month collection points and ADOS-G scores were obtained when the children were 24 months of age. The patterns of relationships in the two groups are distinct and multiple significant associations were observed between MACI scores and MSEL scores collected at 12 months in the TL group; these correlations are displayed in Table 5.1.7 for the TL group and in Table 5.1.8 for the EL group.

In the TL group, caregiver sensitivity, caregiver non-directiveness, infant attentiveness to caregiver, infant positive affect, dyadic mutuality and engagement intensity positively correlated with performance on visual reception. Caregiver sensitivity and dyadic mutuality positively correlated with performance on the fine motor domain. Engagement intensity scores were positively associated with expressive language skills. Higher levels of caregiver sensitivity, dyadic mutuality and engagement intensity were positively related to ELC scores. For the relationship between MACI and ADOS-G scores, higher levels of engagement intensity were associated with improved social functioning and flexible thinking.

Fewer significant associations were observed in the EL group. Surprisingly, higher levels of engagement intensity were associated with lower ELC scores at 12 months among the EL group. However, as expected, infants who showed less attention to their mother scored higher (i.e., displayed more pronounced symptoms) on the communication and flexible thinking domains of the ADOS-G. Infant positive affect

scores were also negatively associated with flexible thinking domain scores (i.e., less positive affect was associated with greater inflexibility in thinking).

No significant associations were observed between any MACI scores at six months and MSEL scores in any domain in either the TL or EL group at 24 months.

Table 5.1.7.*Correlations among the study variables in the Typical Likelihood Group.*

	MACI Variables						
	Caregiver Sensitivity	Caregiver Non-directiveness	Infant Attentiveness to Caregiver	Infant Positive Affect	Infant Liveliness	Dyadic Mutuality	Engagement Intensity
MSEL Variables							
Visual Reception	.28*	.26*	.25*	.25*	.14	.30**	.28*
Fine Motor	.20	.09	.04	.14	.14	.20	.06
Receptive Language	.00	-.02	.05	-.07	.02	.05	.09
Expressive Language	.16	.17	.15	.05	.19	.20	.32*
ELC	.24*	.19	.15	.15	.14	.29**	.20
ADOS-G Variables							
Communication	-.05	-.14	.07	.15	.03	.04	.09
Social	.00	-.20	-.21	-.09	.24	.12	-.43**
Flexible Thinking	.06	.04	-.24	-.18	.13	.03	-.29*
Stereotyped Behaviour	-.13	-.24	-.24	.05	.20	-.25	-.21

Note. MACI scores were collected when the children were 6 months of age, MSEL scores were acquired at 12 months of age and ADOS-G scores were obtained when the children were 24 months of age. *$p \leq 0.05$; **$p \leq 0.01$. (One-tailed).

Table 5.1.8. *Correlations among the study variables in the Elevated Likelihood group.*

	MACI Variables						
	Caregiver Sensitivity	Caregiver Non-directiveness	Infant Attentiveness to Caregiver	Infant Positive Affect	Infant Liveliness	Dyadic Mutuality	Engagement Intensity
MSEL Variables							
Visual Reception	.00	.25	-.09	-.12	-.14	-.13	-.20
Fine Motor	-.02	.09	-.10	-.08	.11	-.04	-.05
Receptive Language	-.03	.05	.03	-.11	-.11	-.16	-.19
Expressive Language	-.26	-.14	.14	-.04	.19	-.25	-.11
ELC	-.13	.13	-.07	-.18	.03	-.21	-.28*
ADOS-G Variables							
Communication	-.15	.08	-.36*	-.17	-.10	-.12	-.07
Social	.03	.07	-.08	-.02	-.04	.07	.05
Flexible Thinking	-.04	.11	-.32*	-.33*	-.01	-.02	-.18
Stereotyped Behaviour	.01	-.01	-.09	-.02	-.24	.15	-.01

Note. MACI scores were collected when the children were 6 months of age, MSEL scores were acquired at 12 months of age and ADOS-G scores were obtained when the children were 24 months of age.

*$p \leq 0.05$. (One-tailed).

Discussion

When the quality of parent-child interaction at six months was compared between the TL and EL groups, the level of engagement intensity was significantly lower in the EL group in comparison to the TL group. This finding partially supports the first hypothesis, which posited that infants with EL of developing autism and their parents would engage in lower quality interactions, characterised by lower levels of reciprocity and less engaging dyadic interactions. Engagement intensity describes the degree of mutual interest, arousal, positivity and excitement expressed by both the mother and infant (Wan et al., 2017). Consistent with the literature, a study by Wan et al. (2013) found engagement intensity at 12 months to predict autism outcomes in children at 36 months of age. It could be that children with an EL of autism found the interaction to be less interesting or rewarding, thus decreasing the intensity of the engagement. Children with autism tend to display lower levels of social behaviour, therefore mothers may adjust their behaviours to ensure they are not overbearing or intense. An engagement that is overly intense may overwhelm the infant causing them to disengage from the interaction (Segars et al., 2020). Conversely, children with an EL for autism may exhibit hypo-reactive patterns of behaviour that lead to a decrease in the number of opportunities for parents to respond to their child and can delay early learning (Grzadzinski et al., 2021). Infants who display hypo-reactive patterns of behaviour may benefit from more intense engagement as long as the parent is sensitive to the interests of the infant (Haebig et al., 2013).

Whilst engagement intensity differed between the TL and EL groups, the groups did not differ significantly on other domains of parent-child interaction including caregiver and infant qualities. Previous studies have demonstrated differences in caregiver qualities in EL groups, such as lower levels of sensitive responding and higher

levels of directive behaviour (Wan et al., 2012). However, other studies have found parental sensitivity to be similar between TL and EL cohorts (Baker et al., 2010; Harker et al., 2016; Schwichtenberg et al., 2019). Infants with EL for autism tend to exhibit fewer and less clear social initiations during episodes of parent-child interaction, but these tendencies are more pronounced in the case of older children in interaction (Pijl et al., 2021). It is possible that the global measure of parent-child interaction used in the present study (MACI; Wan et al., 2017) was not sensitive enough to detect fine-grained differences in interactions at six months of age.

Previous studies comparing groups with EL and TL of developing autism measured the quality of parent-child interaction using the MACI when children were 6 to 10 and 12 to 15 months of age (Wan et al., 2012; Wan et al., 2013). Parent-child interaction measured at 12 to 15 months (but not at 6 to 10 months) was found to significantly predict autism diagnosis at three years of age (Wan et al., 2013). This suggests that the MACI may be more sensitive to the quality of parent-child interaction in older children with EL for autism. Alternatively, when infants are 6 to 10 months old, they may not be displaying symptoms that affect the quality of the parent-child interaction. In turn, this may affect the child's developmental outcome. Wan et al. (2012) identified a larger number of group differences in parent-child interactions when comparing the interactions of those in an TL and EL group using the MACI. The study had a larger sample size and thus increased statistical power compared to the current study. The MACI scores were normally distributed and therefore treated as interval data and infant age was controlled for in their analysis. If the sample size and statistical power of the current study was increased, more differences in parent-child interactions may be observed. Indeed, a number of domains demonstrated borderline effects and the EL group displayed lower levels of caregiver sensitivity, infant attentiveness to caregiver and

dyadic mutuality compared to the TL group. It is possible that increased statistical power, and the inclusion of more children displaying high levels of autism symptoms within the EL group could increase the likelihood of detecting significant group effects on these domains of the MACI. Alternatively, the trends might disappear.

The results of the study indicate that infants in the EL group are susceptible to poorer developmental outcomes compared to those in the TL group. Furthermore, the current study found children with an EL for autism and their parents display characteristically different behaviours during parent-child interaction compared to dyads where the child has a TL of autism. This suggests that changes in behaviour in the EL group can be identified as early as 6 months of age. The detection of emerging autism symptoms at a young age is critical, as early intervention is likely to be the most effective in decreasing impairments (Landa, 2018).

The second hypothesis concerned the relationship between the quality of parent-child interactions at six months and outcome measures at 12 and 24 months and whether this relationship differed between the TL and EL groups. Different patterns of relationships were observed in the TL and EL groups. The findings indicate that parent-child interactions do not impact the developmental outcomes of those in the EL group in the same way as they do for the TL group. In the TL group, multiple positive associations were identified where higher quality parent-child interactions at six months were associated with better developmental outcomes 6 months later and lower ADOS-G scores 18 months later. In the EL group, the infant MACI scores including higher levels of infant positive affect and infant attentiveness to the caregiver were associated with lower levels of autism symptoms, as hypothesised. Our findings are consistent with the trajectory of the autism phenotype (Filliter et al., 2014), as infants who were less attentive to their

mother and displayed lower levels of positive affect displayed higher levels of autism symptoms.

No positive associations were observed between MACI and MSEL scores in the EL group. Instead the opposite effect was found and higher levels of engagement intensity at 6 months was associated with lower ELC scores at 12 months of age. The literature suggests that higher quality parent-child interactions are associated with improved developmental outcomes in children. Wan et al. (2013) found lower levels of engagement intensity to be associated with autism outcome at 36 months. Therefore, this finding is unexpected and runs counter to the study's hypothesis. Engagement intensity describes the quality of the engagement and mutual focus that the parent and child share directly or through focus on a mutual object. Certain qualities of parent-child interactions may have different effects on those with EL and TL of developing autism. In the context of these findings, it could be that higher levels of engagement intensity shared in the parent-child dyad could be less rewarding for children in the EL group, whereas children in the TL group may find the level of intensity to be engaging and enjoyable. However more definitive explanations for this unexpected finding awaits replication by further studies.

Mothers of infants with an EL for autism may have adapted their parenting in response to their older children with autism. Mothers may exhibit more structured and routine-based behaviours in order to accommodate less flexible and more demanding behaviours observed in children on the autism spectrum. Thus, even if engaged in positive mutual activity, this mutual engagement might exhibit different characteristics which may be associated with different domains of development for those on the spectrum. For example, differences in the properties of language used by the mother during the dyadic interaction might be observed. There may be variation in the use of

child directed speech addressed to the infant such as prosody, which in turn has been found to impact child language development and social responsiveness (Cohen et al., 2013; Quigley et al., 2016). Future studies might incorporate microanalytic approaches (Morawska et al., 2014) to explore fine grained characteristics of engagement intensity such as the composition of caregiver speech input, levels of synchrony, joint attention and social reciprocity. The use of microanalytic techniques in conjunction with global measurements of parent-child interaction can increase the precision and sensitivity of the observation of parent-child interaction.

In the literature various measures are used to code parent-child interactions (Lotzin et al., 2015). The lack of standardisation brings into question the reliability of these coding schemes, particularly measures that code global behavioural constructs. A variety of definitions are used to describe specific constructs of parent-child interaction. For instance, directiveness has been negatively associated with favourable interaction styles such as sensitivity and responsiveness. However, recent literature suggests that directiveness can be impacted by different types of verbal directiveness such as follow-in directives (Haebig et al., 2013). Follow-in directives are statements that relate to an activity or object that the child is already focused on and conveys an expectation that the child changes their activity. The use of follow-in directives is linked to improved language outcomes in children with autism one year later (Haebig et al., 2013). Thus, directiveness has distinct effects on child outcomes depending on the definition of directiveness employed. This raises a question of whether directiveness should be considered as a single construct and highlights the importance of how global variables are defined.

Limitations of the study design must be considered when interpreting these findings. The study sample was affected by attrition at multiple time points of assessment

and participants in the EL group were more likely to withdraw. The infant's gestational age, gender, number of siblings, and the mother's age, household income and maternal AQ scores were observed and compared between the completer and non-completer groups. No significant differences were found in any of the sample demographics when comparing the completers to the non-completers. This suggests that demographics and autism traits in mothers had no impact on whether the family would return to participate in the study at a later stage. The parent was less likely to return to take part in the later stages of the study if their infant displayed a distressed or negative state during participation. It could be that some element of the study such as the tasks or being in a laboratory environment prompted negative emotional responses in this group of infants.

A measure of development was not taken at the baseline period when the children were 6 months of age, only parent-child interactions were observed during this phase of data collection. Future research should ensure a baseline assessment is conducted at the time of the parent-child interaction.

The findings of this study are based on a relatively small sample. The use of non-parametric statistics decreases the statistical power of the study and possible covariates such as number of siblings cannot be controlled for. The results are therefore susceptible to false negatives and any sampling effect biases may be amplified; thus the findings must only be viewed as indicative. Due to the heterogeneous nature of autism, studies investigating infant sibling groups require relatively large sample sizes in order to sufficiently represent this cohort (Lombardo et al., 2019). Despite this limitation, the study's prospective repeated measures design helped elevate the statistical power of this study (Bakeman, 2005).

There were significant differences in the family demographics between the EL and TL groups such as maternal age, number of siblings and household income. These

differences may have influenced the results. In this study, mothers of children with EL were on average 2.42 years older than mothers of TL children. Findings from previous studies suggest that older mothers display higher levels of maternal supportiveness and closeness with their child (Barnes et al., 2014). Despite these differences, it is unlikely that this age gap is significant enough to elicit these characteristics, which were between mothers in their teens and early twenties and older mothers in their thirties in the Barnes et al. study. One factor likely linked to elevated maternal age is the difference in the number of siblings between groups. In this study mothers of EL infants had more children than mothers of TL infants. A greater number of siblings has been found to influence how mothers interact with their infant, and in turn has an effect on their children's development. The amount of parent-child quality time the mother can offer to each child decreases as the size of the family expands. This has been linked to lower scores on cognitive tests in younger siblings (Lehmann et al., 2018). Additionally, infants with EL of autism could learn maladaptive behaviours from their diagnosed older sibling, such as non-functional repetitive behaviours (Christensen et al., 2010). Despite significant differences in household income, the median income for EL and TL groups both fall into high income brackets. The families participating in the study generally come from high resource households, which limits the generalisability of results.

Only a small number of children in this sample exhibited high levels of autism symptoms and received a score of 7 or more on the ADOS-G at 24 months of age. None of the children had a formal clinical diagnosis of autism. Seven out of 57 (12.28%) children had high levels of autism symptoms identified using the ADOS-G, with six of the children from the EL group and one child from the TL group. Due to the small number in the high autism symptom group, any group differences that emerge as early signs of autism may have been dampened in the EL group. Further analyses could not be

carried out to see if parent-child interactions and developmental outcomes could predict membership in high versus low autism symptom groups. An explanation for the low level of autism symptoms found could be attributed to relatively equal numbers of males and females in the study. Boys are three times more likely to be diagnosed with autism, and it is harder to measure autism symptoms in girls at a young age (Loomes et al., 2017). It may have been too early to measure autism symptoms using the ADOS-G assessment at 24 months of age, particularly among girls. Given the period of recruitment for the study was before the publication of the ADOS-2 (Lord et al., 2012) and DSM-5 (APA, 2013), the ADOS-G (Lord et al., 2000) was used to measure the level of autism symptoms in the sample and the siblings diagnosed with autism were diagnosed with reference to the DSM-IV (APA, 1994). Future studies should make use of the most up to date measures to ensure the evaluation of autism symptoms and autism diagnosis in sibling probands is reliable.

Despite these limitations, the study has many strengths including its longitudinal design, the assessment of development at two time points, and the observation of developmental outcomes by subdomain. The majority of studies that observe the relationship between parent-child interactions and developmental outcomes report only the ELC score of the MSEL. To our knowledge, this study is one of the first to examine global qualities of parent-child interaction and their relationship to specific developmental subdomains in children with an EL or TL of developing autism.

Study 2a

NE and Joint Attention during Parent-Child Interaction

Introduction

Joint Attention (JA) is a social and communicative event where two or more individuals share their attention towards an activity or object (Bakeman & Adamson, 1984; Yu & Smith, 2013). JA is initiated when an individual directs the attention of a social partner using verbal or non-verbal cues such as pointing or switching their gaze towards an object or event (Schertz et al., 2018). When responding to JA, one member of the dyad follows the gaze, gesture or verbal cue of their communicative partner (Adamson et al., 2010).

JA during parent-child interactions has been studied widely in dyads with typically developing children as well as those in clinical groups (Adamson et al., 2019; Gaffan et al., 2010; Mateus et al., 2019). Studying JA not only allows us to examine children's social cognition, but it also allows us to investigate its impact on child developmental outcomes and how parental behaviours can help facilitate development.

Language and cognitive abilities, alongside the use of eye gaze and gestures, are important skills required by a child to initiate and respond to instances of JA with their parent. In typically developing children, JA skills begin to develop between 6 and 12 months of age and advance through early childhood as children improve their language and social cognition (Charman, 2003; Deak et al., 2013). By the third year of life, children learn to initiate social interactions and cooperate with parental requests, thus improving children's early social development (Olsen & Spelke, 2008).

Child development is facilitated through shared experiences with their parents. JA has therefore been found to optimise a child's capacity to gain knowledge from a social interactive environment and is positively linked with a number of developmental abilities

in children such as language skills (Salo et al., 2018; Tomasello & Todd, 1983), social cognition (Mundy & Newall, 2007; Nelson et al., 2008), visual attention (Striano & Stahl, 2005) and academic success (Mundy & Newall, 2007).

Joint Attention in Clinical Groups

Research indicates that children in clinical groups exhibit characteristic differences in JA due to the potential impact of their condition on skills such as attention shifting and social referencing (Spiker et al., 2002). Examining JA in clinical groups allows us to gain insight into how particular deficits might impact child development, but also it can demonstrate how JA can help facilitate child outcomes, particularly in children at risk of developmental delay.

For example, as a result of preterm birth, children's developmental outcomes can be adversely affected. The extent of developmental delay depends on factors such as gestational age, birth weight and the presence of medical complications during the neonatal period (Aarnoudse-Moens et al., 2009). Preterm children show deficits in attention (De Schuymer et al., 2012), they fixate on stimuli for shorter periods of time (Telford et al., 2016) and appear to be less efficient at initiating, disengaging and shifting their attention compared to full-term children (Van de Weijer-Bergsma et al., 2008). Full-term children respond to bids for JA more frequently than those born preterm (De Schuymer et al., 2011) and are more likely to follow the gaze of an experimenter (Imafuku et al., 2017).

Children diagnosed with autism encounter challenges with communication, social interaction and display restrictive or repetitive behaviours (APA, 2013). Behaviours characteristic to those diagnosed with autism are observed to directly impact the ability to take part in JA including deficits in attention shifting and repetitive behaviours (Elsabbagh et al., 2013; Hudry et al., 2013). Difficulties with attention shifting in children

with autism may impede the child's ability to redirect their attention to parental JA bids, leading to an increase in missed bids and a reduction in the quantity of JA in this clinical group. Repetitive behaviours in children with autism are also related to a reduction in the time spent in JA during parent-child interaction. Children with autism are less able to sustain JA as repetitive behaviours can disrupt attentional focus, and parents are less able to read the child's behavioural prompts and interests (Hudry et al., 2013).

During the neonatal period, preterm children and their parents share similar experiences as those born with NE as both groups will have spent time in the NICU. In the NICU, infants are exposed to bright lights and high frequency sounds during a critical stage of development (El-Metwally & Medina, 2020). Due to these conditions, prolonged periods of stay in the NICU is associated with poorer cognitive and motor outcomes when children are followed up at 9 and 24 months of age (Subedi et al., 2017). Admission to the NICU is also a distressing experience for parents and is linked with high levels of parental stress and anxiety causing shifts in parenting behaviours (Treyvaud et al., 2019). The developmental outcomes of children born preterm are also somewhat similar to those born with NE. Children born preterm are at an increased risk of cognitive, language (Ionio et al., 2016), motor (De Kieviet et al., 2009) and attention difficulties (De Schuymer et al., 2012) which has an observed effect on the aquisition of JA skills (You et al., 2019). Additionally, those born with NE have an elevated likelihood of autism (Badawi et al., 2006; Chalak et al., 2018). Patterns of JA behaviours in these clinical groups may therefore be comparable to those born with NE.

Neonatal Encephalopathy and Joint Attention

Despite the potential impact of NE on the parent-child dynamic, JA has yet to be studied in dyads impacted by the disorder. The adverse outcomes associated with NE means that children born with NE and their mothers might show differences in JA during

parent-child interaction when compared to mothers and their children who do not experience complications during birth. The impact of clinical outcomes associated with NE on the child, combined with the experience of a traumatic birth on the mother could impact the relationship between the dyad, subsequently modifying aspects of JA.

Similar to those born preterm, NE is a major risk factor for children's development. It negatively impacts children's cognitive (Finder et al., 2019), language (Chin et al., 2019) and motor development (Jary et al., 2019). Impairments in attention (Tonks et al., 2019) and behavioural problems have also been found to be related to NE (Edmonds et al., 2020), with difficulties persisting into adolescence (Lindström et al., 2006). Whilst the cognitive, language and motor outcomes of children born with NE are widely studied, the development of social-cognition and social-emotional skills receive less attention. The challenges that these developmental deficits might impose on the child's social environment have yet to be considered.

Differences in the development of children born with NE may have an impact on their ability to engage in JA during parent-child interactions. The ability to initiate and maintain JA with a social partner requires the recruitment of a number of developmental skills (Saxon et al., 2000). For example, receptive and expressive language skills are vital for sustaining JA episodes as they allow the mother and child to communicate during the interaction (Adamson et al., 2019). Language allows the mother or child to initiate JA and to discuss the object or activity they are engaged in with their social partner (Hollich et al., 2000; Tomasello & Farrar, 1986). Motor skills are important as they allow the child to navigate their environment and manipulate objects of interest. They also allow the child to expand their communicative competencies by using gestures such as pointing or handing the mother objects of interest. However, children born with NE show language difficulties (Chin et al., 2019) and they may struggle with verbal turn-taking making JA

less engaging. This could lead to shorter JA episodes that are frequently terminated by the child. Due to issues relating to attention (Tonks et al., 2019), children born with NE may also find it harder to sustain episodes of JA. They may switch their attention more often, ending JA episodes more frequently or may miss their mothers' bids for JA at a higher rate. Children who show signs of motor delay are less likely to gesture compared to those with typical motor development (Longobardi et al., 2014). Thus, children born with NE may gesture less often or their bids for JA may be harder to recognise.

Mothers of children in clinical groups are more likely to exhibit directive behaviours (Green et al., 2013) which may lead to more attempts to initiate JA. In a qualitative study parents were interviewed about their experience of caring for a child born with NE (Herringhaus et al., 2013). Mothers reported that the prolonged separation and lack of contact during treatment in the NICU hampered the initial parent-infant bond. Parents describe their stay in the NICU to be extremely stressful as they feared their child might not survive or will be significantly affected by NE. Studies with other high-risk groups, for example, mothers of children born preterm, also found that they endured high levels of stress during the neonatal period (Forcada-Guex et al., 2011). During parent-child interactions, mothers who reported higher levels of stress during the neonatal period were more directive (De Jong et al., 2017) and used more active and overstimulating behaviours towards their child in later life (Forcada-Guex et al., 2011). The mean duration of JA episodes has been found to be shorter when mothers exhibit higher levels of controlling behaviour (Koşkulu, 2019).

Directive maternal behaviours may show different associations with JA depending on the nature of the directives used by the parent. Directive behaviours are typically characterised as negative and demanding behaviours that intend to disengage the child's attentional focus (Wan et al., 2017). Higher levels of intrusive behaviours and negative

comments are associated with reduced child engagement (Saint-Georges et al., 2011) and higher levels of negative affect (Steiner et al., 2018), thus compromising opportunities for JA. However, other studies have found maternal directiveness to facilitate higher levels of JA (Bottema-Beutel et al., 2018). Children in clinical groups have been found to be responsive to directive behaviours given that it is sensitive to the child's state (Doussard-Roosevelt, 2003). Although intrusive behaviours may disrupt the child's current state of play, a contingent bid from the parent is often followed by higher levels of JA, particularly when the bid is related to the child's current interests (Bottema-Beutel et al., 2018; Masur et al., 2013). In this case, mothers are adapting the interaction to facilitate high levels of JA. Directives help maintain the attention of the child and decreases the likelihood of the child disengaging from the interaction.

The Current Study

The current study investigated the quality and quantity of JA in dyads where the child was born with NE. They were compared to dyads where the child was born without complications. Due to the impact of NE on child and parent behaviours, differences in JA are expected to be observed between the NE and No-NE groups. The following hypotheses were investigated:

Hypothesis 1. It is expected that the total duration and mean duration of JA episodes will be shorter in dyads where the child was born with NE when compared to the No-NE group. However, mothers in the NE group will initiate JA episodes more frequently.

Hypothesis 2. The number of missed and terminated JA episodes will differ between the NE group and the No-NE group. Compared to children in the No-NE group, children born with NE are expected to terminate JA episodes more frequently and to miss maternal bids for JA more often.

Method

Participants

A total of 34 children aged between 48 and 66 months of age participated in the study with their biological mothers. The sample consists of two groups based on the child's NE status. Seventeen children were born with NE (NE group), and 17 were children born without complications (No-NE group). The No-NE group were selected by age using PSM (refer to chapter three for full details). There were no differences in age between the NE (M = 56.35, SD = 6.28) and No-NE group (M= 53.83, SD = 3.02; t = -1.49, p = 0.15) and a similar proportion of males and females were recruited in each group (X^2 = 0.47, p = 0.49). In the NE group, 10 children were male (55.6%) and seven were female (43.80%). In the No-NE group eight were male (44.4%) and nine were female (56.30%). All the mothers in this sample completed second-level education. Overall, 53.30% of mothers with children born with NE and 57.10% of mothers in the No-NE group had a Bachelor's degree. No difference in maternal education was observed between groups (X^2 = 7.11, p = 0.13).

Procedure

During their visit to the laboratory, parent-child interactions between the child and their mother were observed and recorded. The mother-child dyads took part in an episode of semi-structured play. During the semi-structured play interaction, the mother and child were given a bag containing a magnetic construction set. The dyad was left alone to play with the construction set and the interaction was recorded for five minutes. Each interaction was video and audio recorded using specialist recording equipment (see chapter three for technical details).

Measures

Joint Attention Coding. The recorded parent-child interactions were coded for qualitative and quantitative characteristics of JA on a second-by-second basis using a microanalytical coding scheme. Each 5-minute interaction was coded for JA using Mangold INTERACT software (Mangold, International, Arnstorf, Germany).

Episodes of JA were coded using the coding framework outlined by Ataman-Devrim et al. (2023). The coding scheme is adapted from studies of Koskulu et al., (2021a; 2021b), Bakeman and Adamson (1984) and Tomasello and Todd (1983). An episode of JA was identified and coded if (a) either the mother or child attempts to initiate an interaction with their partner, (b) the partner responds to the request within 2 seconds, and (c) both mother and child participated in the same activity or focused on the same object for at least 3 seconds. If the mother or child shifts their attention or disengages from the activity for more than 3 seconds, the JA episode is considered terminated.

Quantitative Features of JA. The frequency, total duration and average duration of JA episodes were calculated as indicators of the quantity of JA during each parent-child interaction. Frequency measures the total number of JA episodes that took place during the interaction. Total duration considers the total time spent in JA, and the average duration of JA episodes is calculated by dividing the total duration of JA by the frequency of JA that took place during the interaction.

Qualitative Features of JA. Specific qualities of each JA episode were also coded for the following: who initiated the interaction and who terminated the JA episode. It was also coded when a bid for JA was missed by either the mother or child.

***Who-Initiated*.** This code specifies the member of the dyad who instigated the JA episode. Either the mother or the child may initiate a bid for JA for example, by pointing to an object of interest. The code is applied when the act to initiate JA begins and finishes

when the behaviour ends. JA episodes are considered child-initiated if the child acts to initiate JA using verbal and/or non-verbal behaviours towards the mother. There are two types of parent-initiated codes including parent-followed and parent-redirected. A JA episode is coded as parent-followed if the mother acts to initiate JA based on an activity or object on which the child is already focused. The episode is considered parent-redirected if the mother initiates an episode of JA using an object or activity of which the child is not currently aware.

Who-Terminated. This code specifies which partner terminates the JA episode. It is applied when the child or mother shifts their attention or disengages from the mutual activity for more than 3 seconds. The interaction is coded as 'Child-terminated' if the child ends the period of JA by shifting their attention away from the mutual activity. If the mother disengages or shifts their attention, it is coded as 'Parent-terminated'.

Missed JA bid. When the child or mother attempts to initiate JA but the social partner fails to respond within two seconds of the bid, a JA episode has been missed. When the parent attempts to initiate JA but the child does not respond within 2 seconds the interaction is coded as 'Child-missed'. If the child produces a bid for JA but the mother does not respond within 2 seconds, it is coded as 'Parent-missed'.

Inter-Rater Reliability. A trained research assistant coded each parent-child interaction for JA. Inter-rater reliability of the coding was established through independent double coding of 25% (n = 9) of randomly selected parent-child interactions. Double coding was completed by the author of the coding scheme. Neither of the coders were blinded to the clinical status of the child. Optimal to excellent inter-rater agreement was established between the coders on each JA characteristic for the NE and no-NE groups. Intra-class correlations for the NE group ranged between r = .93 to 1.00 (child-initiated = .93, parent-followed = .96, parent-redirected = .99, child-terminated = .94,

parent-terminated = .90, child-missed = 1.00, parent-missed = .96). Intra-class correlations for the no-NE group ranged between r = .88 to 1.00 (child-initiated = .99, parent-followed = .90, parent-redirected = .97, child-terminated = 1.00, parent-terminated = .88, child-missed = .94, parent-missed = 1.00). Any disagreements that arose were resolved through discussion.

Statistical Analysis

The analyses for this study were run using SPSS (Version 26). To satisfy the assumptions of parametric statistics, data were checked for normality using Shaprio Wilk's test and homogeneity of variance was checked using Levene's test. Levene's test statistics were not statistically significant for any of the JA variables confirming homogeneity of variances among each group. Independent samples t-tests were computed to compare the quantity and quality of JA between the NE and No-NE group. A Bonferroni correction was applied to reduce the chances of type I error when performing multiple comparison tests, therefore alpha criterion was adjusted to $p<0.01$.

Results

The Quantity of JA during Parent-Child Interaction

The frequency, total duration and average duration of JA episodes was compared between the NE group and No-NE group using independent sample t-tests. Overall, no differences were observed in the quantity of JA episodes in each group (see table 5.2.1.1).

The Quality of JA during Parent-Child Interaction

Specific qualities of JA were measured and compared between the NE and No-NE groups including who initiated the JA episode, the proportion of episodes terminated by the mother and child, and the proportion of JA episodes missed by the mother and child. Independent samples t-tests were computed and found no group differences in any of the qualities of JA (see table 5.2.1.1).

JA during Semi-Structured Play in Children Born with NE compared to a Typically Developing Group

Table 5.2.1.1. *Descriptive statistics of JA variables and independent samples t-test.*

	No-NE Group (n = 17)	**NE Group** (n = 17)	**Group Difference**
	M (SD)	*M (SD)*	*t-value, p-value*
JA Quantity			
Frequency	1.53 (.87)	1.58 (.87)	t = -0.20, p = 0.85
Total Duration (sec)	291.98 (11.88)	279.34 (31.30)	t = 1.56, p = 0.14
Average Duration (sec)	236.12 (89.61)	221.20 (93.39)	t = 0.48, p = 0.64
JA Quality			
Child-Initiated %	.30 (.44)	.42 (.42)	t = -0.80, p = 0.43
Parent-Followed %	.46 (.48)	.21 (.35)	t = 1.78, p = 0.09
Parent-Redirected %	.12 (.28)	.26 (.41)	t = -1.15, p = 0.26
Child-Terminated %	.25 (.41)	.41 (.48)	t = -1.06, p = 0.30
Parent-Terminated %	.16 (.34)	.18 (.35)	t = -0.12, p = 0.90
Child Missed %	.06 (.24)	.00 (.00)	t = 1.00, p = 0.33
Parent-Missed %	.00 (.00)	.00 (.00)	-

Note. %, proportion of total JA episodes

Discussion

The current study examined the quantity and quality of JA between children born with NE and their mothers compared to dyads with typically developing children during parent-child interaction. Overall, NE status was not found to be associated with any features of JA. Contrary to our hypotheses, no statistically significant differences were observed as the quantity of JA did not differ between the NE group and the No-NE group. Similarly, there were no differences in qualities of JA between the two groups such as who initiated JA, the proportion of JA episodes terminated by the mother and child, and the proportion of bids missed by either partner.

Previous research has demonstrated differences in JA when comparing those in other clinical groups to dyads with typically developing children (Kmita et al., 2014; Sansavini et al., 2015). However, some inconsistencies have been reported and other studies only observe differences in JA when additional factors such as the degree of medical risk and maternal characteristics are considered (Bottema-Beutel et al., 2018; De Schuymer et al., 2011; Egotubov et al., 2019; Mateus et al., 2019; Sansavini et al., 2015).

Mateus and colleagues (2019) conducted a meta-analysis of findings that compare the duration and frequency of JA episodes in children born preterm to full term children. It was concluded that preterm birth had a negative effect on JA, but this effect was observed only in those born extremely preterm (Sansavini et al., 2015). Infants born extremely preterm endure higher levels of clinical complications (Patel, 2016) which has been linked to emotional and behavioural problems in later life (Clark et al., 2008). Such behavioural problems may cause these infants to be more challenging social partners consequently compromising the quantity and quality of JA. It is therefore suggested that the degree of neonatal risk and complication must be considered when investigating JA in these clinical groups.

Similar to the outcomes of the current study, Egotubov et al. (2019) did not find a direct link between neonatal complications and characteristics of JA in later life. However, when additional variables such as maternal responsiveness, maternal anxiety and household chaos were taken into consideration, indirect effects were observed. These findings highlight the complex nature of the link between developmental risk factors and their impact on JA behaviours. The differences in JA among clinical groups may not be due to developmental delays alone. The bidirectional nature of JA and the influence of other environmental factors must also be considered (Coldwell et al., 2006; Forcada-Guex et al., 2011; Mendive et al., 2013).

Limitations

The following limitations must be considered when interpreting the findings of this study. The study is based on a small sample, therefore factors that may influence the outcome of the results such as the grade of NE sustained at birth could not be taken into consideration. Most participants in the current sample were diagnosed with moderate NE, a group with the greatest range in developmental outcomes (Van Handel et al., 2007). Further investigations need to consider the impact of NE grade on JA since children born with mild, moderate and severe NE may exhibit different behaviours during parent-child interaction.

The current study observed JA during instances of semi-structured play. However, differences in JA have been found when contrasting behaviours during free-play with semi-structured play episodes (Mateus et al., 2013). During semi-structured play mothers initiated more bids for JA, whereas children showed higher levels of responding and initiating behaviours during free-play (Mateus et al., 2013). It is suggested that semi-structured activities set the agenda and dominate the interaction (Gaffan et al., 2010).

Free-play interactions allow the child to take more initiative and might therefore provide a more representative portrayal of the child's JA capabilities.

Similar to the current study, other studies have investigated JA in preschool children at-risk of developmental delay (Dawson et al., 2004; Hudry et al., 2013; Leekam et al., 2000; Smith & Ulvund, 2003). However, the vast majority of literature that investigates JA typically recruit participants in infancy (Deak et al., 2013; Franchini et al., 2019; Mateus et al., 2019; Nyström et al., 2019). Due to recruitment constraints caused by COVID-19 it was not possible to recruit younger participants to this study. There may be value in investigating JA in infants born with NE so emerging differences from infancy to early childhood can be observed.

Although the selection of videos that were double coded was randomisied, neither of the coders were blinded to the clinical status of the child. Therefore, internal biases may have affected the coding of the parent-child interaction recordings. Whenever possible, neither of the coders should know which group each participant belongs to. Future studies should ensure that one or both of the coders are blinded to the clinical status of the child to ensure the coding is accurate and reliable.

Despite these limitations, this study has a number of strengths. This study uses a detailed coding scheme as both quantitative and qualitative features of JA were coded microanalytically. Although time intensive, this coding system identifies the occurrence of clearly defined behaviours on a second-by-second basis thus increasing the precision of behavioural observations, allowing us to measure JA objectively.

Most studies that investigate the development of children born with NE rely on test scores from standardised assessments of development (Marlow et al., 2021). Despite being widely investigated in other clinical groups, to the author's knowledge, this is the first study to investigate the behaviours of children born with NE and their parents during

parent-child interaction. In particular, few researchers have investigated JA during play in this clinical group.

Study 2b

Joint Attention and Developmental Outcomes in 3- and 4-year olds Born with NE

Introduction

Joint Attention (JA) is a multifaceted skill that develops gradually through early childhood. In typically developing children, the skills essential for JA begin to develop from early infancy and advance through childhood as children improve their communication skills and social cognition (Charman, 2003). The link between JA and children's developmental skills is bidirectional. JA plays an essential role in a child's communication, social interaction and cognitive development (Mundy & Gomes, 1998; Salo et al., 2018; Tomasello & Todd, 1983). Similarly, these skills are also required by the child to initiate and respond to instances of JA (see chapter two for a review of the literature).

Children require a suite of skills in order to initiate, respond to and sustain episodes of JA. Adequate social cognition allows children to identify social cues and bids for JA. The child is required to identify gestures, facial expressions, follow their parent's gaze and recognise tone of voice in order to take part in JA (MacPherson & Moore, 2017; Nyström et al., 2019). Similarly, children need to be able to communicate with their social partner in order to participate and sustain JA episodes. For example, the child might use gestures to point to an object of interest. Language is another vital communicative skill for JA as it allows the child to talk with the parent about the common object or event (Adamson et al., 2019). Back and forth communication helps sustain episodes of JA. To date, the majority of the literature investigates JA during infancy and JA is less frequently investigated in older children (Deak et al., 2013; Franchini et al., 2019; Mateus et al., 2019; Nyström et al., 2019).

Several developmental skills exhibited by 3-year-old and 4-year-old children have been identified to facilitate JA. From 18 months of age children's language abilities tend to accelerate. By the age of three, children are observed to have a larger vocabulary and can therefore form more complex sentences and ask detailed questions during JA (Gilkerson et al., 2017). At ages 3 and 4 years cognitive abilities also progress and children are found to have better memory, problem solving skills and can follow instructions with multiple steps (Miller & Goldsmith, 2017). Four-year-olds show higher levels of attentional focus (Fisher et al., 2012; Miller & Goldsmith, 2017) and can therefore engage in tasks for longer periods, thus extending the amount of time spent in JA. Compared to infants, 3- and 4-year-olds tend to have more refined gross and fine motor skills allowing them to better control their movements and navigate the environment (Kakebeeke et al., 2013; Muluk et al., 2014). For example, they are able to walk to another room to get an object of interest or manipulate small objects with a social partner. As children get older, they are also more likely to show sophisticated social cognition. Older children are better at sharing and turn taking during play (Warneken & Tomasello, 2013). They are able to regulate their emotions appropriately (Montroy et al., 2016) and start to understand other people's thoughts and feelings (Conte et al., 2019). Despite the clear development of these skills observed between infancy and childhood, JA is less commonly investigated in older children particularly in clinical populations where development might be compromised.

Neonatal Risk and Joint Attention

Infants who endure complications at birth are at-risk for developmental delays when they are older. Previous research has established that neonatal risk factors such as preterm birth, low-birthweight and medical problems are associated with atypical social communication (De Groote et al., 2006; De Schuymer et al., 2011; Landry et al., 1990;

Zmyj et al., 2017). These factors have been linked to reduced initiating and responding to JA (Garner & Landry, 1992; Zmyj et al., 2017). An early study by Poehlmann and Fiese (2001) established that parent-child interactions mediated the relationship between neonatal risk and cognitive development in low birthweight infants. It was found that reciprocal and engaging interactions predict higher cognitive scores in this clinical cohort. More recent studies have found a combination of neonatal risk and environmental factors specific to these high-risk groups, such as maternal anxiety and household chaos, to negatively impact JA during parent-child interaction (Egotubov et al., 2020). The degree of medical risk endured during the neonatal period appears to play a role in the development of JA (Poehlmann & Fiese, 2001). Higher levels of neonatal risk are typically associated with developmental delay (Trembath et al., 2016; Poehlmann & Fiese, 2001) which may play a part in the difficulties children experience with JA. When children in clinical groups experience developmental delay, they may find it difficult to initiate, engage in or respond to bids for JA. Therefore, children in these groups have fewer opportunities to practice and develop their skills because they are less likely to take part in JA with a social partner.

NE is linked with deficits in a number of developmental domains such as cognitive (Finder et al., 2019; Schreglmann et al., 2020), language (Herrera et al., 2017; Murray et al., 2009), motor (Van Kooij et al., 2008) and attention difficulties (Tonks et al., 2019). Children born with NE who display cognitive deficits may find tasks difficult to follow and may therefore feel less inclined to take part in a joint activity. This might lead to difficulties with sustaining JA as the child may disengage from JA more frequently. When children's expressive and receptive language skills are negatively affected by NE, they will find it difficult to initiate, respond to and sustain JA. The child may find it harder to communicate with a social partner which reduces the opportunity for

back-and-forth conversation that facilitates JA. If the child's receptive language skills are poor, they may find it difficult to perceive the social partner's communicative intent and the child may miss bids for JA more often or disengage from JA more frequently. Motor delay in children might compromise JA as children may navigate to stimuli less frequently and find it harder to manipulate objects of interest. Parents may also find it harder to understand the child's intent as their gestures might be harder to read. Attention difficulties are prevalent in low birthweight infants (Groen-Blokhuis et al., 2011), those born preterm (Anderson, 2014) and in those born with NE (Tonks et al., 2019; Spencer et al., 2021). Poor attention is likely to have a negative impact on JA as children are more likely to miss their parent's bids for JA. Children might also redirect their attention more frequently making it difficult to sustain JA for longer periods of time.

Few researchers have investigated the relationship between developmental outcomes and JA, particularly in children born with NE. To date, most JA research has concentrated on typically developing children and those in other clinical groups such as those diagnosed with autism (Bottema-Beutel et al., 2018; Charman, 2003; Franchini et al., 2019) or those born preterm (De Schuymer et al., 2011). Infants diagnosed with NE may experience a number of medical complications at birth. NE is a heterogeneous disorder and a number of problems that occur during the antenatal and intrapartum periods have been linked to long-term neurodevelopmental delay (Ahearne et al., 2017; Glass et al., 2018; Jenster et al., 2014; Polat et al., 2013). Despite the impact of NE on child outcomes, characteristics of JA in this cohort have yet to be explored. Due to the bidirectional nature of JA and developmental outcomes, there may be value in studying the relationship between JA and developmental functioning in children born with NE. In doing so, it may help us better understand the development of children born with NE, and whether characteristics of JA facilitate the development of children in this clinical cohort.

The Current Study

The current study investigated the quality and quantity of JA in 3-year-old and 4-year-old children born with NE. The following hypotheses were investigated:

Hypothesis 1. It is expected that the quantity and quality of JA will differ between the 3- and 4-year old children born with NE. Four-year olds will engage in fewer JA episodes of longer duration than 3-year olds. Four-year-olds will initiate JA more frequently than the 3-year-olds, whereas the 3-year olds will miss more bids for JA than the 4-year-olds.

Hypothesis 2. Associations between JA and developmental outcomes will be observed. Less frequent JA episodes that are longer in average duration will be associated with better language and processing speed skills in children. A higher proportion of child terminated and missed JA episodes will be associated with poorer language and processing speed abilities in children born with NE.

Method

Participants

Thirty children born with NE and their mothers took part in the current study. Twelve children aged 36.17 to 46.37 months completed the Bayley-III assessment and 18 children aged 48.10 to 69.40 months completed the WPPSI-IV assessment. A larger proportion of males born with NE took part in this study (66.67%) compared to females (33.33%). The mothers taking part in this study were aged between 24 and 45 years (M = 35.96, SD = 5.79) and all had completed second-level education. Three mothers had a secondary certificate (10.00%), six had completed a diploma (20.00%), 13 mothers obtained a Bachelor's degree (43.33%) and five had a postgraduate degree (16.67%). No difference in maternal education was observed between groups (X^2 = 4.94, p = 0.29).

Procedure

Mothers and their newborn infants were approached to take part in the study if the infant was diagnosed with NE (further information on participant recruitment is outlined in chapter three). Clinical data were collected from the infants throughout their stay in the neonatal unit. Additional data were collected from medical records in order to calculate a neonatal risk index.

When the children turned 3 years of age or older, the families were re-recruited to take part in a follow-up study that took place at the Infant and Child Research Laboratory, Trinity College Dublin. During their visit to the laboratory the mother and child took part in an episode of free-play and the parent-child interaction was observed and recorded. The dyad was given a box containing a number of toys including building blocks, a ball, a Mr Potato Head kit, toy vehicles and animals. The mother was asked to play with their child as they would at home and the interaction was recorded for five minutes (see chapter three for technical details of the recordings).

After taking part in the parent-child interaction each child completed a standardised assessment of development. The measure administered depended on the age of the child at the time of assessment. Children aged 36 to 46 months of age were administered the Bayley-III and children aged 48 months and older were administered the WPPSI-IV.

Measures

Neonatal Risk. The clinical presentation of infants born with NE is variable particularly in those diagnosed with moderate NE. The majority of participants who took part in this study (73.33%) was diagnosed with moderate NE, therefore a neonatal risk index was created based on previous research with NE infants (Ahearne et al., 2017; Ambalavanan et al., 2006; Beligere & Rao, 2008; Glass et al., 2018; Jenster et al., 2014;

Twomey et al., 2010). Clinical data were collected for each infant during their time in the NICU and were used to compute a risk score for each infant. One point was given for the presence of each of the following nine risk factors: an abnormal MRI result, the presence of seizures, a 5 minute Apgar score less than 7, a cord blood pH less than 7.2, whether the infant was intubated, lack of suck and feeding problems, the presence of maternal infection and if the infant was hospitalised for more than ten days. Another point was given if delivery complications arose such as fetal bradycardia, meconium aspiration, maternal haemorrhage, placental abruption or nuchal cord complications. Neonatal risk was measured on a scale ranging from 0 to 9 points; higher scores indicate the presence of more neonatal risk factors. Similar scales have been developed to measure neonatal risk in other clinical groups such as low birth weight infants (Poehlmann & Fiese, 2001) and those with an elevated likelihood of autism (Stein et al., 2006). Such risk indices have been found to predict future cognitive outcomes (Poehlmann & Fiese, 2001) and the quality of parent-child interactions (Fiese et al., 2001).

Joint Attention Coding. The five-minute parent-child interaction recordings were coded for qualitative and quantitative qualities of JA on a moment-by-moment basis using a microanalytic coding scheme. Each free-play session was coded for JA using Mangold INTERACT software (Mangold, International, Arnstorf, Germany). Details of the full coding scheme is outlined in chapter five study 2a. The current study also considered the type of JA used which is described below.

Quantitative Features of JA. The frequency, total duration and average duration of JA episodes were calculated as indicators of the quantity of JA during each parent-child interaction.

***Qualitative Features of JA*.** Select qualities of JA were also coded including: who initiated the JA episode, who terminated the interaction and if either the mother or child missed a bid for JA.

Inter-Rater Reliability. Each parent-child interaction was coded for JA by a trained research assistant. Inter-rater reliability of the coding was established through independent double coding of eight (25%) randomly selected parent-child interactions. Optimal to excellent inter-rater agreement was established between the coders on each JA characteristic, intra-class correlations ranged between r = 1.00 to .89 (child-initiated = .93, parent-followed = .94, parent-redirected = .89, child-terminated = .91, parent-terminated = .90, child-missed = 1.00, parent-missed = 1.00). Any disagreements that arose were resolved through discussion.

Developmental Assessment. All methods and procedures used for the assessment of child developmental outcomes including the administration of language assessments using the Bayley-III are detailed in chapter three. As detailed in chapter four, when compared to those born without complications, children born with NE performed significantly worse on the language scales of the Bayley-III, and the verbal comprehension and processing speed sub-domains of the WPPSI-IV. Due to the importance of language skills and processing speed during JA, these domains are likely to have the most influence on JA and were therefore selected for these analyses.

When assessing language comprehension in the 4-year-old children, two tasks from the WPPSI-IV were administered including the 'Information' and 'Similarities' tasks. The Information task involves asking the child a series of general knowledge questions such as "How many eyes do you have?" and "Tell me the name of a vegetable?". During the Similarities task the child is asked to verbally identify similarities between common objects and concepts. The child is told: "Finish what I say, red and

yellow are both…?" "Cars and trucks are both…?". As the child progresses through each of the tasks the questions increase in difficulty. Each task is discontinued if the child gives three incorrect answers in a row. The score on each task is calculated by adding up the number of correct responses.

Two tasks in the WPPSI-IV scale were used to assess processing speed including the 'Bug Search' and 'Cancellation' tasks. During the Bug Search task, the child was given an 11-page response booklet (excluding a demonstration and practice page) to complete. Each page included six rows of bug pictures containing various types of bugs such as a worm, butterfly, centipede, dragon fly, or snail. At the start of each row a particular bug is pictured in a grey box and is followed by five different bugs including a duplicate of the bug in the grey box. The aim of the task is to use a stamp pen to stamp the bug seen in the grey box. After explaining the instructions to the participant and completing a demonstration and practice run, the child is given 120 seconds to search for as many bugs as possible in the booklet. The final score is calculated by the number of correct answers minus the number of incorrect answers. The second task used to assess processing speed is the cancellation task. The child is given another response booklet consisting of two A3 sheets of paper. The pages each contain 192 pictures of various items such as a dog, scissors, tree, airplane, trousers, t-shirt, socks, dungarees and so on. The child is given a stamp pen and is told to only stamp things that people wear. After a demonstration and practice round the child is given 45 seconds per sheet to stamp as many items of clothing as possible. The total score on this task is calculated by counting the number of correct answers given and subtracting the number of incorrect answers for each page.

Statistical Analysis

The analyses for this study were run using SPSS (Version 26). Data were checked for normality using Shapiro Wilk's test and homogeneity of variance was checked using Levene's test. The JA data violated the assumptions of normality and equal variance, thus non-parametric tests were used to conduct the analyses. Mann-Whitney *U* tests were computed to compare the quantity and quality of JA between the 3-year-old and 4-year-old children. Kendall's Tau correlation coefficients were calculated to analyse the relationships between JA characteristics and child developmental outcomes. Due to the number of comparison tests performed alpha criterion was adjusted to $p<0.01$ to reduce the chances of type I error. However, the alpha level remained at 0.05 for the correlation analyses to reduce the chance of type II error.

Results

JA in 3-year-old compared to 4-year-old children born with NE

The quantity and quality of JA was compared between 3-year-old and 4-year-old children born with NE using a Mann Whitney *U* test. The frequency, total duration and average duration of JA was measured to compare the quantity of JA during free-play interactions in each age group. No differences were observed in the quantity of JA episodes (see table 5.2.2.1).

Specific qualities of JA were measured and compared between the 3-year-olds and 4-year-olds including the type of JA used, who initiated the JA episode, the proportion of episodes terminated by the mother and child, and the proportion of JA episodes missed by the mother and child. Mann Whitney *U* tests found no differences in any of the qualities of JA between the two age groups (see table 5.2.2.1).

Relationship between JA and Developmental Outcomes

Kendall's tau correlation coefficient was used to analyse the relationship between JA during parent-child interaction, neonatal risk and developmental outcomes in the 3-year-olds and 4-year-olds born with NE. Correlations were tested between JA, neonatal risk and receptive and expressive language scores from the Bayley-III assessment when the children were 3 years of age. Correlations were tested between JA, neonatal risk and verbal comprehension and processing speed scores from the WPPSI-IV in the 4-year-old cohort.

No significant associations were observed between JA and language outcomes in children at 3 and 4 years of age (see table 5.2.2.2). However, associations between qualities of JA and processing speed were observed in the 4-year-olds (see table 5.2.2.3). Analyses revealed a negative relationship between parents' initiating JA through following and the child's processing speed abilities. The child terminate and parent terminate variables were mutually exclusive and a negative relationship was observed between the child terminating JA and processing speed abilities (i.e. the child terminating JA is associated with poorer processing speed). Therefore, a positive association between parents terminating JA and processing speed was also observed. Additionally, a relationship between neonatal risk scores and processing speed abilities was observed such that higher levels of neonatal risk were associated with poorer processing speed abilities in children born with NE.

JA during Free-Play in 3-year-old and 4-year-old Children Born with NE

Table 5.2.2.1. *Descriptive statistics of JA variables and Mann-Whitney U tests.*

	3 years (n = 12)	**4 years** (n = 18)	**Group Difference**
	M (SD)	*M (SD)*	*U, p-value*
JA Quantity			
Frequency	7.50 (2.61)	5.78 (2.76)	$U = 69.00$, $p = 0.10$
Total Duration (sec)	236.64 (33.15)	245.37 (33.94)	$U = 91.00$, $p = 0.47$
Average Duration (sec)	40.52 (34.88)	66.21 (67.29)	$U = 0.69$, $p = 0.10$
JA Quality			
Child-Initiated %	.39 (.15)	.44 (.23)	$U = 90.00$, $p = 0.45$
Parent-Followed %	.23 (.12)	.30 (.25)	$U = 93.50$, $p = 0.54$
Parent-Redirected %	.38 (.17)	.26 (.17)	$U = 66.00$, $p = 0.08$
Child-Terminated %	.66 (.30)	.62 (.34)	$U = 103.00$, $p = 0.83$
Parent-Terminated %	.34 (.30)	.38 (.34)	$U = 103.00$, $p = 0.83$
Child Missed %	.63 (.43)	.33 (.49)	$U = 72.00$, $p = 0.09$
Parent-Missed %	.13 (.23)	.11 (.32)	$U = 96.00$, $p = 0.63$

Note. %, proportion of total JA episodes.

The relationship between neonatal risk, JA and language outcomes in 3-year-old children born with NE

Table 5.2.2.2*. Correlations between the variables.*

		1	2	3	4	5	6	7	8	9	10	11	12	13
1	Neonatal Risk	1.												
2	Rec Lang	.11	1.											
3	Exp Lang	.11	.74**	1.										
4	JA Freq	.26	.37	.46	1.									
5	JA Dur	-.26	.00	.02	-.22	1.								
6	JA Ave Dur	-.26	-.26	-.38	-.81**	.42	1.							
7	C-init %	-.16	.02	.20	.46	-.14	-.41	1.						
8	P-follow %	.05	.36	.38	-.06	.64**	.18	-.14	1.					
9	P-redic %	.05	-.23	-.35	-.19	-.36	.09	-.44	-.42	1.				
10	C-term %	.00	.17	.31	.13	.50	-.06	.27	.47	-.63**	1.			
11	P-term %	.00	-.17	-.31	-.13	-.50	.06	-.27	-.47	.63**	-1.00**	1.		
12	C-miss %	.25	.53	.40	.28	.09	-.31	.26	.31	-.53	.34	-.34	1.	
13	P-miss %	.43	-.35	-.34	.05	-.21	-.07	.10	-.26	.12	-.29	.29	-.26	1.

Note. Rec Lang, Receptive Language; Exp Lang, Expressive Language; Freq, frequency; Dur, duration; Ave Dur, average duration; %, proportion of JA episodes; C-initi, child initiated; P-follow, parent followed; P-redic, parent redirected; P-term, parent terminated; C-term, child terminated. **p ≤ 0.01 (two-tailed).

The relationship between neonatal risk, JA, cognitive and language outcomes in 4-year-old children born with NE

Table 5.2.2.3*. Correlations between the variables.*

		1	2	3	4	5	6	7	8	9	10	11	12	13
1	Neonatal Risk	1.												
2	VCI	-.40	1.											
3	PSI	-.60**	.35	1.										
4	JA Freq	.01	.31	-.05	1.									
5	JA Dur	-.21	-.08	-.08	-.50**	1.								
6	JA Ave Dur	-.06	-.25	.05	-.91**	.61**	1.							
7	C-init %	-.25	.37	.18	.32	-.24	-.32	1.						
8	P-follow %	.31	-.32	-.58**	-.28	.41	.27	-.36	1.					
9	P-redic %	-.25	.13	.39	.03	-.11	-.03	-.32	-.34	1.				
10	C-term %	.24	-.18	-.57**	.09	.13	-.06	.03	.46	-.48**	1.			
11	P-term %	-.24	.18	.57**	-.09	-.13	.06	-.03	-.46	.48**	-1.00**	1.		
12	C-miss %	-.35	.25	.34	.19	-.29	-.21	.09	-.32	.36	-.13	.13	1.	
13	P-miss %	.33	.03	.13	.18	-.46	-.23	.35	-.43*	-.12	-.28	.28	-.25	1.

Note. VCI, Verbal Comprehension Index; PSI, Processing Speed Index; Freq, frequency; Dur, duration; Ave Dur, average duration; %, proportion of JA episodes; C-initi, child initiated; P-follow, parent followed; P-redic, parent redirected; P-term, parent terminated; C-term, child terminated. **$p \leq 0.01$ (two-tailed).

Discussion

As JA is robustly linked to children's developmental outcomes, this study examined the relationship between the developmental functioning of children born with NE and the quantity and quality of JA at 3 and at 4 years of age. Overall, no differences in the characteristics of JA episodes were observed between dyads including 3-year-old and 4-year-old children. However, associations between qualities of JA, neonatal risk and children's processing speed abilities were observed in the 4-year-old cohort. Children born with NE who endure more complications at birth are likely to perform worse on the processing speed domain of the developmental assessment. Processing speed abilities are also found to be negatively associated with qualities of JA. As expected, NE children with lower processing speed scores terminated JA episodes more frequently. Additionally, mothers of children with poorer processing speed abilities used a higher number of parent-followed bids to initiate JA.

Contrary to our first hypothesis no differences in JA were observed between the groups of 3 and 4-year-old children born with NE. Despite the progression in developmental abilities between 3- and 4-year-old typically developing children, no studies to date have investigated differences in JA between the two age groups. The majority of JA literature focuses on the study of JA in infancy (Deak et al., 2013; Franchini et al., 2019; Mateus et al., 2019; Nyström et al., 2019). Therefore, it is difficult to establish whether the patterns of JA exhibited by children born with NE in this study differ from or follow the normative developmental trajectory.

Although there was no relationship between the quantity of JA and language outcomes as outlined in hypothesis two, associations between qualities of JA and processing speed abilities were observed in our cohort of children born with NE. As expected, poorer processing speed scores in children were related to a higher number of

JA episodes terminated by the child. Information processing skills have been linked to JA in other clinical groups. A study on children with an elevated likelihood of autism found information processing difficulties to be a marker for poor JA abilities when compared to typically developing children (Mundy et al., 2016). Processing speed is an important cognitive skill that considers the speed at which an individual processes information in order to quickly perform tasks (Fry & Hale, 2000). Assessments that measure processing speed capture the rate at which the participant completes a basic cognitive task such as stimuli identification or discrimination. In the case of this study, the processing speed tasks were completed as part of the WPPSI-IV assessment (Wechsler, 2013).

A number of developmental skills have been found to facilitate processing speed including attention, memory and executive function skills (Fry & Hale, 2000; Gordon et al., 2018; Mulder et al., 2011). Weaknesses in these areas might also contribute to children ending JA episodes more frequently. In order to process information, the ability to sustain attention and focus on a task is important. Children who show difficulties with attention find it difficult to process information quickly and may therefore lose interest and disengage from JA more frequently (Mundy, 2016). Attention (Lindström et al., 2006; Tonks et al., 2019) and memory problems (Van Handel et al., 2012) are widely reported in children born with NE therefore it is a likely explanation for the poorer processing speed scores in this cohort.

The findings of this study also demonstrate that the type of initiation used by the mother to instigate JA is linked to the child's processing speed skills. Parent-initiated JA episodes were identified as either parent-followed or parent-redirected. When a mother initiates JA using an object or activity the child is already focused on, it is labelled parent-followed. If a mother initiates JA using stimuli the child is not currently focussed on it is considered parent-redirected. Overall, mothers of 4-year-old children with poorer

processing speed scores used a higher number of parent-followed bids to successfully initiate JA. Parent-followed bids are considered contingent as the mother initiates JA based on the current interests of the child. Previous research has demonstrated that levels of JA are increased when adult play behaviour closely follows the interests of the child (Lewy & Dawson, 1992; Masek et al., 2021).

This relationship between contingent responsiveness and JA has been observed in both typically developing children and those in clinical groups. In particular, children in clinical groups are found to be more responsive to parent-followed bids. A study by Bottema-Beutal and colleagues (2018) found associations between parent-followed utterances and JA to be stronger in dyads where the child has a diagnosis of autism compared to typically developing children. JA bids allow the individual to share topics and activities with a social partner. Contingency allows the dyad to sustain the interaction using back-and-forth exchanges that impact the longevity of a JA episode (Bakeman & Adamson, 1984). It is suggested that children in clinical groups are more likely to respond to parent-followed bids as children typically orientate towards objects and activities that are within the child's developmental range of understanding. Children are less likely to respond if a bid is above their developmental level (Mahoney & Neville-Smith, 1996). Children with poorer processing speed skills might find it difficult to focus their attention or will reject bids for JA if they are not interested in the stimuli presented to them. However, if the child is familiar with an object or activity, they may already understand the function of the object or objective of the task they are interested in. Therefore, children born with NE are more likely to be responsive to a JA bid that is related to their current interest or focus.

Higher levels of neonatal risk were associated with poorer processing speed abilities in children born with NE at age 4. However, neonatal risk did not show a

significant relationship with qualities or the quantity of JA. Thus, the link between neonatal risk, processing speed and JA could not be further investigated. Some studies have found children born with NE to have slow reaction times and attention difficulties (Spencer et al., 2021; Tonks et al., 2019). However, these findings are not consistently observed during infancy. A study by Zorn et al. (2018) found processing speed in young infants born with NE to be preserved. This suggests that processing speed difficulties are not necessarily present from birth but may develop over time. More research is necessary to enhance understanding about the processing speed abilities in children born with NE and how they may impact the overall developmental functioning of those in this clinical cohort.

Limitations

Despite the findings of this study, the following limitations must be considered when interpreting the results. The current study only included dyads with children born with NE and has not taken into consideration dyads with typically developing children. Future research should compare the relationships between developmental outcomes and JA in both groups in order to establish whether the associations observed in this group of children born with NE differ from or follow the normative developmental trajectory. Comparing the outcomes of children born with NE to typically developing children allows us to establish differences between the two groups. In doing so, we can determine what aspects of behaviour and JA are impacted by NE and how the disorder influences overall development.

This study established a link between parent-followed bids and children's processing speed skills. However, the current study did not take into account the types of initiation used by the parent in the JA bids missed by the child. Therefore, it could not establish whether children with poorer processing abilities missed redirected bids more

frequently. Following on from the findings of this study, it is presumed that children with poorer processing speed skills may miss parent-redirected bids for JA more frequently. Further studies are needed to investigate the influence of parent-redirected bids and the likelihood of the children born with NE to miss or ignore JA bids.

Processing speed in children born with NE was measured in the 4-year-old children but not in the 3-year-old cohort. Therefore, we could not establish whether the relationship between processing speed and JA is consistent throughout early childhood. The developmental assessment administered to the 3-year-old cohort does not take into consideration processing speed. Future studies may also want to assess the relationship between child processing speed and qualities of JA in children born with NE at 3 years of age and younger to see if the association is also present in early childhood.

The sample size of this study was small and therefore its statistical power is limited. The study was impacted by high levels of attrition from the time of birth to follow-up. Issues with recruitment such as restrictions associated with the COVID-19 pandemic also negatively impacted the sample size of this study. Future studies should consider replicating this research with a larger sample size in order to increase statistical power. The power of the study could also be improved if a longitudinal study design was adopted and the participants were assessed at both 3 and 4 years of age.

Despite these limitations, this study looks to be the first to investigate the relationship between JA and child outcomes in dyads where the child is born with NE. The influence of parent and child behaviours has been widely studied in other clinical cohorts but had yet to be examined in those born with NE. In spite of the negative impact of NE on developmental outcomes (Finder et al., 2019; Robertson et al., 1993), not many studies have looked at JA and how it may facilitate or hinder child development. Patterns of behaviours exhibited during JA that are identified as beneficial, such as the use of

parent-followed bids to encourage play, can inform the development of interventions for children affected by NE. These findings allow us to provide parents with simple yet effective advice that can help improve the outcomes of children.

Study 3

Child Directed Speech and Child Speech during Parent-Child Interaction

Introduction

Whilst we are primarily interested in constructs that are inherently dyadic, we must also consider the combination of individual behaviours that shape the dyadic interaction. Speech that occurs during parent-child interactions is shaped by reciprocal processes. When parents communicate with their children, they systematically alter features of their speech to get the attention of their child, and to ensure the child understands what the parent is trying to convey (Rowe & Snow, 2020; Song et al., 2014; Thiessen et al., 2005). Similarly, the child shapes the language input they receive through their behaviour and responses to parental speech. Examination of the speech used by each individual during parent-child interaction gives us an insight into how the linguistic input of parents might influence children's development. It can also give us an indication of children's communicative skills and highlight possible language difficulties.

Previous studies recognise the importance of investigating the speech register used during parent-child interactions (Redmond., 2004; Rowe & Snow, 2020; Snow, 1972). Children born with NE are at an increased risk for adverse neurodevelopmental outcomes. However, the impact of NE on child and parent speech has not been adequately investigated. So far, most research has concentrated on the conversational profiles of typically developing parent-children dyads (Anderson et al., 2021; Rowe, 2012) Additional studies have included the investigation of the speech used by mothers and their children born preterm (Salerni et al., 2007; Suttora et al., 2011) and those diagnosed with autism (Bang & Nadig, 2015; Choi et al., 2020; Warren et al., 2010; Woolard et al., 2021). Investigating the speech used by mothers and their children born

with NE will allow us to explore the child's interactive environment and to determine factors that influence child development in this clinical cohort.

Child-Directed Speech

The language parents use with their children is a salient feature of parent-child interaction that is important for child development. Child-directed Speech (CDS) is the particular speech register used when communicating with children. CDS has been found to have long-term influences on children's development (Anderson et al., 2021; Pancsofar & Vernon-Feagans, 2006). As outlined in a review of the literature in chapter two, both the quantity and quality of CDS during parent-child interaction can be measured and has been found to influence child development. Mixed findings have been reported in previous investigations of the linguistic input experienced by children in clinical groups. The majority of studies have found no differences in the quantity of speech used by mothers towards their children born preterm (Salerni et al., 2007) and in those diagnosed with autism (Bang & Nadig, 2015; Watson, 1998). When measuring the quality of parental speech, such as language complexity, findings are mixed with some studies reporting differences compared to parents of typically developing children (Brisson et al., 2014; Choi et al., 2020) and others observing no differences (Bang & Nadig, 2015; Suttora et al., 2020). Inconsistent findings across studies warrant further research investigation of CDS in a cohort where the child is born with NE.

Although no consistent differences in the quantity and quality of CDS used with children in clinical groups is observed when compared to the CDS utilised with typically developing children, numerous studies have reported significant positive associations between characteristics of parental speech and child outcomes in both clinical and typically developing groups (Caskey et al., 2014; Choi et al., 2020; Rowe et al, 2009; Warren et al., 2010). While no studies to date have examined parental CDS with children

born with NE, a study by Rowe and colleagues (2009) investigated the parental speech register used with children who endured early brain injury longitudinally when children were 14 to 46 months of age. The speech of parents whose children had sustained early brain injury was compared to the CDS used towards typically developing children over a period of nine visits. Similar to studies on other clinical cohorts no differences in the quantity and quality of CDS were observed between the groups. Further, positive associations between the quality of parental speech and children's syntactic growth were observed for both groups. Overall, it was found that parents who used more complex language and more diverse vocabulary had children with more advanced language outcomes. Additionally, it was observed that the quality of parental language was a more potent predictor of the syntactic growth in children with early brain injury compared to typically developing children.

Parents systematically alter their speech according to the perceived developmental level of their child (Snow, 1995). Due to the nature of NE, parents of children in this clinical cohort may show different patterns of speech compared to parents of typically developing children. Children impacted by NE have been found to have delayed or impaired language development (Chin et al., 2019; Herrera et al., 2017; Murray et al., 2009). Furthermore, the cohort of children in this study born with NE have been found to score significantly lower on assessments of language abilities when compared to typically developing children (findings outlined in chapter four). Parents may therefore adapt the speech register they use when communicating with their child by using shorter utterances, simpler vocabulary and reduced linguistic complexity in order to attune to their child's language abilities. NE can also impact a child's cognitive functioning and attention abilities (Spencer et al., 2021; Tonks et al., 2019). Thus, in order to promote engagement and gain the child's attention during interaction, parents may alter the linguistic input to

which their child is exposed more than usual. For example, they may use shorter but more frequent utterances to break down play tasks into smaller steps to sustain the child's attention.

Child Speech

When investigating the development of children born with NE, a number of studies indicate that those impacted by the condition show poorer language abilities when compared to children born without complication (Chin et al., 2019; Herrera et al., 2017; Murray et al., 2009). Standardised assessments of development, such as the Bayley-III (Bayley, 2006) and the Wechsler (Wechsler, 2013) scales, are routinely used by researchers and clinicians to evaluate children's developmental skills. However, such instruments are considered to be relatively blunt and have been criticised for their limited precision in assessing children's abilities as they are found to underestimate developmental delay (Anderson et al., 2010; Brito et al., 2019; Calder et al., 2023). In order to obtain a comprehensive understanding of the language abilities of children born with NE, it may be useful to analyse the naturalistic use of speech as opposed to focusing on the acquisition of specific milestones such as the knowledge of individual words or grammatical constructions. It is important to analyse the language use of children within the context of communication during interactive situations that involve a social partner in an environment that facilitates meaningful conversations.

Parent-child interactions play an important role in children's language development as they provide a context where children can verbally and non-verbally engage in back and forth communication with a social partner. This allows children to build upon their language and communication skills (Tamis LeMonda et al., 2001). Previous studies have investigated children's language abilities and vocabulary size following NE (Chin et al., 2019). However, only one study has used conversational speech analysis to investigate

the speech used by children in this clinical cohort during naturalistic interactions with their parents (Cepanec & Ljubesic, 2006). Analysing children's speech during episodes of parent-child interaction gives us the opportunity to better characterise the language profile and communicative intent of children born with NE and how these skills relate to the developmental functioning of children.

Due to the nature of NE, characteristic differences in the speech used by children in this clinical cohort may be observed when interacting with their caregiver. The presence of language delay in those impacted by NE (Herrera et al., 2017; Murray et al., 2009) means that children exhibit slower acquisition of vocabulary and grammar compared to children born without complications. Children with limited vocabulary and knowledge of grammatical constructs are more likely to use shorter and simpler utterances and less likely to understand complex sentences used by parents. NE has also been linked to impairments in cognition and attention (Finder et al., 2019; Spencer et al., 2021; Tonks et al., 2019), and such difficulties can also impact language abilities and the child's ability to recognise social cues. Children experiencing attention problems as a result of NE may find it difficult to attend to social cues and encounter challenges in participating and maintaining conversations, which in turn might limit their language development. Poor attention can also affect verbal fluency, resulting in struggles with articulation, and word retrieval, resulting in disruptions in speech flow or disorganised and fragmented speech (Andersen et al., 2013; Masek et al., 2021; Spencer et al., 2021). The accuracy of children's speech might also be influenced by attention problems as children may make mistakes with word choice and grammar usage.

To date, only one other study has analysed the language used by children born with NE during parent-child interaction (Cepanec & Ljubesic, 2006). The study analysed the complexity of children's speech by calculating the mean length of utterance (MLU) at a

single time point in children aged 1.6 to 3.6 years old. Variation in MLU was observed among the participants, with some children at the early stages of word combination and others speaking in complete sentences. However, it is important to note that the study did not include a comparison group, therefore, the speech of children born with NE was not compared to those born without complications. Furthermore, the results of the study must be interpreted with caution due to the wide age range of the participants and the study's small sample size of just seven children.

When investigating the speech used by children in other clinical groups, characteristic differences are observed when compared to the speech of typically developing children. Children born preterm have been found to be more passive during sessions of parent-child interaction and were observed to vocalise less often than their peers born full-term (Salerni et al., 2007). Similar findings are also observed in cohorts of children who sustained early brain injury (Rowe et al., 2009) and in those diagnosed with specific language impairment (Redmond, 2004). When taking into consideration the quality of speech used by children in clinical groups, differences have also been observed. When compared to typically developing children, children with recognised language impairment (Redmond, 2004; Rescorla et al., 2001), early brain injury (Rowe et al., 2009) and those born preterm (Suttora et al., 2020) use less complex language when communicating with their parents. A recent study investigated the speech used by school aged children whilst interacting with artificial intelligence (AI) stimuli. Overall, it was established that those diagnosed with autism produced shorter utterances and fewer verb types when compared to a group of typically developing children. In addition, children with ADHD and comorbid autism demonstrated less complex language by using fewer nouns, verbs and tokens than their typically developing peers (Boo et al., 2022).

Child-Parent Conversational Balance

In addition to analysing the speech of each individual speaker, it may be productive to examine the speech of both the parent and child jointly. Analysing real time speech will allow us to evaluate the contribution of each speaker to the interaction and to measure the overall conversational balance. A conversation is defined as a sequence of "mutually contingent vocal exchanges" (Bornstein et al., 2015; Stern, 1985) that involve back-and-forth turns that alternate between the speakers (Dunn & Brophy, 2005). Successful conversations rely on turn-taking and a more balanced interaction occurs when parents and their children contribute an approximately equal number of turns of a similar duration (Kelly, 2022). Conversational turn-taking also serves as an indicator of a child's ability to engage in prolonged and meaningful conversations (Leech & Rowe, 2020).

A number of studies of dyads with typically developing children have found conversational balance during parent-child interactions to be associated with children's cognitive, language and social-emotional development (Gilkerson et al., 2017; Gomez & Strasser, 2021). Multiple longitudinal studies have provided evidence that conversational turn-taking between parents and their children is a significant predictor of children's later language abilities (Ferjan Ramirez et al., 2020; Gilkerson et al., 2017; Zimmerman et al., 2009). These findings highlight the positive association between conversational balance and language abilities in children as they get older. When parents and their children take part in back-and-forth conversations it allows parents to appraise the developmental capabilities of the child. This enables parents to adjust the complexity of their language to align with the child's developmental abilities, thus allowing the parent to optimise the child's learning (Vygotsky, 1978). Additionally, balanced conversations give children the opportunity to practice their language skills. The more parents and children engage in

back-and-forth conversation, the more the complexity of these conversations evolves (Edmunds et al., 2019).

As conversational turn-taking involves the contribution of both parents' and children's responsive vocalisations, it is possible that the occurrence of NE may have a negative impact on child-parent conversational balance. Children born with NE have a higher likelihood of experiencing difficulties with their expressive and receptive language abilities (Chin et al., 2019; Herrera et al., 2017; Murray et al., 2009). This can impact children's ability to actively participate in conversation causing the parent to be more talkative relative to the child. Difficulties in receptive language can make it difficult for the child to understand and respond during conversations. Children may need extra time to process their parents' speech leading to pauses or interruptions in turn-taking, which in turn disrupts the conversational balance. Attention difficulties are another prominent feature that impacts those born with NE (Spencer et al., 2021; Tonks et al., 2019) which can make it challenging for children to sustain focus during interaction. Children may struggle to take turns appropriately or pay attention to social cues, leading to disruption of conversational dynamics. Parents of children born with NE may recognise that their child is experiencing developmental difficulties which can also alter the child-parent conversational balance. In an attempt to support their child, the parent may become more directive, leading the interaction and verbalising more frequently than the child.

Previous research has demonstrated preterm children to exhibit more passive and less responsive behaviour towards verbal cues from caregivers during parent-child interaction. Conversely, mothers have been observed to display an active and responsive role (Salerni et al., 2007). Given the reduced activity observed in preterm children, the mother's contribution to the interaction appears to play a compensatory role and is believed to strengthen the dyadic exchange and children's communicative development.

No studies to date have investigated the conversational balance between parents and children impacted by NE.

The Current Study

The current study investigated the quantity and quality of parent and child speech produced during a period of interactive play. The first analysis aims to compare the speech of mothers and their children born with NE (NE group) to the speech used by typically developing children and their mothers (no-NE group). The second analysis will investigate the speech used by 3- and 4-year-old children born with NE, and the relationship between characteristics of speech and children's other developmental outcomes. Results presented in chapter four highlight the association between language and motor abilities in children born with NE. A clear relationship between language and motor domains has already been established in the child development literature more broadly (Iverson, 2010; Valla et al., 2020; see literature review in chapter one). Due to the impact of NE on children's motor development (Jary et al., 2019; Van Kooij et al., 2008), exploring the link between motor skills and speech and language abilities in those born with NE is warranted. The following hypotheses were investigated:

Hypothesis 1. There will be no difference in the quantity (total number of utterances and words) of maternal speech between the NE and the no-NE group. However, a difference in maternal speech quality will be observed in that mothers of children born with NE will produce shorter utterances (MLU) and a less diverse vocabulary (VOCD) compared to mothers in the no-NE group. Given the poorer language abilities exhibited by children born with NE in comparison to typically developing children, conversational turn-taking within the NE group is expected to be less balanced compared to the no-NE group.

Hypothesis 2. Children born with NE will vocalise less frequently than children in the no-NE group. The NE group is predicted to produce lower quality speech compared to the no-NE group, specifically shorter utterances (MLU) and a less diverse vocabulary (Type Token Ratio; TTR[2]). Additionally, children born with NE are predicted to use grammatically less complex language (indicated by fewer verbs per utterance).

Hypothesis 3. In the cohort of 3- and 4-year-old children born with NE, it is expected that the speech of mothers and their children will differ as a function of age. Mothers of 4 year olds will display a more diverse vocabulary and use more grammatically complex language than mothers of 3 year olds. As children develop their language abilities and become more responsive with age, it is expected that conversational turn-taking will be more balanced in the cohort of 4-year-olds compared to 3-year-olds. Similarly, it is expected that the cohort of 4-year-old children will produce more utterances, turns and words compared to 3-year-olds. The quality of speech will also be higher in older children in that 4-year-olds are expected to use more diverse vocabulary and demonstrate more grammatically complex language by producing more verbs in their speech, compared to 3-year-olds.

Hypothesis 4. Previous research has demonstrated a link between CDS and child developmental outcomes. Therefore, it is expected that positive associations between the quantity and quality of parent speech and developmental outcomes in children will be observed. Mothers who talk more, produce a broader vocabulary and use more grammatically complex language will have children with higher developmental assessment scores. Greater conversational balance will also be positively associated with children's developmental abilities. When analysing the speech of children born with NE,

[2] TTR is a measure of vocabulary diversity.

the quantity and quality of speech will be positively correlated with higher scores on developmental assessments.

Method

Participants

Participating families in the NE group were originally recruited at the time of the child's birth from one of three maternity hospitals in Dublin, Ireland including the National Maternity Hospital, the Coombe Women and Infants Hospital and the Rotunda Hospital. Mothers of infants diagnosed with NE were approached and asked to take part in the research study (see chapter 3 for full details). When the children were 3 years of age or older, families were re-contacted by phone or post and were invited for follow up.

A total of 46 children aged 36 to 66 months of ages participated in the current study with their mothers. For the first set of analysis, comparing NE and no-NE children, the sample comprised 34 families with children aged 45 to 66 months of age, half of whom were born with NE (n = 17) and half of whom are typically developing children born without complication (no-NE group, n = 17). Children in the no-NE group were drawn from a group of typically developing children born without complications, they were matched and selected with the NE children based on child age (refer to chapter three for full details). No differences in age were observed between the NE (M = 56.35, SD = 6.28) and no-NE group (M=53.12, SD = 2.37; t = -1.99, p = 0.06). The NE group comprised 10 males (58.8%) and 7 females (41.2%) and the no-NE group included 8 males (47.1%) and 9 females (52.9%). A similar proportion of males and females were recruited in each group (X^2 = 0.47, p = 0.49). All participating mothers completed secondary level education. In total, 47.1% of mothers with children born with NE had a bachelor's degree compared to 52.9% of mothers in the no-NE group. No difference in maternal education was observed between the groups (X^2 = 6.83, p = 0.15).

The second set of analyses involving NE 3- and 4-year old children includes 29 families all with children born with NE. The sample was divided into two groups for comparison based on child age. The 3-year-old cohort included 12 children aged 36 to 46 months of age (M = 40.64, SD = 3.40) and the 4-year-old+ group included 17 children aged 49 to 66 months old (M = 56.35, SD = 6.28). The group of 3-year-old children born with NE comprised 9 males (75%) and 3 females (25%) and the 4-year-old+ cohort included 10 males (58.8%) and 7 females (41.2%). A similar proportion of males and females were recruited in each group (X^2 = 0.82, p = 0.37). All mothers had completed at least secondary level education and no difference in maternal education was observed between the groups (X^2 = 5.09, p = 0.28).

Procedure

Mothers and their children were invited to the Infant and Child Research Laboratory at Trinity College Dublin, Ireland for follow-up. During the visit to the laboratory the mother and child took part in an episode of semi-structured play and the parent-child interaction was observed and recorded. To initiate the parent-child interaction the mother and child were given a construction set and the dyad was left alone to play with the toy. A Zoom H2n Handy Recorder was used to capture high quality audio recordings of the parent-child interaction. The microphone was concealed in the corner of the room to prevent distraction and the speech of the mother and child was recorded for the duration of the five-minute interaction.

After taking part in the parent-child interaction, each child in the NE group completed a standardised developmental assessment. The assessment administered to the child depended on the age of the child on the day of the test. Children aged 36 to 46 months of age completed the Bayley-III (Bayley, 2006) and children aged 47 months and

older were administered the WPPSI-IV (Wechsler, 2013) and the Movement-ABC (Henderson et al., 2007).

Measures

Speech Transcription and Analysis. Once recorded, the speech used by the parent and child during the structured play interaction was transcribed by trained research assistants at the utterance level according to the Codes for Human Analysis of Transcripts (CHAT) conventions (MacWhinney, 2000). All speech was transcribed verbatim and each transcript was reviewed by a senior transcriber. Prior to conducting the speech analysis, CLAN's CHECK and MOR functions were used to confirm the transcripts were accurate and that words followed the correct morphological structure. Measures of parent and child speech were then extracted and analysed from the transcripts using the CLAN programme (Computerised Language Analysis; MacWhinney, 2000).

Maternal Speech. The following variables were used to measure the quantity of the mother's speech: total number of utterances, turns and words, the number of words per turn, utterances per turn and the number of words per utterance. The quality of mother's speech was examined through the analysis of: Mean length of utterance in morphemes (MLUm), vocabulary diversity (VOCD), number of verbs per utterance and child-parent MLT (mean length of turn) ratio.

Utterance. Utterances are defined as speech units which are separated by a pause, change in intonation, and/or grammatical structure.

Turn. A conversational turn is defined as a sequence of utterances spoken by a single speaker. While the same speaker is talking, each utterance is part of the current turn. During a turn the conversational partner does not talk or overlap between the current speaker's utterances.

Mean Length of Utterance in Morphemes (MLUm). MLU is an index of language complexity. Language complexity is calculated by dividing the total number of morphemes or words used by the total number of utterances. Longer utterances (indicated by a higher MLUm) demonstrate the use of more complex grammatical constructs (Brown, 1973).

Vocabulary Diversity (VOCD). VOCD is an index of lexical diversity. VOCD considers the probability of new vocabulary being used in longer samples of speech. It is calculated by summing the number of unique word stems spoken by the mother during the length of the parent-child interaction using random sampling. Lower values of VOCD indicate more repetition and less lexically rich speech.

Child-Parent MLT (Mean Length of Turn) Ratio. The child-parent MLT ratio is a measure of conversational balance. MLT considers the ratio of utterances over turns for the child and the mother. The child-parent MLT ratio is then calculated by dividing the child's MLT by the parent's MLT. An MLT ratio closer to 1 indicates greater conversational balance, whereas a score closer to 0 indicates that the parent was more talkative relative to the child.

Child Speech. The following variables were used to quantify child speech: the total number of utterances, turns and words used by the child during the interaction. The mean number of utterances used per turn and the number of words per utterance was also calculated. The quality of child speech was established by measuring MLUm, Type-Token Ratio (TTR), the number of verbs per utterance, the proportion of nouns and the proportion of verbs used by the child.

Type-Token Ratio (TTR). TTR is a measure of vocabulary diversity. TTR is calculated by dividing the number of different word types used by the total number of words. Higher values of TTR indicate higher levels of lexical diversity.

Developmental Assessment

The Bayley Scales of Infant and Toddler Development – Third Edition (Bayley-III; Bayley, 2006). In the cohort of 3-year-old children, cognitive, language and motor abilities were assessed by a trained researcher using the Bayley-III. All methods and procedures used to assess development using the Bayley-III are detailed in chapter three.

Wechsler Preschool and Primary Scale of Intelligence – Fourth Edition (WPPSI-IV; Wechsler, 2013). In the cohort of 4-year-old children the WPPSI-IV was used to assess children's cognitive and language abilities. Due to the importance of language and processing speed abilities in children born with NE, the verbal comprehension index and processing speed index were selected for the analysis. All procedures for the administration of the WPPSI-IV are detailed in chapter three and in chapter five study two.

Movement Assessment Battery for Children – Second Edition (Movement-ABC; Henderson et al., 2007). The Movement-ABC assessment was administered to assess children's motor abilities in the cohort of 4-year-old children born with NE. All procedures describing the use of the Movement-ABC are detailed in chapter three.

Statistical Analysis

Data from this study were analysed using SPSS (version 26). A series of independent samples t-tests was conducted to investigate the mean differences in CDS and child speech between the NE and no-NE group, and in 3- and 4-year-old children born with NE. A Bonferroni correction was applied to reduce the chances of type I error when performing multiple comparison tests, therefore alpha criterion was adjusted to 0.01. To satisfy the assumptions of parametric statistics, data were assessed for normality using Shapiro Wilk's test and homogeneity of variance was checked using Levene's test.

When checking the assumptions of the NE and no-NE groups, variables indicating the number of utterances, turns and utterances per turn used by children did not meet the assumption of normality. A Mann-Whitney *U* test was therefore computed to compare the quantity and quality of child speech between the two groups. Pearson's correlation coefficients were calculated to analyse the relationships between characteristics of parent speech and developmental outcomes. Although multiple comparisons were conducted in the correlation analyses, alpha criterion remained at 0.05 to reduce the chance of type II error.

Results

CDS during Parent-Child Interactions: NE vs no-NE Sample

The Quantity of CDS during Parent-Child Interaction. A series of independent samples t-tests were used to compare the quantity of CDS in the NE and no-NE groups. Overall, mothers in the NE group tended to use less words per turn and less utterances per turn (see table 5.3.1). There were no other significant differences in the quantity of speech used such as the total number of utterances, turns or words used by the mothers in each group. Due to the adjusted p-value of 0.01 there was also no difference in utterances per turn.

The Quality of CDS during Parent-Child Interaction. Overall, no differences were observed in the quality of CDS used by mothers across groups (see Table 5.3.1).

Table 5.3.1. *Characteristics of CDS of mothers in the NE and no-NE groups.*

	NE (n=17)	**No-NE** (n=17)	**Group Difference**
	M (SD)	*M (SD)*	*t-value, p-value*
Parent Speech Quantity			
Total no. utterances	74.94 (18.96)	81.65 (18.08)	t = 1.06, p = 0.30
Total no. turns	42.59 (10.49)	37.65 (5.51)	t = -1.72, p = 0.10
Total no. words	409.53 (168.78)	489.18 (156.20)	t = 1.43, p = 0.16
Words per turn	9.61 (3.06)	13.12 (4.16)	t = 2.80, p = 0.01*
Utterances per turn	1.80 (0.42)	2.20 (0.53)	t = 2.45, p = 0.02
Words per utterance	5.33 (1.25)	5.91 (1.04)	t = 1.46, p = 0.16
Parent Speech Quality			
MLUm	5.74 (1.34)	6.35 (1.08)	t = 1.44, p = 0.16
VOCD	45.04 (6.39)	42.84 (5.89)	t = -1.04, p = 0.31
Verbs per utterance	1.02 (0.28)	1.20 (0.20)	t = 0.94, p = 0.35
Child-Parent MLT Ratio	0.86 (0.47)	0.61 (0.21)	t = -1.98, p = 0.06

Note. No. = Number; MLUm= Mean Length of Utterance in Morphemes; VOCD = Vocabulary Diversity; MLT = Mean Length of Turn.

*$p \leq 0.01$

Child Speech during Parent-Child Interactions: NE vs no-NE Sample

The Quantity of Child Speech during Parent-Child Interaction. A series of Mann-Whitney *U* tests was used to compare the quantity of speech used by children in the NE and no-NE groups. There were no significant differences in the quantity of speech used by children in either group (see table 5.3.2).

The Quality of Child Speech during Parent-Child Interaction. A series of Mann-Whitney *U* tests revealed no significant differences in MLUm, Type-Token Ratio or proportion of verbs. However, the speech of children born with NE included a higher proportion of nouns compared to children in the no-NE group (see table 5.3.2).

Table 5.3.2. *Speech characteristics of children in the NE and no-NE groups aged 4 years.*

	NE (n=17)	**No-NE** (n=17)	**Group Difference**		
	Median (IQR)	*Median (IQR)*	*U*	*Z*	*p-value*
Child Speech Quantity					
Total number of utterances	52.00 (28.50)	48.00 (14.00)	98.00	-1.61	0.11
Total number of turns	43.00 (13.50)	39.00 (7.00)	97.00	-1.64	0.10
Total number of words	216.00 (135.50)	169.00 (52.00)	101.50	-1.48	0.14
Utterances per turn	1.26 (0.37)	1.28 (0.26)	120.00	-0.84	0.40
Words per utterance	3.37 (1.78)	3.40 (1.33)	142.00	-0.09	0.93
Child Speech Quality					
MLUm	4.14 (1.66)	3.82 (1.64)	130.50	0.48	0.63
Type-Token Ratio	0.38 (0.11)	0.43 (0.11)	120.00	-0.84	0.40
Verbs per utterance	0.70 (0.42)	0.74 (0.26)	140.00	-0.16	0.88
Proportion of Nouns used	14.85 (7.11)	10.00 (5.03)	64.00	-2.77	0.01*
Proportion of Verbs used	18.98 (4.16)	18.68 (4.64)	137.50	-0.24	0.81

Note. MLU = Mean length of utterance in morphemes.

*$p \leq 0.01$; significance level adjusted to 0.01 to correct for multiple analyses.

CDS and Child Speech in 3- and 4-year olds Born with NE and their Mothers

The Quantity and Quality of CDS used by Mothers of 3- and 4-year-old Children Born with NE. A series of independent samples t-tests indicated no significant differences between mothers of 3- and 4-year old children in the quantity of CDS including the total number of utterances, turns and words spoken and the number of words per turn. When comparing the quality of CDS used by mothers of 3- and 4-year-old children born with NE, mothers of 4-year-old children used more diverse vocabulary (VOCD). No significant differences in MLUm, verbs per utterance and Child-Parent MLT ratio was observed between the groups (see table 5.3.3).

The Quantity and Quality of Speech used by 3- and 4-year-old Children Born with NE. A series of independent samples t-tests was used to compute differences in the quantity and quality of speech used by 3- and 4-year-old children born with NE. Overall, no differences were observed in the quantity or quality of speech used by children across the groups (see Table 5.3.4).

Table 5.3.3*.Characteristics of CDS: Mothers of -3 and 4-year-old children born with NE.*

	3-year-olds (n=12)	**4-year-olds** (n=17)	**Group Difference**
	M (SD)	*M (SD)*	*t-value, p-value*
Parent Speech Quantity			
Total number of utterances	77.58 (19.31)	74.94 (18.96)	$t = 0.38$, $p = 0.72$
Total number of turns	40.83 (11.97)	42.59 (10.49)	$t = -0.42$, $p = 0.68$
Total number of words	344.25 (88.13)	409.53 (168.78)	$t = -1.22$, $p = 0.23$
Words per turn	8.82 (2.71)	9.61 (3.06)	$t = -0.72$, $p = 0.48$
Words per utterance	1.20 (0.62)	1.80 (0.42)	$t = -2.02$, $p = 0.05$
Parent Speech Quality			
MLUm	4.84 (0.79)	5.74 (1.34)	$t = -2.09$, $p = 0.04$
VOCD	38.03 (7.62)	45.04 (6.39)	$t = -2.69$, $p = 0.01$*
Verbs per utterance	0.92 (0.14)	1.02 (0.28)	$t = -1.17$, $p = 0.25$
Child-Parent MLT Ratio	0.68 (0.20)	0.86 (0.47)	$t = -1.26$, $p = 0.22$

Note. MLU= Mean Length of Utterance in Morphemes; VOCD = Vocabulary Diversity; MLT = Mean Length of Turn.

*$p \leq 0.01$; significance level adjusted to 0.01 to correct for multiple analyses.

Table 5.3.4. *Characteristics of speech of 3- and 4-year-old children born with NE.*

	3-year-olds (n=12)	**4-year-olds** (n=17)	**Group Difference**
	M (SD)	*M (SD)*	*t-value, p-value*
Child Speech Quantity			
Total number of utterances	50.50 (15.46)	58.59 (19.41)	t = -1.20, p = 0.24
Total number of turns	40.50 (12.00)	42.41 (10.40)	t = -0.46, p = 0.65
Total number of words	151.08 (51.95)	210.59 (97.99)	t = -1.92, p = 0.07
Utterances per turn	1.25 (0.16)	1.41 (0.44)	t = -1.20, p = 0.24
Words per utterance	3.05 (1.07)	3.56 (1.19)	t = 1.20, p = 0.24
Child Speech Quality			
MLUm	3.32 (1.12)	3.94 (1.18)	t = -1.41, p = 0.17
Type-Token Ratio	0.42 (0.09)	0.40 (0.09)	t = 0.58, p = 0.57
Verbs per utterance	0.53 (0.20)	0.67 (0.27)	t = -1.58, p = 0.13
Proportion of Nouns used	18.06 (4.61)	15.36 (6.04)	t = 1.30, p = 0.20
Proportion of Verbs used	16.87 (3.13)	18.09 (3.46)	t = -0.97, p = 0.34

Note. MLU= Mean Length of Utterance in Morphemes.

The Relationship between CDS and Developmental Assessment Scores in 3- and 4-year-old Children Born with NE

Tables 5.3.5 presents bivariate correlations between parent language variables during structured play and child outcome scores on the cognitive, language and motor scales of the Bayley-III measured in the cohort of 3-year-old children. Table 5.3.6 includes correlations between parent language variables and child outcomes scores on the VCI and PSI scales of the WPPSI-IV, and Movement-ABC scores in the cohort of 4-year-old children. Overall, no significant relationships were observed between the quantity and quality of CDS and child developmental scores for either age group.

Table 5.3.5. *Correlations between CDS and developmental assessment scores in 3-year-olds born with NE (n=12).*

	Developmental Assessment Scores				
	Cognitive Composite Score	Receptive Language Scaled Score	Expressive Language Scaled Score	Language Composite Score	Motor Composite Score
Parent Speech Quantity					
Total Number of Utterances	-.52	-.43	-.41	-.44	-.20
Total Number of Turns	-.30	-.23	-.43	-.34	-.37
Total Number of Words	-.39	-.24	-.55	-.37	-.31
Words per Turn	-.01	.04	.00	.05	.20
Utterances per Turn	-.12	-.20	.02	-.11	.22
Words per Utterance	.27	.43	-.07	.25	-.13
Parent Speech Quality					
MLUm	.24	.46	-.03	.28	-.11
VOCD	.14	.08	-.05	.00	-.10
Verbs per utterance	.18	.43	.00	.28	-.16
Parent-Child MLT Ratio	-.23	-.21	-.03	-.13	.21

Note. MLU= Mean Length of Utterance in Morphemes; VOCD = Vocabulary Diversity; MLT = Mean Length of Turn.

Table 5.3.6. *Correlations between CDS and developmental assessment scores in 4-year-olds born with NE (n=17).*

	Developmental Assessment Scores		
	VCI Score	PSI Score	M-ABC Score
Parent Speech Quantity			
Total Number of Utterances	-.12	-.15	-.21
Total Number of Turns	-.04	.05	.15
Total Number of Words	-.02	-.16	-.12
Words per Turn	-.02	-.21	-.27
Utterances per Turn	-.12	-.22	-.38
Words per Utterance	.07	-.12	-.05
Parent Speech Quality			
MLUm	.04	-.16	-.06
VOCD	.29	.12	.29
Verbs per utterance	.01	-.21	-.12
Parent-Child MLT Ratio	-.18	-.23	-.34

Note. MLU= Mean Length of Utterance in Morphemes; VOCD = Vocabulary Diversity; MLT = Mean Length of Turn; VCI = Verbal Comprehension Index; PSI = Processing Speed Index; M-ABC = Movement-ABC.

The Relationship Between Child Speech and Developmental Assessment Scores in 3- and 4-year-old Children Born with NE

Table 5.3.7 presents bivariate correlations between the quantity and quality of child speech used during a structured play interaction, and cognitive, language and motor abilities measured using the Bayley-III in the cohort of children aged 3 years born with NE. Overall, no significant associations between child speech and developmental abilities were observed in this age group. Table 5.3.8 includes correlations between child language variables and scores on the VCI and PSI scales of the WPPSI-IV, and Movement-ABC

scores in the cohort of 4-year-old children born with NE. The analyses indicate positive associations between children's VCI, PSI and Movement-ABC scores and the quantity and quality of their speech during parent-child interaction. A positive relationship was observed between VCI scores and children's words per utterance, language complexity (as measured by MLUm) and verbs per utterance. Children who used a higher total word count and more words per utterance were more likely to receive higher scores on the PSI scale of the WPPSI-IV assessment. Positive associations were also observed between Movement-ABC scores and the total number of words, words per utterance and the proportion of verbs used in speech.

Table 5.3.7*. Correlations between child speech and developmental assessment scores in 3-year-olds born with NE (n=12).*

	Developmental Assessment Scores				
	Cognitive Composite Score	Receptive Language Score	Expressive Language Score	Language Composite Score	Motor Composite Score
Child Speech Quantity					
Total Number of Utterances	-.17	-.22	-.32	-.30	-.32
Total Number of Turns	-.32	-.25	-.45	-.37	-.40
Total Number of Words	.04	.12	.04	.07	-.11
Utterances per Turn	.35	.04	.16	.09	.03
Words per Utterance	.23	.46	.42	.48	.20
Child Speech Quality					
MLUm	.19	.42	.37	.43	.12
Type-Token Ratio	-.08	-.18	.06	-.09	.20
Verbs per utterance	.00	.41	.36	.41	.02
Proportion of Nouns used	.34	.19	.28	.24	.20
Proportion of Verbs used	-.42	-.14	-.05	-.14	-.36

Note. MLU= Mean Length of Utterance in Morphemes.

Table 5.3.8. *Correlations between child speech and developmental assessment scores in 4-year-olds born with NE (n=17).*

	Developmental Assessment Scores		
	VCI Score	PSI Score	M-ABC Score
Child Speech Quantity			
Total Number of Utterances	.15	.27	.34
Total Number of Turns	-.03	.07	.15
Total Number of Words	.47	.49*	.51*
Utterances per Turn	.20	.21	.26
Words per Utterance	.65**	.51*	.50*
Child Speech Quality			
MLUm	.63**	.44	.47
Type-Token Ratio	-.32	-.33	-.42
Verbs per utterance	.48*	.39	.54*
Proportion of Nouns used	-.19	-.14	-.37
Proportion of Verbs used	.05	.12	.53*

Note. MLU= Mean Length of Utterance in Morphemes.

*p≤ 0.05; **p≤ 0.01.

Discussion

Both parents and children play active roles during interaction as each participant influences the other's behaviour, thus, shaping the course of a conversation. The speech profile used by the mother and child during interaction is dynamic and has been linked to children's development (Anderson et al., 2021; Rowe, 2012). Despite the risk of adverse neurodevelopmental outcomes, the impact of NE on child and parent speech had yet to be adequately investigated. This study examined the quantity and quality of speech that parents and children impacted by NE used during a period of interactive play.

The first hypothesis sought to compare the quantity and quality of CDS used by mothers of children born with NE and mothers of typically developing children born

without complications. Contrary to our hypothesis, no differences were observed in quality of CDS used by mothers during parent-child interaction. The only difference observed was that mothers of children born with NE used less words per turn however, there was no difference in the total number of turns used by mothers in each group. This suggests that mothers in the NE group used shorter turns during interactions with their children. Due to the language and attention difficulties common in children born with NE (Chin et al., 2019; Tonks et al., 2019) mothers may be pacing their own responses to accommodate for these difficulties. This may encourage the child to speak more often or assist in sustaining the child's attention by breaking down play tasks into steps. The use of shorter turns might also indicate that more concise speech patterns are being used, as succinct language is easier for children to understand.

Despite this difference in maternal speech quantity, the current study failed to identify any differences in the quality of CDS used by mothers in the NE and no-NE groups. The existing literature on the quality of CDS used in clinical groups compared to typically developing groups presents mixed findings. Some studies suggest there are differences in the quality of parental speech and have found mothers of children in clinical groups to use less complex language (Brisson et al., 2014; Choi et al., 2020). Other studies have observed no such differences (Bang & Nadig, 2015; Suttora et al., 2020). Factors such as the age of the child at assessment and the study sample size seem to influence findings when comparing the methodologies of studies that do and do not find differences in the quality of CDS. Studies that established differences between clinical and non-clinical groups tended to have larger sample sizes (Choi et al., 2020) and assessed the speech of parents directed towards younger children aged 6 to 24 months (Brisson et al., 2014; Choi et al., 2020). On the other hand, studies that found no differences in CDS had smaller sample sizes and the samples tend to include older

children ranging from 30 to 85 months of age (Bang & Nadig, 2015; Suttora et al., 2020). In accordance with the studies reporting null findings, the current study included a small sample size and assessed the CDS directed towards older children aged 45 to 66 months of age. It is possible that as children get older, any distinctive features of parents' CDS used towards children in clinical groups diminishes. However, using higher quality speech may not necessarily match the developmental level of children in clinical groups and may have differing associations with their developmental abilities (Snow, 1995).

When child-parent conversational balance was compared between NE and no-NE groups no statistically significant differences were observed, although the finding of the NE group displaying greater conversational balance when compared to the no-NE group approached significance. Given that differences were observed in utterances per turn used by the mothers but not by the children, the mothers' MLT may be driving this greater conversational balance among the NE group. This further supports the finding that mothers of children born with NE are pacing their speech in order to encourage their child to speak, thus, promoting conversational turn-taking.

The second hypothesis sought to compare the quantity and quality of speech used by children born with NE and typically developing children born without complications. Overall, no differences were observed in the quantity and quality of speech, except for the proportion of nouns used. Given that the children in the NE group scored significantly lower in developmental assessments of language (see chapter four) these findings are unusual. This suggests that children in the NE group struggle with language assessments but not necessarily with the demands of real time conversation and pragmatics. It may also reflect something about the nature of developmental assessments and implies that such instruments measure language abilities differently compared to the analysis of naturalistic speech patterns (Brito et al., 2019; Calder et al., 2023; Litt & Church, 2023).

Developmental assessments of language rely not only on speech abilities but also on cognitive processes and comprehension, which may pose challenges for children born with NE (Finder et al., 2019; Schreglmann et al., 2020; and chapter four findings on processing speed).

When comparing the speech of children in the NE group and the no-NE group during parent-child interaction, the only notable difference was the proportion of nouns used in speech. The results showed that children born with NE used a higher proportion of nouns compared to the children born without complications. Using more nouns in speech is typically referred to as 'noun bias' (Bates et al., 1994; Frank et al., 2021; Gentner, 1982). Noun bias is a phenomenon common during early language learning where young children use more nouns compared to other word types such as verbs or adjectives. Nouns are words that represent objects and entities in the immediate environment. It is suggested that nouns are easier for children to acquire because objects are recognisable and easy to describe in comparison to actions and abstract concepts (Markman, 1994). At around 3 to 4 years of age noun bias tends to fade. As language development advances, children acquire a more diverse vocabulary that includes a wider range of word types such as verbs and adjectives. If noun bias persists in children born with NE it may indicate difficulties in acquiring and using more advanced language structures and word types.

The third hypothesis concerned the quality and quantity of speech used by children and mothers impacted by NE. Speech was compared between dyads that included 3-year-old and 4-year-old children born with NE. As hypothesised, mothers of 4-year-olds used higher quality speech and used more diverse vocabulary when compared to the mothers of 3-year-old children born with NE. Mothers of 4-year-olds also demonstrated greater lexical complexity and used more words per utterance (despite

being statistically non-significant, the findings align with the hypotheses). As observed in samples including typically developing children, parents systematically alter the quality of their speech as their child's language abilities develop over time (Anderson et al., 2021). The transactional model of development suggests that children have an active influence on the behaviours of those with whom they interact (Sameroff, 2009). Thus, factors such as child age, developmental level and the quantity and quality of the child's speech will have a dynamic influence on the speech used by the parent. Previous studies have demonstrated that qualities of parental speech such as the complexity of CDS and parental vocabulary are associated with child speech abilities during parent-child interaction (Bohannon & Warren-Leubecker, 1985; Dale & Spivey, 2006; Quigley & Nixon, 2019). Although there were differences in CDS between the two age groups, no difference in the quantity and quality of child speech was observed between the 3- and 4-year-old children born with NE. This perhaps indicates that the language abilities of children born with NE follows a non-linear pattern of development (Ko, 2012). Alternatively, these findings might be attributed to methodological flaws such as the small sample size, which will be discussed in the limitations section of this chapter.

Hypothesis four sought to investigate associations between the speech of children and parents impacted by NE and children's developmental abilities. It was predicted that higher parental speech quantity and quality would be associated with higher child scores on standardised assessments. However, no significant associations were observed between any of the variables. These null findings may also be attributed to factors related to the study's methodology. Due to the small sample size, it is possible that the limited number of participants has reduced the statistical power of the study to detect any significant associations. Additionally, this analysis focused on dyads impacted by NE and did not take into consideration dyads with typically developing children born without

complications. Future research should look to compare the relationship between characteristics of CDS and child outcomes in both groups to determine whether the associations observed in children born with NE deviate from, or follow, the trajectory of typically developing children (Choi et al., 2020; Rowe et al., 2009). Comparing the influence of CDS on child outcomes in both groups allows us to identify aspects of parental speech that are impacted by NE and whether they have differing associations with children's developmental abilities.

When analysing the association between the quantity and quality of child speech and children's developmental scores in 4-year-olds born with NE, the results showed significant positive relationships between the variables as hypothesised. Positive associations between the child's MLUm, words per utterance and verbal comprehension scores were observed. This suggests that children who use more complex language and longer utterances are better at understanding and processing verbal information. A positive association was also found between the number of verbs used per utterance and VCI scores. Verbs are an essential component of language that describe actions or events (Givon, 1984). Children who use more verbs in their speech demonstrate a higher level of lexical sophistication (Frank et al., 2021). As children incorporate more verbs into their speech, they are applying grammatical concepts and expressing complex ideas that can also be linked to their verbal comprehension skills. Overall, these findings indicate that speech quality and verbal comprehension in children born with NE is closely linked, as children who display superior language skills also display better comprehension abilities.

Further associations between child speech quantity and processing speed were established in 4-year-old children born with NE. It was found that children who used more words and words per utterance had higher scores on the processing speed scale of the WPPSI-IV. The PSI scale measures how quickly and efficiently the child can process

and respond during a cognitive task (Wechsler, 2013). Previous studies on children born preterm found that children with better processing speed abilities are better word learners (Marchman et al., 2022). Our findings support the literature and indicate that children born with NE who produce more speech have faster cognitive processing abilities as they are able to retrieve and express information more rapidly. This suggests that language expression and cognitive functions are linked in this cohort of children born with NE.

As hypothesised, positive associations between children's motor abilities and characteristics of speech were also observed. Overall, children with higher scores on the Movement-ABC tended to use more words, more words per utterance, used a higher proportion of verbs and more verbs per utterance. This suggests that children with better motor abilities speak more and use higher quality language. The link between language and motor abilities in children has been widely researched (Iverson, 2010; Valla et al., 2020). It has been established that the development of motor skills allows children to navigate their environment and encounter new opportunities for learning (Hofsten, 2009). Children born with NE who have better motor abilities may have more opportunities for social interaction and engagement. Greater participation in activities allows children to practice their language abilities leading to higher levels of speech being used. As verbs are words that describe actions, motor abilities can influence the acquisition and use of verbs in children (Levi, 2013). Children with better motor skills are more likely to take part in physical activities, recognise actions, imitate and name them which can lead to more diverse verb usage.

Additional limitations in the study's methodology must be taken into consideration when interpreting the results. A number of null findings present in the study may be attributed to the study's small sample size. For example, associations were found between child speech and developmental outcomes in the cohort of 4-year-olds born with

NE but not in the smaller sample of 3-year-old children. The limited number of participants has impacted the statistical power of the study and has restricted the present analysis. If a larger sample was recruited, regression analyses could be conducted to establish the predictive power of speech quantity and quality on child developmental outcomes. However, issues with recruitment such as attrition and restrictions associated with the COVID-19 pandemic has impacted the sample size of this study. Future studies will benefit from replicating this research with a larger sample size and a longitudinal study design, thus increasing the statistical power of the study to detect significant relationships.

The heterogeneity of the NE group may have also played a role in these findings. Varying degrees of neurodevelopmental abilities has been observed in children born with NE, with some children experiencing delay (Herrea et al., 2017; Mitra et al., 2018; Murray et al., 2005) and others performing as well as typically developing children (Azzopardi et al., 2014; Natarajan et al., 2016). This makes it difficult to identify consistent associations across the cohort. Due to the small sample, factors such as the grade of NE could not be taken into consideration despite being an important predictor for child outcomes. Future investigations should take into account the impact of the grade of NE since children born with mild, moderate and severe NE are likely to demonstrate varying levels of language ability.

Despite the negative impact of NE on children's language abilities (Chin et al., 2019; Herrera et al., 2017; Murray et al., 2009), no studies have previously investigated the language of parents and their children born with NE in detail. This is the first study to compare the speech of parents impacted by NE with the speech of parents of typically developing children. It is also the first study to report associations between characteristics

of speech during parent-child interactions and child developmental scores in those born with NE.

To date this is only the second study to investigate the speech of children born with NE. Some of the methodological flaws of the first study have been addressed in the current study by including a larger sample size and taking into account child age. Overall, we have identified characteristics of speech that parents adopt to facilitate child speech, such as pacing their speech and using more concise utterances. We have also further characterised the language used by children born with NE and how qualities of speech relate to children's developmental outcomes.

Chapter Six

General Discussion

Summary of Rationale and Findings

To date, research investigating the development of children born with NE has primarily focused on the clinical outcomes of those impacted by the disorder. Standardised assessments of development are the most common tool used to assess the development of children born with NE and are typically used as end-point assessments for neonatal trials. However, due to the heterogeneity of NE, the developmental profile of NE is still relatively undefined and the underlying processes that influence the development of children affected by the disorder had yet to be investigated. The investigation of parent-child interactions is a well-established approach that has been widely used to examine the developmental processes that influence child development. The parent-child interactions of typically developing children and of those in other clinical groups have been widely studied and characterised, however, this approach has yet to be applied in the context of those born with NE. In an attempt to provide further insight into the impact of NE on child development this thesis also examined the association between interactions between mothers and children born with NE and children's developmental abilities.

Specifically, this thesis characterised quantitative and qualitative features of JA episodes and of the speech used by mothers and their children during dyadic interaction. Mother-child dyads where children were born with NE and dyads with children born without complications were compared. Participating families were recruited for this study when the child was aged 2 years or older and consisted of two groups including children born with NE and typically developing children born without complications. Naturalistic interactions of children and their parents at play were recorded and an age-appropriate

standardised assessment of development was administered to assess children's developmental abilities. This chapter summarises the key findings of this thesis and will be discussed in the context of the existing literature. The strengths and limitations of this research are outlined and recommendations for future research are provided.

The study outlined in Chapter 4 sought to characterise the developmental profile of a group of preschool children born with NE by comparing their developmental assessment scores to those of a group of typically developing children born without complications. Developmental risk factors such as neonatal risk, autism traits, executive functioning and sleep difficulties were measured in those born with NE and the associations with developmental assessment scores were examined. Overall, children born with NE were found to perform significantly worse on assessments of language abilities. However, no differences were observed in domains of cognition apart from processing speed where differences were observed in the cohort of children aged 4 and older. Numerous studies have identified cognitive deficits in children born with NE (Finder et al., 2019; Pappas et al., 2015; Schreglmann et al., 2019). Therefore, these results continue to highlight the variability in the developmental abilities of children born with NE. NE is a heterogenic disorder with multifactorial aetiology, thus, inconsistent findings are common. Studies have reported that cognitive difficulties may become more apparent in later life (Lindström et al., 2006) and that a single developmental assessment score is not an accurate predictor of outcomes at school age (Anderson et al., 2010; Chalak et al., 2014; Hack et al., 2005). Therefore, it is suggested that children born with NE are followed up into later childhood and at multiple time points (Neel et al., 2023).

A sizeable proportion of the children born with NE exhibited a high number of autism traits and EF deficits. More than half of children who showed these developmental risk factors also received below average scores on assessments of language and motor

abilities. In line with the literature, these findings suggest that children who exhibit autism traits and EF difficulties are more likely to experience delays in language and motor development (Gooch et al., 2016; Liu & Breslin, 2013; Reindal et al., 2021). The practical implications of these findings are further discussed below.

The results of this study also established that language and motor abilities in children born with NE are highly correlated. This finding is in line with the literature indicating that language and motor abilities in children are interrelated (Iverson, 2010; Valla et al., 2020). A previous study on children born with NE found that performance on cognitive assessments is linked with children's motor abilities (Jary et al., 2019). The results of the current study adds to the body of literature suggesting that deficits in motor functioning caused by NE are linked to difficulties in other domains of development (Jary et al., 2019; Martinez-Biarge et al., 2012; Van Kooij et al., 2010).

Chapter 5 included three studies that investigated the association between parent-child interactions and the developmental abilities of children in clinical groups including those born with NE. Study 1 examined the association between global characteristics of parent-child interactions and the developmental outcomes of children with an elevated or typical likelihood (EL or TL) of developing autism. Parents and children with an EL for autism show characteristic differences during parent-child interactions compared to TL dyads (Pijl et al., 2021; Wan et al., 2012). This preliminary investigation demonstrates that there are also differences in the association between parent-child interaction quality and developmental outcomes of children in the EL and TL groups. The global coding scheme used to measure the quality of parent-child interactions in this study (MACI; Wan et al., 2017) may not have been sensitive enough to detect differences in parent-child interactions between the two groups. Therefore, microanalytic coding techniques were used to investigate the parent-child interactions of children born with NE.

Study 2 investigated characteristics of JA in dyads where the child was born with NE. Study 2a compared the quantity and quality of JA episodes between the NE and no-NE group. The study did not find any significant differences in JA between the two groups. Previous studies comparing characteristics of JA of those in other clinical groups to dyads with typically developing children find differences in JA are only present when additional factors such as the degree of medical risk or other environmental factors such as household chaos are taken into consideration (Egotubov et al., 2019; Mateus et al., 2019; Sansavini et al., 2015).

Study 2b examined characteristics of JA in the NE group and how the quantity and quality of JA episodes are associated with children's developmental abilities. Overall, relationships between children's processing speed abilities and characteristics of the mother and child during JA were observed. Firstly, it was found that children with poorer processing speed abilities terminated episodes of JA more frequently. Previous studies have shown that children born with NE are more likely to exhibit attention difficulties and poorer processing speed abilities (O'Connor et al., 2017; Tonks et al., 2019). Children with attention difficulties find it difficult to process information quickly and therefore are more likely to lose interest and disengage from JA more frequently (Mundy, 2016).

The study also found that mothers of children with worse processing speed abilities used a higher number of parent-followed bids to successfully initiate JA. This means that mothers initiated JA by referring to an activity on which the child was already focused or interested in. This finding has been previously demonstrated in other clinical groups and it has been observed that children with autism are more responsive towards parent-followed bids for JA (Bottema-Beutal et al., 2018). It might be difficult for children with poor processing speed skills to focus on bids for JA if they are not

interested in the stimuli presented. However, if the child is familiar with an object or activity they may already understand the objective of the bid. Therefore, children born with NE might be more likely to respond to a bid that is relevant to their interests.

Study three examined the quantity and quality of speech used by parents and children during parent-child interaction. The first analysis compared the speech used by mothers and their children born with NE to the speech used by typically developing children and their mothers. It was found that mothers of children born with NE used fewer words per turn compared to mothers of children born without complications. As children born with NE experience difficulties with language and attention (Chin et al., 2019; Tonks et al., 2019) it is suggested that mothers may be pacing their speech to accommodate their child's difficulties. This speech pattern may help encourage the child to speak more often or assist in sustaining the child's attention by breaking down play tasks into steps. The current study also demonstrated that children born with NE used more nouns in their speech compared to typically developing children born without complications. In typically developing children noun bias tends to fade at 3 to 4 years of age (Bates et al., 1994; Frank et al., 2021). It is possible that noun bias is persisting in children born with NE which may indicate difficulties in acquiring and using more advanced language structures and word types.

The second analysis examined the association between characteristics of parent and child speech and the developmental abilities of children born with NE. No relationships were observed between parent speech and child developmental outcomes. However, multiple associations were observed between characteristics of child speech and children's developmental abilities at 4 years of age. Children who scored higher on the verbal comprehension domain of the WPPSI-IV were found to use longer utterances and more complex language. This indicates that speech quality and verbal comprehension

in children born with NE is closely linked as children who use more advanced language skills also possess better comprehension abilities. Children who received higher scores on the processing speed domain of the WPPSI-IV used more words and words per utterance in their speech. Previous research has demonstrated that preterm children with better processing speed abilities were better word learners (Marchman et al., 2022). It is suggested that children with faster cognitive processing abilities produce more speech as they are able to retrieve and express information more rapidly. The current study also found positive associations between the motor abilities of children born with NE and characteristics of speech in that children with higher scores on the Movement-ABC spoke more often and used higher quality language. This finding further supports the link between language and motor abilities in children (Iverson, 2010; Valla et al., 2020), particularly in this cohort of children born with NE.

Study Strengths

The present research reports a number of novel findings that contribute to the literature describing the development of children born with NE. The current study combined the use of standardised assessments that are commonly administered to assess the development of children born with NE with observational methods that have not been previously applied in this clinical cohort. The parent-child interactions of those in other clinical groups have been widely studied (Festante et al., 2019; Wan et al., 2018); however, research had not yet investigated parent-child interactions in the context of NE. Previous research has found parents to also be negatively impacted by NE (Lemmon et al., 2017; Herringhaus et al., 2013); however, the link between parenting behaviours and the development of children in this clinical cohort remained to be investigated. Therefore, this research offers a new perspective as it is the first study to examine qualities such JA and CDS in relation to the development of children born with NE.

There are strengths in the design of the current study. The environment in the Infant and Child Research Lab was designed to prevent distractions and promote naturalistic behaviours during the administration of the developmental assessments and parent-child interactions. The laboratory is a clean, quiet, well-lit room with minimal decorations that provides an ideal setting for the administration of standardised assessments (Del Rosario et al., 2020). The wall-mounted cameras in the laboratory were discreetly placed which allowed parents and their children to engage in naturalistic interactions. The lab environment also enabled the stimuli and environmental factors to be controlled across the participants which facilitated comparability across the study sample (De Barbaro et al., 2013).

Standardised assessments are the most common tool used to assess the development of children born with NE; however, a large proportion of studies do not compare these developmental assessment scores to appropriate control groups (Herrera et al., 2018; Pappas et al., 2015; Perez et al., 2013; Robertson & Finer, 1985). The majority of studies use the normative sample and developmental threshold scores established by the measure to identify developmental delay, but research has found these norm-based threshold scores to underestimate developmental delay (Anderson et al., 2010; Green et al., 2023; Jary et al., 2013). The current study recruited a control group of typically developing children born without complications which allowed the direct comparison of developmental assessment scores to those born with NE. Rather than focusing on the attainment of cut-off scores, the present study was able to identify significant differences in developmental abilities between the two groups.

Another strength of the current study is the analysis of developmental outcome scores by subdomain and the fine-grained analysis of children's speech. The majority of studies that use standardised assessments to measure the development of children born

with NE focus on global development and tend to report only composite scores (Finder et al., 2019; Van Kooij et al., 2010; Van Schie et al., 2015). Domains such as cognition, language and motor abilities are made up of a number of different skills which composite scores may fail to capture. Reporting assessment scores by subdomain and using speech sample analysis provides a more detailed picture of the development of children born with NE. It allows researchers to identify specific areas that children in this cohort need support with and enables the development of NE specific interventions.

Study Limitations

Limitations of the studies in this thesis should also be noted. The main limitation of this research is the small sample size which impacted the statistical power of the study and limited the analysis of the data. The limited sample size meant that more sophisticated analyses such as regression models could not be conducted to estimate the predictive value of the variables investigated in this research. Confounding factors that may have an influence on the results of this study also could not be taken into consideration such as the grade of NE diagnosed. The grade of NE sustained at birth is one of the most important predictors for the development of children born with the condition (Van Handel et al., 2007; Robertson & Finer, 1985). However, NE grade could not be taken into consideration in the current study due to the small sample size and uneven group sizes in each category of diagnosis, with the majority of participants being diagnosed with moderate NE. Previous research proposes that children born with mild NE perform just as well on developmental assessments as typically developing children born without complications (Polat et al., 2013; Robertson & Finer, 1985). However, more recent research suggests that even those with mild NE are at-risk of developmental delay (Conway et al., 2018) and therefore, it was important that children with mild NE were included in the study.

The study's small sample size is partially attributed to high rates of attrition. Over 100 infants born with NE were recruited during the first phase of this research project at the time of diagnosis. For the current study, families who had signed up for the study at birth were re-contacted for follow-up when the children turned 2 years of age or older. Families dropped out of the study for a number of reasons. Many families recruited for the study lived outside the county of Dublin and could not attend the assessment due to the distance of the laboratory from their homes. Some families had moved abroad, others did not remember signing up for the research study and some were uncontactable. Future studies should implement specific measures to maintain rapport with families who sign up for research studies such as sending newsletters to families updating them on the research projects they are part of. Doing so could help improve the follow-up rates within future studies (Bode et al., 2014).

The research project was significantly impacted by the COVID-19 pandemic, which further contributed to the study's small sample size. Fieldwork and data collection for this study was ready to commence just as the pandemic started to take effect. This meant that data collection had to be postponed for over 16 months. When recruitment was permitted several restrictions were implemented which impacted the willingness of families to participate in the study. During some periods the public were not allowed to travel outside their county of residence, therefore only those who lived in Dublin could attend an appointment at the laboratory. Several public health and safety measures were implemented which may have deterred families from travelling to take part in the research study. The initial protocol intended to re-contact and recruit participants to the study as soon as the child turned 2 years of age. However, by the time data collection could resume some of the children were already 4 years of age, which explains the variation in the ages of the children who participated in this study.

Another limitation is the lack of generalisability in the study sample. The majority of the sample were White, highly resourced, well-educated families. This may explain why there were no differences in the quality of CDS and characteristics of JA episodes between the NE and no-NE groups. Furthermore, the current sample was self-selected, particularly those in the no-NE group. Research demonstrates that families who take part in research show characteristic differences to those who do not take part in research. Families who take part in research are typically highly resourced families who have time to participate in research studies (Yu et al., 2020). Parents of children with more severe difficulties may be less inclined to take part in the study as they may not have the time or resources to take part in the research.

Some methodological limitations may have impacted the outcomes of this study. The researcher was not blinded to the medical status of the child and was aware of which participants were born with NE. Some unconscious biases may have occurred during the administration of the developmental assessment. Additionally, the brief play interactions initiated in the lab may not fully represent the daily experience of the parent and child. Whilst there are advantages to using a laboratory setting, they do not replicate the home environment. For example, the child may have siblings that alter the dynamic between the parent and child in day-to-day life. Mothers are aware of being recorded and may therefore act differently in the lab environment, such as being more directive, asking the child more questions or speaking more frequently than they do at home (Bergelson et al., 2018). Despite these limitations, the observation of parent-child interactions is a technique that is widely used in the literature.

Theoretical and Practical Implications

From a family systems perspective, it is suggested that NE does not just impact the individual child but may also implicate the family as a whole (Minuchin, 1974).

Although the clinical symptoms may affect the child on an individual level, the experience of a traumatic birth, the interruption of the parent-infant bond during treatment and uncertainty surrounding the child's outcome (Lemmon et al., 2017) may play a role in how parents and children interact with each other during childhood. This thesis adds to the current literature by investigating fine-grained behaviours that may be characteristic of parents and children born with NE. We find that mothers of children born with NE may display behaviours that support their children's development such using more concise speech patterns so it is easier for children to understand, and using contingent bids to encourage JA.

The majority of previous research focused on the use of standardised assessments to measure the development of children born with NE. This study attempted to examine additional developmental risk factors that may impact child development such as autism traits, EF and sleep problems. Whilst these characteristics have been established in cohorts of children born with NE (Badawi et al., 2006; Zareen et al., 2001) the association of these traits with developmental outcomes had yet to be investigated. The current study found language and motor difficulties to be common amongst those with autism traits and EF difficulties. This adds further insight into the developmental profile of children born with NE and suggests that developmental delay might be linked with behavioural problems in those in this clinical cohort.

The link between language and motor abilities in children born with NE is another prominent finding in this thesis. The relationship between motor and language development in typically developing children has been thoroughly researched (Iverson, 2010; Valla et al., 2020) and motor skills are intrinsically linked with the development of language and communication skills. NE has been found to have a negative impact on children's motor functioning even in those without a diagnosis of a movement disorder

(Jary et al., 2019). The current study has established a link between children's motor abilities and their performance on standardised assessments of language, and also with the quantity and quality of speech used during interaction. Children who perform better on standardised assessments of motor development were also more likely to receive higher scores on the domain of language abilities. Children born with NE who receive superior scores on assessments of motor development are also found to speak more often and use higher quality speech patterns during parent-child interaction. These findings contribute to the literature indicating a connection between children's motor skills and language abilities. Based on these findings, it is also possible to inform intervention plans and it is suggested that children born with NE are offered support with their motor development if early developmental difficulties are observed.

The findings in this thesis have further practical implications that can inform procedures for clinical follow-up and intervention design for children born with NE. The current study identified a subset of children born with NE who exhibit autism traits and EF difficulties using parent-report questionnaires. Children who exhibited these behavioural difficulties were also likely to receive below average scores on developmental assessments. Whilst there are some disadvantages to using parent report questionnaires (Johnson et al., 2008), the use of screening questionnaires may be helpful in some contexts such as assessing children born with mild NE. Some healthcare providers do not have the resources to provide developmental assessments to all children born with NE, however it is imperative that those with developmental delay are identified as early as possible. Screening questionnaires are a quick and inexpensive method that can be used to identify early markers of developmental delay in children born with NE.

Previous research has established that parents of children in clinical groups are less concerned about general prognostic information and are more interested in specific

goal orientated advice to support the development of their children (Maitre & Duncan, 2023). The investigation of parent-child interactions can help inform the future of high-risk follow up as it helps to uncover specific environmental and social determinants that might facilitate the development of children born with NE. In this study we found that children with processing speed difficulties were responsive to contingent bids for JA and that mothers pace their speech when speaking to their child. The investigations of these fine-grained behaviours can help the formulation of specific behavioural interventions for children impacted by NE and can offer parents simple yet effective advice that can improve child outcomes.

Future Directions

The present research aimed to characterise the developmental profile of children born with NE and is one of the first studies to examine parent-child interactions within the context of child NE. Future studies investigating the parent-child interactions of families impacted by NE should take into consideration the grade of NE sustained at birth. The grade of NE has been found to be an important predictor for the outcomes of children (Van Handel et al., 2007; Robertson & Finer, 1985), therefore it would be of interest to see how varying grades of severity are associated with different features of parent-child interaction.

Due to the small sample size of this study it was not possible to use sophisticated analytical methods to analyse the association between parent-child interaction data and developmental assessment scores in this cohort of children born with NE. Previous research has found parent-child interactions to mediate the relationship between neonatal risk and cognitive outcomes of children in other clinical groups (Poehlmann & Fiese, 2001). Regression analyses have also been applied to analyse qualities of parent-child interaction that predict future developmental outcomes (Wan et al., 2013). It would be

useful for future studies to recruit a larger cohort of children born with NE in order to conduct more advanced analyses on the potential mediating pathways. Such analyses would further inform the influence of parent behaviours on child development in those born with NE.

Conclusion

The collection of studies outlined in this thesis adds to the literature that aims to characterise the developmental profile of children born with NE. In order to gain a comprehensive idea of how NE might affect child development, the influence of NE on child and parent behaviours must also be considered. Thus, research analysing the parent-child interactions of those impacted by NE provides an insight into the underlying processes that influence child development. The studies presented in this thesis are some of the first to compare the fine-grained behaviours shown by children born with NE and their parents to typically developing children and their parents. Overall mothers of children born with NE are found to exhibit behaviours that support their children's development such as using more concise speech patterns and using contingent bids to encourage JA. While studies with small sample sizes must be interpreted with caution, this research contributes to the literature seeking to understand the impact of NE on child development.

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Appendices

Appendix A. Contribution to Work

Children born with NE were recruited at birth by medical doctors completing PhDs including Dr. Mary O'Dea (M.O) and Dr. Tim Hurley (T.H) at one of three collaborating maternity hospitals in Dublin, Ireland. When the children were two years of age or older, the author of this thesis, Chelo Del Rosario (C.D.R), contacted families for a follow-up appointment at the Trinity College Dublin (TCD) Infant and Child Research Laboratory. During the follow-up appointment a standardised assessment of development was carried out with the child and parent-child interactions were recorded by C.D.R with the assistance of lab members and PhD candidates Merve Ataman Devrim (M.A.D) and Sarah Coughlan (S.C). Some Bayley-III assessments were carried out by developmental specialist Marie Slevin (M.S) at the Department of Neonatology, NMH and scores were retrieved from children's medical records. The control group consisting of typically developing children born without complication were recruited and assessed by Dr. Linda Kelly (L.K).

Appendix B. Ethical Approval

An tOspidéal Náisiúnta Máithreachais
The National Maternity Hospital
Founded in 1894

Sráid Holles, Baile Átha Cliath 2 • Holles Street, Dublin 2, D02 YH21
Telephone: (01) 6373100. Fax: (01) 6766623. Web: www.nmh.ie

Máistir/Master: Prof. Shane Higgins

PRIVATE AND CONFIDENTIAL

Prof Eleanor Molloy
Neonatology Consultant & Prof. of Paeds,
Trinity College
Dublin

Tuesday 26th November

Re: Neonatal Inflammation and Multiorgan dysfunction and Brain injury research group (NIMBUS)
Our ref: (EC 14. 2016)

Dear Eleanor,

Thank you for your letter regarding the above project. In your letter you are requesting an amendment to include carrying out Bayley's Assessment Scores in infant and toddlers. The request was considered by the Research Ethics Committee on Monday 25th November 2019. This request has received ethical approval.

However, in relation to Holles Street, I would be grateful if you could link up with Marie Slevin, Developmental Psychologist, as some of the children in the study will have been treated at Holles Street. Marie sees all these children for follow up and performs a Bayley's Scrore. It would seem to be unnecessary and perhaps a burden for both the child and the family to have two Bayley's Scores undertaken.

I would be grateful if you could link-up with Marie in due course.

Kind regards,

Yours sincerely,

Dr. John Murphy
Chairman,
Ethics Research Committee

Coombe Women & Infants University Hospital
Excellence in the Care of Women and Babies
Foirfeacht i gCúram Ban agus Naíonán
Cork Street
Dublin 8
telephone +353·1·408 5200
fax +353·1·453 6033
www.coombe.ie

Prof Eleanor Molloy
Chair and Professor of Paediatrics and Child Health
Trinity College Dublin
eleanor.molloy@tcd.ie

6th January 2021

Re: Study No. 2 – 2020 – FIREFLY: Follow up of Inflammatory Responses and multiorgan outcomes FoLlowing neonatal brain injury – revised application

Dear Prof Molloy,

I reviewed the documents supplied by your team on 10th December 2020 for your study entitled '**FIREFLY: Follow up of Inflammatory Responses and multiorgan outcomes FoLlowing neonatal brain injurY'.** Your study is now fully approved by the Research Ethics Committee (REC) in the Coombe Women and Infants University Hospital (CWIUH).

Yours sincerely

Professor Jan Miletin (IMC 241348)
Consultant Neonatologist
Chairman of the Research Ethics Committee
Coombe Women and Infants University Hospital (jmiletin@coombe.ie)

cc. *Dr Johana Isaza-Correa, Research Fellow, The Children´s Research Laboratory, Trinity College Dublin (ISAZACOJ@tcd.ie)*
Prof Afif El-Khuffash, Director of Paediatrics, The Rotunda Hospital (afifelkhuffash@rcsi.ie)

Ospidéal an Rotunda
Cearnóg Parnell, Baile Átha Cliath 1, Éire.
The Rotunda Hospital
Parnell Square, Dublin 1, Ireland.
T: +353 1 817 7700 / F: +353 1 872 6523
www.rotunda.ie

18th June, 2020.

Professor Eleanor Molloy
Consultant Paediatrician and Neonatologist
Paediatrics and Child Health,
Trinity College,
Dublin 2.

Our ref: **REC-2020-005** *(please quote this reference on all correspondence)*
Re: **FIREFLY: Follow-up of Inflammatory Responses and multiorgan outcomes following neonatal brain injury.**

Dear Eleanor,

Many thanks for the amended documentation received in relation to the above research. I am pleased to advise that the requirements set out by the Committee in respect of your study have now been met. This being the case, ethical approval for the research is granted and it may now commence.
You are requested to submit a progress report to the Committee in twelve months, and annually thereafter as applicable.
Please advise us by email to research@rotunda.ie when you have completed your research. We would also like to know when and where you publish or present your results. Please be aware of your responsibilities with respect to the Rotunda Hospital's good research practice policies and guidelines, copies of which are available on the Q-Pulse system.

Kind regards.

Yours sincerely,

Professor Michael Geary,
Chairman,
Research Ethics Committee.

Coláiste na Tríonóide, Baile Átha Cliath
Trinity College Dublin
Ollscoil Átha Cliath | The University of Dublin

F.A.O. Chelo Del Rosario
Approval ID: SPREC092014-01

School of Psychology Research Ethics Committee

SCHOOL OF PSYCHOLOGY
Arás an Phiarsaigh
Trinity College
Dublin 2

28th January 2020

Dear Chelo,

The School of Psychology Research Ethics Committee recognises you as a named researcher on the project entitled "Parent-child interactions and child development outcomes" which received ethical approval in September

Adverse events associated with the conduct of this research must be reported immediately to the Chair of the Ethics Committee. The safety protocols attached must be observed.

Yours sincerely,

Richard Carson
Chair,
School of Psychology Research Ethics Committee

Safety protocol for adults

Scoil na Síceolaíochta
Dámh na nEolaíochtaí Sóisialta agus Daonna,
Áras an Phiarsaigh,
Coláiste na Tríonóide,
Baile Átha Cliath,
Ollscoil Átha Cliath,
Baile Átha Cliath 2, Éire.

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Faculty of Arts, Humanities and Social Sciences,
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Appendix C. Information Sheet

Health Research Board

Trinity College Dublin
Coláiste na Tríonóide, Baile Átha Cliath
The University of Dublin

PANDA PROJECT: **P**sychological **a**nd **N**euro**d**evelopmental **A**ssessment of Neonatal Encephalopathy

You and your child are invited to join a research study to help understand the development of children born with Neonatal Encephalopathy. The study is being conducted by researchers at the Schools of Psychology and Medicine at Trinity College Dublin. Please read this information leaflet carefully and please get in touch with us if you have any questions about participating in the research. The decision for yourself and your child to join, or not to join, is totally up to you.

Contact us at:

Chelo Del Rosario infantresearch@tcd.ie

Dr Elizabeth Nixon enixon@tcd.ie – 01 896 2867

Dr Jean Quigley quigleyj@tcd.ie- 01 -896 2697

Site	Trinity College Dublin (TCD)
Principal Investigator and Co-Investigators	Dr. Elizabeth Nixon & Dr. Jean Quigley, Assistant Professors in Psychology, TCD Prof. Eleanor Molloy, Professor of Pediatrics, School of Medicine, TCD. Chelo Del Rosario, Postgraduate Student, TCD
Data Controller	Trinity College Dublin
Data Protection Officer	Data Protection Officer Secretary's Office Trinity College Dublin, Dublin 2.

This leaflet provides information on the research study and aims to answer any questions you may have

Part 1 – The Study

Who are we and what do we do?

We are a team of lecturers, researchers and psychology students at the School of Psychology, Trinity College Dublin. We do research in our Infant and Child Lab on child development and parenting.

What is the study about?

We are carrying out a study of families affected by Neonatal Encephalopathy (NE). This is a condition that affects a very small number of children born every year. The effects of NE on children's development is not very well understood and that is why we want to assess the development of children affected by NE. We have called the project the PANDA project. The Health Research Board has funded a programme of research on NE (called the NEPTUNE project). PANDA is one of the projects and it stands for **P**sychological **a**nd **N**euro**d**evelopmental **A**ssessment of Neonatal Encephalopathy

What is involved in the taking part in the study?

Your child will complete a standard developmental test (we measure children's language, thinking skills, and motor skills (by this we mean being able to crawl, walk, pick up objects, etc). Some of the tasks that we ask your child to do will be very easy for them and some will be very hard – this assessment will take up to 90 minutes to complete but we make sure that you and your child get a lot of breaks so that your child does not get too tired. We will also observe your child and the child's mother playing with each other, as you ordinarily would at home, and will ask to video record you and your child playing with each other. We will provide specific toys at certain times, and at other times you will be given a box of toys where you and your child can choose what to play with and how to play. In total, the session will last approximately 2 to 3 hours. You will be present with your child at all times. Before we invite you in for the assessment, we will send you a series of questionnaires that we will ask you to complete online. The questionnaires will include questions about yourself, your child and your partner, for instance on your stress levels, your mood, the quality of your relationship with your partner (if relevant), your child's personality and behaviour.

We may want to follow-up with you and your child to see how things are progressing, after one year. We will re-contact you after the data collection to see if you would be interested in taking part in another assessment. However, just because you took part in one stage of the study does not mean that you have to take part in a later stage – you can change your mind

about taking part at any time. We will also ask permission to access the medical records held on your child.

We will contact you the day before your appointment and we will ask you to complete a screening questionnaire 24 hours and on the morning of visit prior to your visit to ensure you or your child are not experiencing any symptoms of COVID-19. We will also store your contact details for 28 days for the purpose of contact tracing and we require you to download the HSE contact tracing app. During the assessment you will be required to wear an FFP2 mask which we will provide for you. Your child is not required to wear a face covering.

Are there any risks involved in taking part in the study?

No risk is foreseen for you and your child's participation in the study beyond those experienced in everyday life. The room in which we conduct the study is a child-friendly space. There is a risk your child may become tired and frustrated with some of the tasks – we can stop at any time and you can take breaks whenever you need. Some of the questionnaires may seek information that is personal and sensitive – you do not have to answer any questions that you do not wish to.

Do I have to take part?

You don't have to take part in this study. It is entirely voluntary - it is entirely your choice whether or not to participate. You can leave the study at any time without giving a reason and without consequence.

What if I change my mind about taking part?

To leave the study you just need to inform the researchers involved in the study, either while the study is taking place or after it has finished. Our contact information is included on this form. If you decide to leave the study, we will delete all information we store about you.

What will happen to the findings of the study?

All information which we collect will be treated in the strictest of confidence and will be stored safely and securely (see the next section for details on what we do with your data). The results arising from the study will be included in a thesis and will be used in publications and presentations. Names and other personally identifiable information will not be used in any outputs arising from the research.

Will I get feedback about the study?

When the study is completed, we will provide an update to participants on the results of the study, which will be in summary (average) form.

Will the research benefit me and my child?

You or your child will not benefit directly from taking part in this research project.

Part 2 – Data Protection

What information about me (personal data) will be used as part of this study?

For this study, we will gather personal information such as your name, your email address and phone number. We will also video-record you and your child interacting and playing with each other –this is classified as personal data because your and your child's voice and your face will be recorded. We will also ask you to complete questionnaires about yourself and your child, relating to things such as your stress levels, your mood, the quality of your relationship with your partner (if relevant), your child's personality and behaviour

What will happen to my personal data?

We will use the personal information (names and contact details) to keep in touch with you about the study and to contact you regarding follow-up meetings. We will retain personal data and the video recordings for a period of seven years after collection. This ensures that we have enough time to analyse the very rich data that we have collected and can continue to use the data in our follow-up studies.

Who will access and use my personal data?

Only the members of the study team listed on this information sheet and TCD students who have been recruited to work under their supervision listed will have access to your personal data. Anybody who works with the data will be Garda Vetted and have undergone training in ethics and data protection. No personal information will ever be shared with individuals outside the institution of TCD.

Will my data be kept confidential? How will my data be kept safe?

Your privacy is very important to us. We take many steps to make sure that we protect your confidentiality and keep your data safe.

- Any information we collect from you is entirely confidential and is used only for the purpose of the research. A unique study ID code will be assigned to you and your study information will be gathered, stored, and accessed using this ID code only. Personal data (names, postal addresses, email address, phone numbers) will be stored separately in a password-protected and encrypted database. Only researchers who are part of this study team will have access to these data. Signed consent forms will be stored in a locked cabinet in a locked office.
- It is not possible for us to anonymise the data – we do not want to delete your names and contact details because we will want to be able to contact you about follow-up in the future (you are not obligated to take part in any follow-up, unless you want to). Also,

it is not possible to anonymise the video-recordings of your data because your voice and face will be heard/seen.

- However, we will pseudo-anonymise the data - that is, we will separately store the file that links your unique study ID code with participant identity.
- Analyses will be conducted on aggregate or anonymised data - no information that could potentially identify you will be included in any reports or presentations.
- We will never share data with researchers at any institution other than TCD.
- Training in data protection law and practice has been provided to those individuals who are responsible for the research (Dr. Elizabeth Nixon and Dr. Jean Quigley).

Will the confidentiality of my data ever be breached or shared with a third party?

There are certain circumstances under which confidentiality may be breached. First, if you tell us something or behave in a way that indicates you or your child are at risk, we will need to let the appropriate services know (e.g., Tusla, the Gardaí). This is required by law. We will not do this without informing you first.

Another circumstance is where disclosure is required as part of a legal process or Garda investigation. In such instances, information may be disclosed to appropriate third parties without permission being sought. Where possible, a full explanation will be given to you regarding the necessary procedures and the actions that may need to be taken.

What are my rights in relation to my data?

You are entitled to:

- Access to your data and receive a copy of it.
- Restrict or object to processing of your data.
- Object to any further processing of the information we hold about you (except where it is fully anonymised).
- To have inaccurate information about you corrected or deleted.
- Receive your data in a portable format and to have it transferred to another data controller.
- Request deletion of your data.

You can exercise these rights by contacting the study team or the Trinity College Data Protection Officer, Secretary's Office, Trinity College Dublin, Dublin 2, Ireland. Email: dataprotection@tcd.ie. Website: www.tcd.ie/privacy.

Part 3 – Costs, Funding, and Approval

Has this study been approved by a research ethics committee?

This study has been approved by the School of Psychology Research Ethics Committee (SPREC) at Trinity College Dublin on 29th January 2020 and by [name of hospital ethics committee] on [DATE].

Who is organising and funding this study?

The study is funded by the Health Research Board, but the study is independently conducted by the team of researchers at Trinity College Dublin.

Will I be paid for taking part? Will it cost anything to take part?

There is no payment for participation in this study, nor are there any costs, aside from any costs incurred in travelling to Trinity College.

Part 4 - Future Research

Will my personal data be used in future studies?

No, not without your explicit consent. As stated above, the research we do involves tracking children and their families over time, so that we can understand how children's early circumstances influence how they get on at a later age. Thus, we may want to contact you to in the future to see how your child is getting on. We will never use any of your data for future studies without contacting you and seeking your permission first.

Part 5 - Further Information

Who should I contact for information or complaints?

If you have any concerns or questions, you can contact:

- *Principal Investigators:* **Dr. Elizabeth Nixon***,* School of Psychology, Trinity College, Dublin 2. Email: enixon@tcd.ie. Phone: 01 8962867; **Dr. Jean Quigley**, School of Psychology, quigleyj@tcd.ie; 01 896 2697.
- *Data Protection Officer, Trinity College Dublin: Data Protection Officer, Secretary's Office, Trinity College Dublin, Dublin 2, Ireland. Email: dataprotection@tcd.ie. Website: www.tcd.ie/privacy*.

Under GDPR, if you are not satisfied with how your data is being processed, you have the right to lodge a complaint with the Office of the Data Protection Commission, 21 Fitzwilliam Square South, Dublin 2, Ireland. Website: www.dataprotection.ie.

What should I do now?

Within a few days of receiving this information sheet, we will be in touch with you to discuss your potential participation. If you wish to contact us before then, you can email us at infantresearch@tcd.ie or call the study Principal Investigators (Dr Elizabeth Nixon or Dr Jean Quigley) on 01 896 2867.

Appendix D. Consent Form

CONSENT FORM

PANDA PROJECT: **P**sychological **a**nd **N**euro**d**evelopmental **A**ssessment of Neonatal Encephalopathy

Site	Trinity College Dublin
Principal Investigator and Co-Investigators	Dr. Elizabeth Nixon & Dr. Jean Quigley, *Assistant Professors in Psychology,* Trinity College Dublin Prof. Eleanor Molloy, *Professor of Pediatrics,* Trinity College Dublin Chelo Del Rosario, *Postgraduate Student,* Trinity College Dublin
Data Controller	Trinity College Dublin
Data Protection Officer	Data Protection Officer Secretary's Office Trinity College Dublin Dublin 2

There are 4 sections in this form. Each section contains a number of statements. You are asked to write your initials in the box beside the statement if you agree. If you do not agree with a statement, please leave the box blank. The end of this form is for the researchers to complete.

If you have <u>any</u> questions, please don't hesitate to call or email the lead researchers – Elizabeth Nixon (01 8962867; enixon@tcd.ie) or Jean Quigley (01 896 2697; quigleyj@tcd.ie). Thank you for participating.

GENERAL	**Initials**
I confirm I have read and understood the **Information Leaflet** for the above-named study. I have been given the contact details of the lead researcher to ask questions. Where I had questions, they have all been answered to my satisfaction.	
I understand that participating in this study **is entirely voluntary, and if I decide that we do not want to take part, we can stop taking part in this study at any time without giving a reason**.	

I understand that we **will not be paid for taking part in this study** and that there will be no direct benefits to us from taking part in the study.	
I know how to **contact the research team** if I need to.	
I agree to take part in this study with my child having been fully informed of the **risks, benefits and alternatives** which are set out in full in the information leaflet with which I have been provided. This includes risk associated with the COVID-19 pandemic, including the transmission of the virus. However, every effort will be made to mitigate these risks, including temperature checking, physical distancing, mask-wearing and hand-sanitising."	
I agree that I and my child can be **contacted by researchers by email and phone** as part of this research study.	
I agree to be video-recorded playing with my child.	

DATA PROCESSING	**Initials**
I give my permission for my own and my child's data to be processed in line with the aims of the research study, as outlined in the information leaflet.	
I agree that the researchers may be granted access to the medical files held about my child's NE.	
I understand that I am entitled to access any data that is held about me or my child.	
I understand that **personal data will be collected and stored as part of this research project.** The consent forms, my contact data, the video-data, and the completed questionnaires/medical data (if applicable) will be stored separately from each other, and a unique study ID to which only the lead researchers have access will be used to link the different pieces of data.	
I understand that the personal information collected in the study will be kept **strictly confidentia**l and will only be made available to researchers who are part of the **study team**.	
I understand that **confidentiality may be breached in circumstances in which**: 1. The research team has a strong belief or evidence exists that there is a serious risk of harm or danger to either the participant or another individual. This may relate to issues surrounding physical, emotional and/or sexual abuse, concerns for child protection, rape, self-harm, suicidal intent or criminal activity. 2. Disclosure is required as part of a legal process or Garda investigation. In such instances, information may be disclosed to significant others or appropriate third parties without permission being sought. Where possible, a full explanation will be given to the participant regarding the necessary procedures and also the intended actions that may need to be taken.	

I understand that **I can withdraw my permission for me/my child to take part in this study** at any time without giving a reason. I understand that in this case, the researchers will delete all information related to us and we will be removed from the study.	

RETENTION OF INFORMATION	
I understand that my data may be retained by the study team for seven years and that we may be contacted within the seven years about follow-up stages of the research.	

SHARING OF INFORMATION	
Data will not be shared with researchers outside of the study team. The data will only be used for research projects related to the goals of this study.	

Child Name (Block Capitals)

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Parent/Guardian Name (Block Capitals) Parent/Guardian Signature Date

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Lead Researcher Name (Block Capitals) Lead Researcher Signature Date

To be completed by the Principal Investigator or nominee and retained by parent/guardian

I, the undersigned, have taken the time to fully explain to the above participant the nature and purpose of this study in a way that they could understand. I have explained the risks and possible benefits involved. I have invited them to ask questions on any aspect of the study that concerned them.
I have given a copy of the information leaflet and consent form to the participant with contacts of the study team.

Researcher name:

Title and qualifications:

Signature:

Date:

Tel: 01 8962867

Email: infantresearch@tcd.ie

This sheet to be retained by guardian/parent of participant