University of Southampton

Faculty of Environmental and Life Sciences

School of Psychology

Maternal Birth-Related Stress and School-Age Emotional and Behavioural Outcomes of Children with Hypoxic-Ischemic Encephalopathy

by

Jasmine Victoria Slinger

Thesis for the degree of Doctorate in Educational Psychology

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Abstract

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Chapter One introduces the researcher's background, research context and theoretical paradigm. Against the background of an emerging evidence base, Chapter Two of this thesis outlines a systematic literature review of the existing evidence which investigates the association between birth-related post-traumatic stress (PTS) and parent-child relationship quality in the first two years of life. A comprehensive, systematic search identified 24 quantitative studies that examined parental birth-related PTS in relation to indicators of parent-child relationship quality. Sample sizes ranged from 19 to 2802 parents and studies utilised a range of measures and designs. The 24 studies included both general population samples and high-risk birth populations, such as parents of pre-term infants. Several domains of parent-child relationship quality were investigated including: bonding, maternal behaviours and child behaviours. Some evidence suggested that parental birth-related PTS is associated with less optimal parent-child relationship quality, such as less maternal warmth and greater maternal anxiety towards the infant. However findings were inconsistent across all domains of parent-child relationship quality. The inconclusive findings and the heterogeneity of the reviewed papers as a whole limits the conclusions that can be drawn. Further and more rigorous research is needed to advance our current understanding of how parental birth-related PTS may influence the parent-child relationship. Further study is also needed to advance our current understanding of partner's birth-related PTS on the parent-child relationship, as current evidence is scarce.

The empirical paper (Chapter Three) assessed maternal birth-related stress and school-age emotional and behavioural outcomes in children with neonatal hypoxic-ischemic encephalopathy (HIE) treated with therapeutic hypothermia (TH) compared with typically developing peers aged six to eight years (mean age 6.94) participated. Mothers completed measures of birth-related stress. Children's emotional and behavioural difficulties were completed by parents (n = 45 for the HIE group, n = 28 for the control group) and teachers (n = 28 for the HIE group, n = 21 for the control group). Mothers of

children with HIE reported significantly greater levels of birth-related PTS (p>.001). Parental and teacher reports of children's emotional and behavioural difficulties found that children displayed significantly greater internalising and externalising difficulties at home but not at school. Maternal birth-related distress fully mediated the association between HIE and parental reported internalising difficulties, but not externalising difficulties. Our findings indicate that HIE is associated with increased risk of birth-related distress in mothers, and increased risk of emotional and behavioural difficulties in children. Implications for Educational Psychologists (EPs) and key stakeholders are discussed.

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Research Thesis: Declaration of Authorship

Print name: Jasmine Slinger

Title of thesis: Maternal birth-related stress and school-age emotional and behavioural outcomes of children with hypoxic-ischemic encephalopathy

I declare that this thesis and the work presented in it are my own and has been generated by me as the result of my own original research.

I confirm that:

1. This work was done wholly or mainly while in candidature for a research degree at this University;
2. Where any part of this thesis has previously been submitted for a degree or any other qualification at this University or any other institution, this has been clearly stated;
3. Where I have consulted the published work of others, this is always clearly attributed;
4. Where I have quoted from the work of others, the source is always given. With the exception of such quotations, this thesis is entirely my own work;
5. I have acknowledged all main sources of help;
6. Where the thesis is based on work done by myself jointly with others, I have made clear exactly what was done by others and what I have contributed myself;
7. Parts of this work have been published as:-

   Collins, F., Cianfaglione, R., & Vollmer, B. (January 2022). *Behaviour in toddler- and school-age children with a history of neonatal hypoxic-ischaemic encephalopathy* [Poster presentation]. Royal College of Paediatrics and Child Health, Southampton, UK.

Signature: ..... Date: 04.06.2022

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Finally, I would like to thank my family and friends, for their support, patience, and belief in me throughout my educational psychology journey.

Definitions and Abbreviations

α ..... Chronbach's alpha reliability statistic

b ..... Unstandardised regression coefficient

CBCL ..... Child Behavior Checklist

CBCL-I ..... Child Behavior Checklist – internalising problems

CBCL-E ..... Child Behavior Checklist – externalising problems

CI ..... Confidence interval

d ..... Cohen's d, measure of effect size

EP ..... Educational Psychologist

ERGO ..... Ethics and Research Governance Online

HIE ..... Hypoxic-ischemic encephalopathy

M ..... mean

MAI ..... Maternal Attachment Inventory

MIBS ..... Mother-to-infant Bonding scale

MORS ..... Mother's object relation scale

MPAS ..... Maternal Postnatal Attachment Scale

n ..... Number of participants

NENAH ..... Neurodevelopmental trajectories and neural correlates in children with neonatal HIE

NICU ..... Neonatal intensive care unit

P ..... Probability value

PBQ ..... Postpartum Bonding Questionnaire

PCDI ..... Parent-child Dysfunctional Interaction

PIPE ..... Paediatric Infant Parent Exam

PPD ..... Postpartum depression

PPQ ..... Perinatal Post-traumatic Stress Disorder Questionnaire

PRISMA ..... Preferred Reporting Items for Systematic Reviews and Meta-Analysis

PTS ..... Post-traumatic stress

PTSD ..... Post-traumatic Stress Disorder

r ..... Spearman's correlation coefficient

SD ..... Standard deviation

SE ..... Standard error

SDQ ..... Strengths and Difficulties Questionnaire

STROBE ..... Strengthening the reporting of observational studies in epidemiology

t ..... test statistic for independent t-test

TH ..... Therapeutic hypothermia

TRF ..... Teacher's Report Form

TRF-I ..... Teacher's Report Form – internalising problems

TRF-E ..... Teacher's Report Form – externalising problems

Chapter 1 Introduction

1.1 Overview

The overarching aim of this thesis was to explore school-age outcomes for children and parents following hypoxic-ischemic encephalopathy (HIE). A systematic literature review (Chapter 2) aimed to investigate whether the current literature evidences a relationship between parental birth related post-traumatic distress (PTS) and parent-child relationship quality. My empirical project (Chapter 3) was part of a wider project which aims to improve understanding of school-age outcomes in children with HIE and who received therapeutic hypothermia (TH) who survive without major neuro-disability. The specific focus of my empirical project was on maternal birth-related PTS and children's internalising and externalising behaviours, as reported by parents and teachers.

1.2 Context for research

HIE is a key contributor to infant mortality, brain injury and cerebral palsy (Gale et al., 2018; Kurinczuk et al., 2010 Wu et al., 2006). HIE is reported to occur in approximately 2.96 per 1000 live births in England and Wales (Shipley, Gale & Sharkey, 2021). In 2010, therapeutic hypothermia (TH) became standard of care for infants with moderate to severe HIE (NICE, 2010), following evidence highlighting TH to improve the outcomes of infants with HIE (Azzopardi et al., 2014; Jacobs et al., 2013). However, most of this research has predominantly examined outcomes in infancy. Investigating the school-age outcomes of this population has become an emerging, and important area of interest in research.

1.3 The wider project

My empirical paper is part of a larger research study: neurodevelopmental trajectories and neural correlates in children with neonatal HIE (NENAH). As a member of the research team, I have been involved in various aspects of the NENAH study that do not specifically relate to my empirical project.

For example, although my empirical project utilised data from parent and teacher questionnaires, I also supported with collecting data for the wider project, such as completing assessments with children at Southampton General Hospital and via Microsoft Teams. The assessments I helped to carry out included subtests of the following: Wechsler Intelligence Scale

for Children – V; Wechsler Individual Achievement Test; Rivermead Behavioural Memory Test for Children; Children's Memory Scale; Neuropsychological Assessment – 2nd Edition; The Pictorial Scale of Perceived Competence and Acceptance for Young Children; Self Perception Profile for Children. The assessments within the hospital were usually carried out for either a half or full day, as appropriate. The assessments completed via Microsoft Teams took approximately 60 to 90 minutes to complete, as it only involved assessments that were suitable to complete online (Wechsler Individual Achievement Test and the appropriate assessment of self-esteem). Initially, I observed the study co-ordinator, so that I could familiarise myself with the assessments. I then carried out assessments under the supervision of the study co-ordinator. Following this, I completed assessments independently. In total, I have been involved in approximately 17 assessments in the hospital (a mixture of half and full days) and 16 assessments via Microsoft Teams.

Another aspect of the study I was involved in was supporting with the recruitment of control participants through contacting eligible schools. In addition, I assisted with contacting teachers regarding questionnaire measures and monitored their responses. I then focused on completing data entry for the measures used in my empirical project (Chapter Three), as well as the measures of self-esteem.

Data collection for the wider project is ongoing. Thus, for the empirical paper, the results are based on the data that was collected at the time of data analysis (April 2022). Based on G* Power 3.1 software, a minimum anticipated sample size of 64 per group is required in order to detect a medium effect size (0.50; Cohen, 1988) for .80 power. Thus, the current data analysis for parental reports is based on 70.31% and 43.75% of the anticipated HIE and control sample respectively. I plan to incorporate the full dataset from the final sample into my empirical paper for publication.

1.4 Researcher's background and rationale for engagement

My earliest memories of a keen interest in the psychology related to children's social and emotional development and the role of the family relate to my watching of the BBC documentary 'The House of Tiny Tearaways'. Consequently, I completed an undergraduate psychology degree, where I was fascinated to learn about how early life experiences can have a profound influence on children's development. The focus of my dissertation aligned with these interests and involved exploring friendship quality and mental health difficulties in looked-after children. Upon completing my undergraduate degree, and prior to becoming a Trainee Educational Psychologist (EP), I worked on a 1:1 basis with a looked-after child in a school. Here, I was able to use and apply

my knowledge of Attachment Theory (Bowlby, 1969, 1982) as a way of understanding and supporting the child's behaviour. Within this role, it became apparent that there was a lack of knowledge about attachment amongst the wider school staff team. I realised that part of my responsibility in advocating for the child included sharing this knowledge and information more widely. I was able to see first-hand just how beneficial this understanding was to effectively support the child and help them to access their education. This was a marked motivation for wanting to advocate for children who had experienced traumatic events early in life and enhance wider understanding among school staff.

Therefore, within my current practice as a Trainee EP, I aim to adopt a holistic view of the child within their context and gain an understanding of their journey so far. The Integrated Factors Framework (Frederickson & Cline, 2009) has been a central aspect of my practice in guiding my hypotheses. Over the course of my training, I have further learnt to appreciate the importance of asking parents about the child's birth and early life experiences within consultations. For example, as a Trainee EP, I have been involved with pieces of casework where social, emotional, and mental health needs have been raised as a concern, and where parents and carers have shared important information about the family background during early childhood, such as the child being exposed to traumatic experiences, or parental experiences of postnatal mental health difficulties. Therefore, understanding the child's behaviour within the context of these early life experiences has been crucial; had this information not been shared, a different, and potentially inappropriate formulation may have been developed.

I reflected that, although I had some awareness of postnatal mental health difficulties such as postnatal depression, my knowledge of birth-related post-traumatic stress disorder (PTSD), was far more limited. I therefore used my systematic review as an opportunity to increase my understanding of the association between birth-related post-traumatic stress (PTS) and parent-child relationship quality in the first two years of life. I decided to focus on early parent-child relationship quality, given the vast evidence demonstrating its relationship with children's emotional and behavioural development; should there be support for an association between birth-related PTS and indicators of less optimal parent-child relationship quality, it would be plausible to hypothesise that birth-related PTS could be associated with greater emotional and behavioural difficulties. Furthermore, I decided to focus on PTS rather than PTSD, as I wanted to include parents who may be experiencing elevated levels of distress and post-traumatic symptoms related to the birth, but who may not have received a formal diagnosis.

Research as part of my review demonstrated pregnancy and birth complications to be a key risk factor for PTSD following childbirth (Yildiz et al., 2017). However, within my systematic search,

only a few papers explored this relationship in the context of 'high-risk' births, and this was limited to parents of pre-term and low birthweight infants. This therefore highlighted the lack of recognition, or evidence relating to the prevalence of birth-related PTS in parents of infants with HIE, let alone its potential implications for child development. As such, the empirical paper offered a chance to contribute novel research to this evidence base.

Prior to my thesis, I had never heard of HIE. However, when my supervisor informed me of the wider project and presented me with the opportunity to join the project, I was interested to learn more about this group of children and their families. The research I have helped to conduct, and the associated reading I have carried out, has highlighted to me just how vulnerable this group of children and their families may be, both in relation to parental mental health and child development. Interestingly, when discussing my research with other EPs, most have also been unfamiliar with the term HIE. I therefore view my thesis as a piece of research which makes an important contribution to Educational Psychology practice and helps to raise awareness of this group of children.

Taken together, these two studies were conducted to inform understanding around parental birth-related distress and its association with both parent-child relationships and child emotional and behavioural development. While the systematic review focused on the relationship between parental birth-related PTS and parent-child relationship quality, the empirical project specifically focuses on parental birth-related PTS and its association with child outcomes in children with HIE. Developing this understanding has potential scope to improve awareness, support, and outcomes for parents, as well as children with HIE. The results have implications for school staff, EPs, and health practitioners.

1.5 Research paradigm

Within my thesis, I adopted a post-positivism research paradigm. Post-positivism balances both positivist and interpretivist stances (Panhwar et al., 2017). Whilst a post-positivist stance tries to test hypotheses, it also maintains that research findings do not result in an overall 'truth'. Furthermore, it recognises the complexity of experiences (Ryan, 2006).

A post-positivist paradigm was adopted in the current thesis, and as such, informed my theoretical position as a critical realist. Critical realism (Bhaskar, 1975) assumes that our knowledge of the external world remains fallible and is open to revision (Collier, 1994). By adopting a critical realist position in my research, I endeavoured to identify relationships that would allow me to understand how HIE influences maternal mental health and children's development. I then wanted to use these findings to make wider generalisations and predictions

about the impact of HIE on children and parents. However, I appreciated that the findings of my empirical project (whether they proved or disproved my hypotheses) would not represent a 'truth' for all children with HIE and their parents. Future research may not replicate the findings of my empirical project and may indeed provide conflicting evidence. Furthermore, I recognised that my findings may be influenced by several other factors that were not specifically accounted for within my project.

1.6 Reflective learning

There have been many benefits to working as part of a team for my thesis and I am grateful to have had the opportunity to work alongside a dedicated and knowledgeable team of others. However, there have also been hurdles that I have had to overcome, which have been important learning points for myself. For example, I have learnt to become more assertive and put boundaries in place. Being assertive, particularly within my professional life, is not something that comes easily to me. I am someone who always aim to please others and therefore I do not like to say 'no'. However, through my thesis journey, I have learnt the importance of asserting my boundaries. Given the large scope of the wider project, there were several demands placed upon me, which I did not always feel were feasible for me to fulfil within the time I had allocated to my thesis. I therefore had to negotiate with the team what was going to be realistic for me to assist with, whilst also ensuring that this met their expectations. Being able to assert my boundaries and negotiate my work will be an important part of my role as a qualified EP, and although it has not always been easy for me, it is a skill I will be able to take forward into my practice.

Seeking both practical and emotional support from others has also been crucial throughout my journey. This has included support from my supervisors, personal tutor, family and friends, and fellow Trainee EPs. Moving forward into my role as an EP, I feel confident to seek such support, and recognise the importance of this, both for my professional development, as well as to maintain a sense of positive wellbeing.

1.7 Dissemination plan

I have written two research papers in this thesis with an intention to publish in peer-reviewed journals. As such, the papers have been written in the style required for submission to the journals I am currently considering. The journal I am considering submitting my systematic literature review to is '*Infant Mental Health*'. This is a peer-reviewed journal with a focus on influences on early social and emotional development, including caregiver-infant interactions. The journal I am considering submitting my empirical paper to is '*Child Development*'. This is also a

peer-reviewed journal, with a broader focus on various topics within the field of child development. This journal was chosen due to its multidisciplinary target audience, including educational psychologists.

Chapter 2 What is the association between birth-related post-traumatic stress and parent-child relationship quality in the first two years of life? A systematic review.

2.1 Introduction

The birth experience can be a distressing and/or traumatic event for some women, particularly if there is perceived threat of serious injury or death to the mother or infant. As a result, childbirth can be a trigger for post-traumatic stress disorder (PTSD). Even without adverse obstetric emergencies, subjective distress during birth has been evidenced as a significant risk factor for childbirth-related PTSD (Ayers et al., 2008; Garthus-Niegel et al., 2013).

In addition to exposure to actual or threatened death or serious injury (which can be direct, witnessed or indirect), PTSD consists of experiencing symptoms which are characterised by distinctive clusters: intrusion, avoidance, negative cognitions and alterations in arousal. A diagnosis of PTSD is based symptoms which have lasted for at least a month (American Psychiatric Association, 2013). Prevalence rates of childbirth-related PTSD in women are greater in high-risk samples than general population samples, with rates of around 18.5% in comparison to 4% (Yildiz et al., 2017). High-risk samples include those who delivered babies at a low birthweight, pre-term, or who had severe pregnancy or birth complications (Yilzdiz et al., 2017). However, it has been highlighted in the literature that many women may not meet the diagnostic criteria for PTSD, yet still experience significant symptoms. Thus, current reported rates may under-estimate the prevalence of post-traumatic stress (PTS) symptoms experienced by women following birth (McKenzie-McHarg et al., 2015).

Women's partners who are present at the childbirth can also be affected by a traumatic experience. While limited literature has investigated prevalence rates, there is evidence which highlights that a small proportion of fathers may experience PTS symptoms in the first year following their child's birth (Bradley & Slade, 2011). Qualitative research by Etheridge and Slade (2017) found that men who experienced the birth as traumatic described it as a "rollercoaster of emotions" (p.11), with feelings of uncertainty, anxiety and helplessness. To the author's knowledge, no research has specifically explored the prevalence of PTS in partners who were not present at the birth (i.e., indirect exposure).

The experience of childbirth-related PTS in parents may have negative implications for the child's development. Postnatal maternal mental health and distress is associated with poorer developmental outcomes in a variety of domains (e.g., cognitive, social, emotional and behavioural) in childhood through to adolescence (Kingston & Tough, 2014; Murray et al., 2010; Murray et al., 2011; Netsi et al., 2018). Longitudinal research has also found father's postnatal distress to be significantly associated with later emotional and behavioural difficulties in children (Giallo et al., 2014; Ramchandani et al., 2008). It must be noted that research has predominantly focused on postpartum depression (PPD) in parents. However, the impact of PTS symptoms and PTSD related to birth on child development is currently unclear.

An important mediator of the association between parental postnatal mental health difficulties and child outcomes is the quality of the parent-child relationship (Giallo et al., 2014; Rominov, et al., 2016). Attachment theory (Bowlby, 1969;1982) places emphasis on the quality of early parent-child interactions for children's development. Specifically, infants are dependent on responsive and sensitive caregiving in order to have their needs met and gradually make sense of the world around them. The theory has since been supported by a large body of research which indicates significant associations between children's early attachments and their later social, emotional and cognitive development (Groh, et al., 2012; Madigan et al., 2013). Parental warmth and sensitivity (i.e., affection, comfort, concern, nurturance; Lee et al., 2018), core features of responsive caregiving required for a secure attachment (Ainsworth et al., 1978), have consistently been found to be associated with better psychological adjustment in children (Khaleque, 2013; Raby et al., 2015).

The emphasis on the importance of early experiences for development in attachment theory has since been supported by neuroscientific findings, which demonstrate significant brain growth and neural pruning during the first two years of life (Fox et al., 2010; Winson & Chicot, 2016). The brain is considered to be particularly sensitive and responsive to experience during this period with both brain structure and function being shaped by experience in important ways (Malekpour, 2007; Schore, 1998; Schore, 2001). This experience-dependent nature of brain development (Meany & Szyf, 2005) coupled with the infants' dependence on caregivers, highlights the central role that early parent-child dyad relationships play for human development.

The experience of postpartum mental health difficulties can hinder the personal psychological resources of the parents required to care for and meet the needs of the child, a crucial factor contributing to the parent-child relationship (Belsky, 1984). Research has consistently found PPD to be related to suboptimal parental behaviours, including lower sensitivity and affection towards their child (Bernard et al., 2018; Stanley et al., 2004). In a review

of the literature, Stein et al., (2014) identified longitudinal evidence which demonstrated parents experiencing psychological distress in the postpartum period to be less able to consistently and sensitively respond to their child's cues, as well as understand their child's thoughts and feelings. Qualitative research has also explored the challenges experienced when parenting with PPD. In a study by Barr (2008), women shared their difficulties adapting to the role of a mother. While they were able to complete caregiving tasks for the infant, they did so in an "automatic and non-thinking manner" (p.366). In line with this, PPD has been found to be associated with reduced rates of secure attachment (Martins & Gaffan, 2000), and increased rates of insecure mother-infant attachment (Stewart & Vigod, 2016). It is important to note that there is limited literature on the impact of paternal PPD. Where research has included fathers, this has mainly consisted of exploring associations between maternal PPD and the father-child relationship (Goodman, 2008).

There is also some research which draws attention to the effects of PTSD on parent-child relationships. In a systematic review of the literature, Van Ee et al. (2015) found that, in most studies, parents experiencing PTSD symptoms were less sensitive and responsive, and more avoidant, overprotective, hostile and controlling when interacting with their children, in comparison to those without PTSD symptoms. While some of the studies within the review included parents experiencing PTSD following childbirth, research that has specifically explored the impact of birth-related PTS on parent-child relationship quality remains relatively scarce.

The nature of the interplay between the effect of a traumatic birth and associated PTS and their impact on the parent-child relationship can be conceptualised in several ways. Firstly, the overwhelming experience of PTS symptoms could lead to parents being less able to attend to and soothe their infant's distress (Erikson et al., 2019), which could influence the attachment relationship. However, as previously noted, PTSD symptoms form different clusters, which may differentially influence the parent-child relationship. For instance, the 'avoidance' cluster relates to individuals avoiding reminders of the traumatic event, which can include decreased affective responsiveness (Lyons-Ruth & Block, 1996). In the case of childbirth-related PTSD, parents may associate the infant with the traumatic birth experience, resulting in avoidance behaviours towards their baby. This could be particularly detrimental for the parent's availability and responsiveness that is required to bond.

Alternatively, increased levels of arousal could result in hypervigilant behaviours reflected by overprotective or intrusive parenting, a parenting style which has also been found in the literature to increase children's risk of greater internalizing difficulties (McShane & Hastings, 2009). Early literature into birth-related PTSD suggests that parents may even 'resent' the infant for 'causing' the trauma (Allen, 1998). Such feelings could potentially result in more hostile

parenting practices (Christie et al., 2019). Despite this, parents may also want to engage in behaviours to "prove themselves" and "make amends to the infant" (p.233) following a traumatic childbirth (Beck & Watson, 2008). Taken together, parental birth-related PTSD may influence the parent-child relationship in a number of ways, involving disrupting the parent's ability to provide warm, sensitive and responsive caregiving.

The existing literature offers some support towards these hypotheses, and it appears that bonding styles of parents experiencing birth-related PTS vary between an overprotective and anxious style, or an avoidant and rejecting style (Nicholls & Ayers, 2007). For example, overprotective parenting has been identified as a common response in parents of high-risk populations. Day et al. (2018) found that adults born at extremely low birthweight retrospectively reported greater overprotective parenting practices compared to matched controls. Adama et al. (2016) conducted a qualitative synthesis of research into the experiences of parents of pre-term infants. Here, overprotection was identified as a key theme across the studies, which was reported by parents to be a way in which to shield their infant from further harm.

Fenech and Thomson (2014) conducted a qualitative synthesis of research into women who experienced a traumatic childbirth, but who were not considered a high-risk population (e.g., pre-term or NICU admissions). In their study, differential parenting behaviours were adopted as a response to the traumatic birth. While some mothers noted a sense of needing to be overprotective of their child in order to "make up" for the difficult birth experience, other women reported feeling "totally detached" from their child. Nonetheless, a systematic review of quantitative research examining the impact of birth-related PTS on child outcomes reports inconclusive findings in relation to the mother-infant relationship (Cook et al., 2018).

To summarise, existing data suggest that poor postpartum mental health in mothers negatively affects the quality of their responsiveness and availability to their infant's needs. However, current research has predominantly investigated this in the context of postnatal mental health difficulties such as depression. Childbirth-related PTSD is a relatively recent area of research (Horesh et al., 2021). Research which has explored these outcomes in the context of birth-related PTS or PTSD has primarily focused on the subjective experiences of parents (see Fenech & Thomson, 2014, for a review). Additionally, the impact of PTS in women's partners remains at a preliminary stage within the literature. Previous systematic reviews have focused on the impact of maternal PTSD on child outcomes in the period from the beginning of pregnancy to one year following childbirth (Cook et al., 2018) or parental PTSD more broadly (Christie et al., 2019). Therefore, this review aimed to systematically review and summarise the current evidence for an association between parental childbirth-related PTS and the indicators of the quality of the

parent-child relationship during infancy (i.e., birth until two years). Given the relatively limited literature in this area, a range of measures relating to the parent-child relationship were included: the parent's behaviour towards the child (e.g., warmth, hostility, availability, overprotection); the child's behaviour towards the parent; attachment and bonding.

2.2 Methods

2.2.1 Inclusion and exclusion criteria

The articles from the search were included if they: a) were published in an academic journal b) available in English c) explored a direct relationship between quantitative measures of the parent-child relationship and birth-related PTS and d) variables were assessed during infancy (from birth up to and including two years old). Papers were excluded from this review if they: a) measured post-traumatic stress not related to childbirth b) measured post-traumatic stress combined with other mental health measures (e.g., depression, anxiety), and where it was not possible to separate the effects of PTS.

2.2.2 Search strategy

The protocol is registered with the International Prospective Register of Systematic Reviews database (Registration No. CRD42021277713).

The papers in this review were identified from a systematic search of the literature using the following online databases: PsycInfo via EBSCO, CINAHL via EBSCO, Medline and Web of Science. See Figure 1 for the PRISMA diagram (Moher et al., 2009) representing the different phases of the article-selection process. The search terms were generated from a scoping search of papers within this topic area and related to childbirth-related post-traumatic stress (childbirth OR labor OR labour OR delivery OR birth OR post-partum OR "post partum" OR postpartum AND PTSD OR "post traumatic stress disorder" OR "posttraumatic stress*" "post-traumatic stress*" OR "post traumatic stress*" OR "traumatic stress" OR trauma*) and parent-child relationships ("mother-child* relationship" OR "father-child* relationship" OR parenting OR "parent-child relationship" OR "parent-child bond" OR "parent-infant relationship" OR "mother-infant relationship" OR "father-infant relationship" OR attach* OR bond*). Searches were conducted for the entire time periods for which the databases are available up until August 2021.

The search was conducted in August 2021. The search strategy identified a total of 2413 studies. After duplicates were removed, titles and abstracts of 1,398 studies were screened based on predetermined eligibility criteria. The titles and/or abstracts of each article were reviewed by

the main review author and another review author based on inclusion and exclusion criteria. Disagreements related to inclusion or exclusion were resolved by discussion and consensus between the two authors was reached. 1364 were excluded following the title and abstract screening, and a further 10 were excluded during the full-text eligibility search, due to failing to meet the inclusion criteria. A total of 24 papers were included for this review. See Appendix A for the data extraction table of the 24 included papers.

2.2.3 Quality assessment

The quality of each article was evaluated using the 'Strengthening the reporting of observational studies in epidemiology' (STROBE) checklists, which is a validated quality appraisal tool widely used in observational research (Von Elm et al., 2014) and covers items that should be addressed in observational studies. As no randomised controlled trials met inclusion criteria, Version 4 of the STROBE checklist for cohort, case-control, and cross-sectional studies (combined) was used (see Appendix B). Limitations of the papers that were identified through the quality assessment process were included in the data extraction table (Appendix A). The main review author and another author independently assessed each criterion. Discrepancies were resolved through discussion. Overview of the results and additional information relating to the checklists are found in Appendix C.

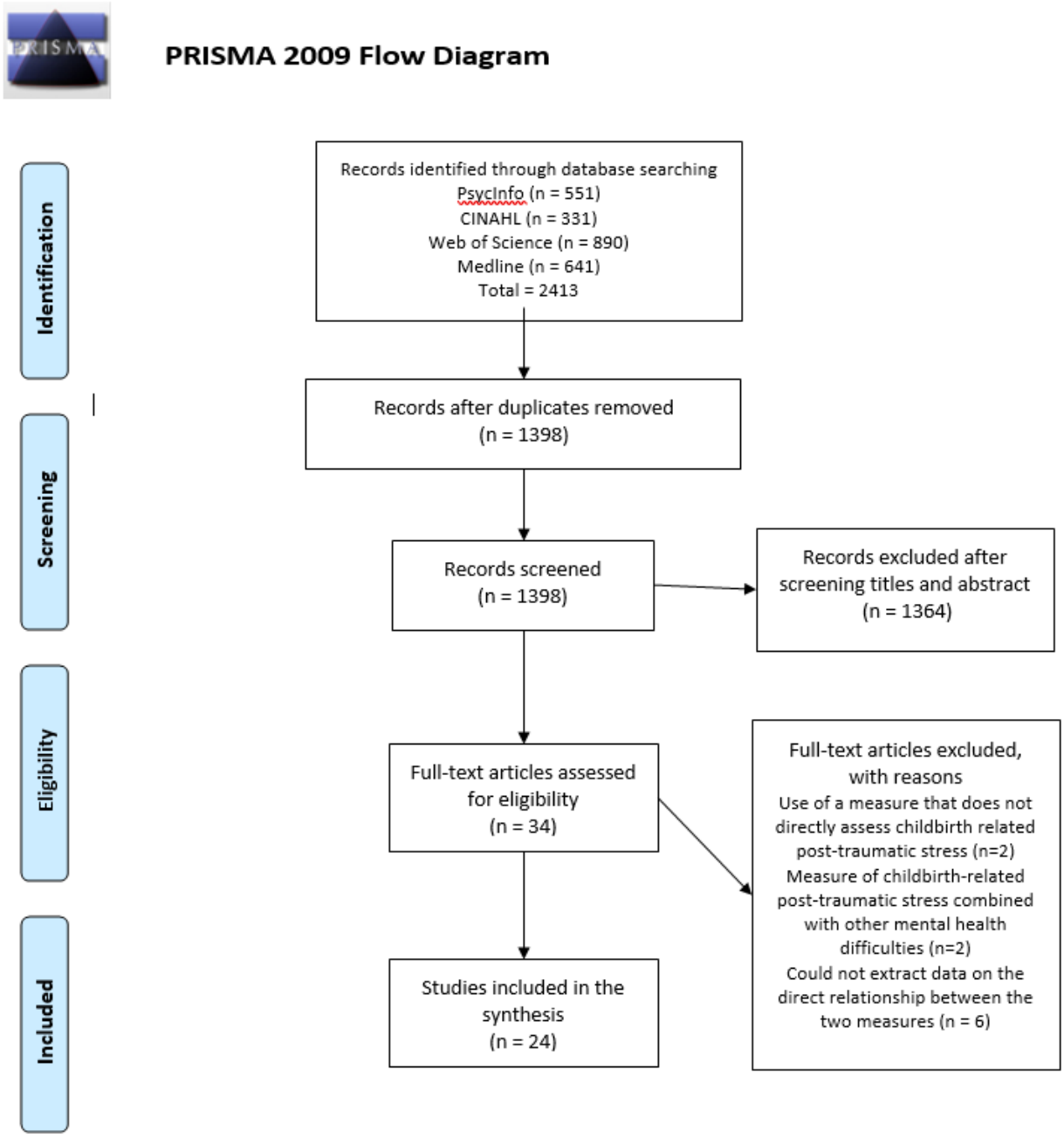


Figure 1 *PRISMA Flow Diagram (Moher, Liberati, Tetzlaff, & Altman, 2009) of the Systematic Search Process*

2.3 Results

2.3.1 Participant and study characteristics

The details pertaining to the sample design, measures and findings of the 24 studies identified in the current review can be found in Appendix A. Across studies, a total of 8892 parents (8566 mothers, 326 partners), with a range of 19 to 2802 participants per study were included. The studies were conducted across Europe (n = 16), Asia (n = 4), America (n = 2) and North America (n = 2). Five of the studies collected data from partners as well as mothers (Ayers et al., 2008; Ertan et al., 2021; Ionio et al., 2017; Parfitt & Ayers, 2009; Stuijfzand et al., 2020). Seven studies investigated outcomes within high-risk groups, including pre-term samples (Forcada-Geux et al., 2011; Ionio et al., 2017; Muller-Nix et al., 2004; Petit et al., 2016; Suttora et al., 2021), low birthweight samples (Feeley et al., 2011) and babies delivered during COVID-19 (Mayopoulos et al., 2021). The remaining studies (n=17) collected data from general population samples, recruited through hospital populations (e.g., maternity wards, parents attending prenatal courses or antenatal appointments) and online samples (e.g., social media, postpartum websites). Some of these studies employed strict inclusion and exclusion criteria to ensure that samples were 'low-risk', such as parents with no previous psychological diagnosis, low-risk pregnancies and/or births, and healthy infants (Camicasa et al., 2017; Davies et al., 2008; Ionio & Di Blasio, 2014; McDonald et al., 2011; Ponti et al., 2020; Smorti et al., 2021; Suetsugu et al., 2020).

2.3.2 Research design

The included studies used self-report questionnaires (n = 23) and interviews (n = 1) to assess birth-related PTS. A range of approaches were used to investigate parent-child relationships. Eight of the studies used coded-observational methods, 17 of the studies adopted parental self-report measures and one included an interview measure. 11 studies were cross-sectional and 13 were longitudinal. Of the seven studies that investigated high-risk groups, five utilised a case-control design.

2.3.3 Parental perceptions of the parent-child bond

18 of the studies assessed parent-child relationship quality through parental self-reported perceptions of the bond with their baby; 17 of these were carried out through self-report questionnaires, and one through an interview. The most commonly employed measure of the parent-child bond was the Postpartum Bonding Questionnaire (PBQ; Brockington et al., 2006),

which was used in seven of the studies. Other measures included the mother-to-infant bonding scale (MIBS; Taylor et al., 2006) (Ertan et al., 2021; Mayopoulos et al., 2021; Stuijfzand et al., 2020), maternal attachment inventory (MAI; Müller, 1994) (Dekel et al., 2019; Mayopoulos et al., 2021), maternal postnatal attachment scale (MPAS; Condon & Corkindale, 1998) (Davies et al., 2008; Ponti et al., 2020; Smorti et al., 2021), the 'parent-child dysfunctional interaction' (PCDI) subscale of the parenting stress index short form (Abidin, 1995) (Camicasa et al., 2017; McDonald et al., 2011), an adapted version of the Bethlehem Mother-Infant Interaction Scale (Pearce & Ayers, 2005) (Ayers et al., 2008) and the Working Model of the Child Interview (Zeanah & Beniot, 1995) (Forcada-Geux et al., 2011). Two papers included measures of father's perceived bond with their baby (Ayers et al., 2008; Stuijfzan et al., 2020) and in one paper partners reported their perceptions of the mother-to-infant bond.

2.3.3.1 Mothers

The papers assessing the perceived mother-infant bond offer some support that birth-related PTS has the potential to influence this bond, however the findings are extremely mixed. Firstly, some of the findings suggest that general PTS may be more predictive of parent-child bonding than birth-related symptoms specifically. Within both cross-sectional (Handelzalts et al., 2019) and longitudinal (Handelzalts et al., 2019; Nacik Rados et al., 2020) designs, correlational analyses found stronger associations between general PTS symptoms than with birth-related PTS symptoms. Nonetheless, correlations between birth-related PTS and the PBQ were positive and significant in Handelzalts et al. (2021) (r=.36, p<.01) and Nacik Rados et al. (2020) (r=.30, p<.01 for infants aged between one and six months, r=.29, p<.01 for infants aged between seven and 12 months). Still, mediational analyses in Nacik Rados et al. (2020) demonstrated that while greater general PTS symptoms had a direct and indirect (via depressive symptoms) on poorer bonding, birth-related PTS symptoms did not have a direct, nor indirect effect on bonding. In Handelzalts et al. (2019) cross-sectional study, only increased scores on general PTS symptoms were significantly associated with lower bonding scores. Such findings suggest that it may be more important to consider parental PTS symptoms more broadly than those specific to the childbirth.

Many of the papers highlight the reduced predictive value of birth-related PTS on parent-child bonding once other variables are considered. For instance, two longitudinal studies identified that any initial associations between birth-related PTS and bonding were no longer significant once maternal psychological distress postpartum (here, conceptualised as anxiety and depression) was accounted for (McDonald et al., 2011; Stuijfzan et al., 2020). Mediation models exploring the PTS symptom clusters (intrusion, avoidance, hyperarousal) in Stuijfzan et al. (2020) demonstrated that both the intrusion and hyperarousal subscales were indirectly related to

poorer bonding via psychological distress; no direct relationship between any of the PTS clusters and bonding was found. In McDonald et al. (2011), prior to the inclusion of maternal distress in the model, only reported PTS at three months postpartum (and not six weeks postpartum) was significantly positively correlated with parental perceived difficulties interacting with their child in McDonald et al. (2011) at two years postpartum.

In a cross-sectional study, Davies et al. (2008) categorised mothers' scores of birth-related PTS symptoms at six weeks postpartum, comparing 'non-symptomatic', 'partially symptomatic' or 'fully symptomatic'. Group comparisons highlighted that, compared to non-symptomatic mothers, those who were categorised as partially or fully symptomatic reported a significantly lower quality of attachment towards their baby. Significant correlations were found between many of the PTS clusters (re-experiencing, avoidance, increased arousal) and total PTS score with the quality of parental attachment to their baby. Nonetheless, once PPD was considered within the model, there was no significant effect of birth-related PTS on the parent-child bond.

Two other longitudinal studies found additional variables to impact the predictive value of birth-related PTS on the mother-infant bond. This included parental insecure attachment styles and general PTS symptoms in Handelzalts et al. (2021), alongside PPD. Within Suetsugu et al.'s (2020) longitudinal study, while a significant and positive correlation was found between concurrently reported birth-related PTS and the total PBQ score at four months postpartum, PTS at one month was not predictive of bonding difficulties at four months. Instead, later bonding difficulties were predicted by a maternal dismissive attachment pattern and family functioning. Finally, Mayopoulos et al. (2021) found that mother's acute stress response at birth was significantly associated with both bonding difficulties and birth-related PTS symptoms. However, acute stress response was accounted for within the analysis, and birth-related PTS was not significantly related to bonding difficulties; the direct relationship between birth-related PTS and bonding before accounting for acute stress was not reported.

In a large cross-sectional study, Ertan et al. (2021) found greater PTS symptoms to be significantly related to greater difficulties within the mother-infant bond and this held true for general and birth-related symptoms. However, it must be acknowledged that Ertan et al.'s (2021) analysis did not control for any possible confounders. Two cross-sectional studies, which also established significant associations between increased birth-related PTS and a poorer perceived mother-infant bond, failed to consider any indicators of postpartum mental health within their analysis (Hairston et al., 2018; Smorti et al., 2021). As highlighted above, such factors may impact the relationship between PTS and bonding, and so the conclusions that can be drawn from these papers is limited.

Regardless, other papers have found significant positive associations between birth-related PTS and parent-child bonding problems, once possible confounders have been considered. In Dekel et al.'s (2019) cross-sectional study, self-reported maternal attachment quality was significantly lower in mothers experiencing birth-related PTSD than mothers experiencing general PTSD, or no PTSD. Importantly, even after possible confounding pre-childbirth variables (e.g., mental health and trauma history) and childbirth variables (e.g., complications in childbirth, acute stress at birth) were adjusted for, PTSD related to childbirth, but not general PTSD, was significantly negatively associated with the mother-to-child emotional attachment.

Two of the papers identified a significant relationship between greater birth-related PTS and poorer bonding after PPD was considered. This was apparent when this relationship was considered longitudinally in Ponti et al. 2020, whereby birth-related PTS was directly related to the mother-infant bond, and indirectly through PPD. Additionally, Parfitt & Ayers' (2009) cross-sectional study found a specific influence of birth-related PTS, though the effect size was small (d=.20).

In a recent large longitudinal study, Kjerulff et al. (2021) identified that, compared to women who reported no PTS symptoms, women who reported one or more birth-related PTS symptom(s) at one month postpartum were more likely to report poorer bonding with their infant at one, six- and 12-months postpartum. Furthermore, this association remained after several possible confounding variables were controlled for, including maternal characteristics, social support, and PPD. Nonetheless, the findings in Kjerulff et al. (2021) should be interpreted with caution, given the brief nature of the measure used to assess PTS. Additionally, the dichotomous categorisation of PTS symptoms within the analysis in Kjerulff et al. (2021) offers limited explanation as to whether a greater PTS symptoms results in poorer mother-infant bonding.

Forcada-Geux et al. (2011) used semi-structured interviews to measure maternal attachment representations of their child for mothers of pre-term and full-term infants. Mothers were grouped into three categories: full-term, low-stress pre-term and high-stress pre-term. Interview responses were categorised as balanced, disengaged, or distorted. Here, compared to mothers of full-term infants, mothers of pre-term infants (irrespective of low- or high-stress classification) showed significantly less balanced representations of their infants; there was no difference between the two pre-term groups. Forcada-Geux et al.'s (2011) findings suggest that other factors within this population (i.e., related to a pre-term birth) are likely more important in the development of the parent-child relationship than distress related to the birth.

Contrary to findings noted above, both longitudinal and cross-sectional research within this area of literature have also found a lack of an association between birth-related PTS and the

parent-child relationship. Camicasa et al. (2017) found no relationship between reported birth-related PTS at 87 hours and three months postpartum and mother's perceptions of the emotional quality of the relationship with their child at 17 months postpartum. Only hyperarousal PTS symptoms at 17 months were significantly correlated with concurrently reported PCDI.

In Ayers et al. (2008), none of the birth-related PTS symptoms (intrusion and avoidance) were related to the mother-baby bond. However, it is highlighted that the measure used to assess the parent-baby bond had not yet been validated against other measures. Furthermore, Ayers et al. (2008) note that the scale focuses more on the behavioural aspects of the parent-infant relationship (e.g., reactivity of infant to parent, the amount the parent and baby like looking at each other), suggesting that the scale may more accurately measure 'care of the baby', rather than the 'emotional' bond.

Taken together, these findings across the papers suggest that other indicators of parental postpartum psychological distress (e.g., general PTS symptoms, PPD, acute stress response) and parental factors (e.g., maternal attachment styles, family functioning, sociodemographic variables) may potentially be stronger predictors of the parent-child relationship than distress specific to the birth experience.

2.3.3.2 Fathers

The relationship between partner's PTS and parent-child bonding is even less clear than that for mothers. Ertan et al. (2021) identified a significant positive correlation between partner's total PTS symptoms and their perception of mother-infant bonding difficulties, though this was not apparent for specific birth-related symptoms. However, it is not stated whether partners were present at the birth. Stuijfzan et al. (2020) explored the longitudinal relationship between fathers' childbirth-related PTS and mothers' self-reported bonding with their child; greater paternal PTS symptoms were significantly related to greater mother-infant bonding difficulties.

Neither Stuijfzan et al. (2020) nor Ertan et al. (2021) identified a significant relationship between fathers' birth-related PTS symptoms and father-infant bonding. However, the limited validity of the measure assessing bonding in Ertan et al. (2021) means that conclusions that can be drawn from this study are tentative. As this relatively novel area of literature, it is not yet clear as to whether the experience of childbirth-related PTS in fathers may influence mother-infant or father-infant bonding.

2.3.4 Maternal behaviours

2.3.4.1 Positive affect during interaction

Two studies assessed mothers' positive affect during interaction, through self-report and observational measures (Davies et al., 2008; Petit et al. 2016). Davies et al. (2008) identified that 'fully symptomatic' mothers (i.e., mothers who reported symptoms fulfilling a criteria of PTSD) reported significantly less pleasure in interaction with their infant than partially symptomatic or non-symptomatic mothers. Furthermore, significant correlations were found between certain PTS clusters (avoidance, increased arousal, total score) and less pleasure in interaction, though none remained significant once PPD was considered in the analysis.

Petit et al. (2016) analysed pre-term mother-infant interactions using the Paediatric Infant Parent Exam (PIPE; Fiese et al., 2001), which scores the degree of interactional reciprocity and positive affect observed; a lower score indicates a more favourable interaction. Here, PTS was assessed at three time points: before hospital discharge, six months postpartum and 12 months postpartum. The only significant association found with the PIPE score was self-reported PTS at six months, which demonstrated a moderate positive correlation with PIPE scores (r=.034). Mothers' anxious and depressive symptoms also demonstrated some correlations with PIPE scores, but no analysis was undertaken to investigate the impact of PTS on PIPE scores once these measures of maternal mental health were taken into consideration.

2.3.4.2 Maternal sensitivity

Four papers assessed maternal sensitivity through observational measures (Feeley et al., 2011; Forcada-Geux et al., 2011; Ionio et al., 2017; Muller-Nix et al., 2004). All four papers included mothers considered to be in 'high-risk' groups (i.e., mothers of pre-term and very low birthweight infants). In Ionio et al. (2017), fathers completed measures of PTS but were not recorded interacting with their child.

Feeley et al. (2011) analysed maternal sensitivity within mother-infant interactions of very low birthweight infants. Correlational analysis indicated that mothers who reported greater PTS symptoms were observed to be significantly less sensitive when playing with their child. Ionio et al. (2017) recorded mother-infant interactions for mothers of pre-term and full-term infants at three months of age. Fathers completed measures of PTS but were not recorded interacting with their child. Correlations were performed for the pre-term sample only; within the pre-term sample, neither maternal or paternal birth-related PTS was associated with mothers' sensitivity.

In a longitudinal study, Muller-Nix et al. (2004) recorded mother-infant interactions in mothers of pre-term and full-term infants at both six- and 18-months postpartum, whereby mothers were grouped into three categories: full-term, low-stress pre-term and high-stress pre-term. At six months, maternal sensitivity was found to be significantly lower in high-stress mothers than full-term mothers. Furthermore, partial correlations highlighted a stronger influence of PTS on maternal sensitivity than the infant's severity of risk (i.e., perinatal difficulties such as gestational age and weight). Nonetheless, at 18 months, no significant differences in observed maternal sensitivity were identified. It is important to note that birth-related PTS was only reported retrospectively 18 months postpartum, and so differences in maternal behaviours at six months may not accurately reflect differences in birth-related PTS.

Forcada-Geux et al. (2011) identified that the percentage of sensitive mother and cooperative infant dyads was significantly lower in pre-term dyads (regardless of PTS status) than full-term dyads, once again highlighting that other factors within this population may influence the parent-child relationship.

2.3.4.3 Maternal mind-mindedness

Two of the papers analysed observed maternal mind-mindedness (Camicasa et al., 2017; Suttora et al., 2021). Mind-mindedness refers to the mother's ability to interpret the infant's cues accurately (Meins et al., 2001) and is considered a prerequisite to maternal sensitivity (Meins, 1999).

Camicasa et al. (2017) demonstrated that birth-related PTS symptoms (both at 87 hours and 3 months postpartum) were not significantly associated with observed maternal mind-mindedness at 17 months. However, hyperarousal symptoms at 17 months were significantly negatively associated with maternal mind-mindedness. Suttora et al. (2021) examined mother-infant dyads for pre-term and full-term infants at six months of age. Here, no significant differences were found between the two groups on PTS or mind-mindedness. Correlational analysis was computed for the entire sample only. No significant correlations were established between PTS symptoms and mothers' use of mind-related comments, though it is worth noting that PTS clusters, which may have yielded further results as seen in Camicasa et al. (2017), were not investigated in this study.

2.3.4.4 Maternal hostility and rejection

Three papers assessed maternal hostility/rejection through self-report measures (Davies et al., 2008; Hairston et al., 2018; Suetsugu et al., 2020) and one through observational measures (Feeley et al., 2011).

When this relationship was assessed using cross-sectional designs, there appears to be a significant relationship. Davies et al. (2008) identified that, at 6 weeks postpartum, partially or fully symptomatic mothers reported a significantly greater infant-directed hostility than asymptomatic mothers. Furthermore, increased scores on the PTS symptom clusters (re-experiencing, avoidance, increased arousal) and total PTS score were associated with greater infant-directed hostility. In a cross-sectional questionnaire at four to 12 weeks postpartum, Hairston et al. (2018) identified that higher levels of birth-related PTS were significantly correlated with higher scores on the 'anger and rejection' subscale of the PBQ.

However, more rigorous study designs which have assessed maternal hostility and rejection (i.e., longitudinal designs or observational measures) do not indicate such a relationship. Suetsugu et al. (2020) measured birth-related PTS and parent-child relationship measures at both one and four months postpartum. Associations between PTS and the subscale of 'anger and rejection', and 'risk of abuse' on the PBQ were not significant, concurrently nor longitudinally. Feeley et al. (2011), found at 6 months' postpartum, there was no significant relationship between self-reported PTS and observed maternal hostility for mothers of low birthweight infants.

Nonetheless, Ionio & Di Blasio (2014) longitudinally investigated this relationship through scrutinising maternal behaviours towards their child at three months postpartum, using the Still Face Paradigm (Tronick et al., 1978). Regression analyses highlighted that, during the play phase (i.e., where the mother and child were engaged in free play), PTS symptoms at two months postpartum were positively associated with mothers' describing their child's status in a negative way. However, the small sample size (19 women attended all phases of the study), and the lack of inclusion of possible confounders, limits the generalisability of these results.

2.3.4.5 Maternal intrusiveness and control

Four papers included analysis of indices of maternal intrusiveness or control via coded observations (Forcada-Geux et al., 2011; Ionio et al., 2017; Ionio & Di Blasio, 2014; Muller-Nix et al., 2004).

Although mothers of pre-term infants in Ionio et al. (2017) were found to be significantly more intrusive during interactions with their child, this cannot be attributed to PTS, as no differences were found between the groups on any of the PTS subscales. Within the pre-term sample, none of the PTS subscales were associated with mothers' behaviour with their infant. Paternal birth-related PTS was not associated with maternal intrusiveness.

Conversely, Muller-Nix et al. (2004) identified maternal control to be significantly higher in high-stress pre-term mothers than full-term and low-stress pre-term mothers at six, but not 18

months postpartum. In line with this, Forcada-Geux et al. (2011) reported that significantly more 'control-compliant' dyads (i.e., a controlling mother and compliant infant) were observed in the high-stress pre-term group than in the full-term group. However, as differences in PTS scores are not reported in Forcada-Geux et al. (2011), it is not possible to draw comparisons between the two groups.

Within the play phase of the Still Face Paradigm in Ionio & Di Blasio (2014), higher levels of PTS were associated with the mother singing, making sounds, and touching their child. During the reunion phase, mothers with greater PTS symptoms were more likely to make sounds to catch their child's attention, and less likely to keep a 'typical' interaction distance from their child. The authors conclude that women experiencing PTS may adopt more intrusive behaviours when interacting with their child, though it could be argued that some of the above behaviours are not necessarily less optimal. Furthermore, the limitations of this study as previously noted limit the generalisability of the results.

2.3.4.6 Maternal anxiety

Two papers investigated the 'infant-focused anxiety' subscale of the PBQ (Hairston et al., 2018; Suetsugu et al., 2020). Hairston et al.'s (2018) cross-sectional study identified birth-related PTS to be significantly and positively correlated with mother's infant-focused anxiety (e.g., "I am afraid of my baby"), when reported between four and 12 weeks postpartum. Similarly, Suetsugu et al.'s (2020) longitudinal study highlighted higher birth-related PTS to be significantly related to greater maternal infant-focused anxiety at one month postpartum. However, no such association was apparent at four months postpartum. Perhaps the experience of PTS related to childbirth impacts mother's feelings of anxiety in the early weeks following birth but does not persist as the child develops. Nonetheless, the limited number of papers here mean that purely tentative hypotheses can be made.

2.3.4.7 Maternal unavailability

Four studies investigated indicators of maternal unavailability during observed interactions with their child (Feeley et al., 2011; Ionio et al., 2017; Ionio & Di Blasio, 2014; Muller-Nix et al., 2004). One study included fathers' reports of birth-related PTS (Ionio et al., 2017).

Two papers investigated this within pre-term populations. Ionio et al. (2017) found mothers of pre-term infants to be significantly more distant during interactions with their child than mothers of full-term children, though as previously noted, this cannot be attributed to PTS. Birth-related PTS subscales were not associated with mothers' behaviour with their infant. Interestingly, fathers' reported avoidance PTS symptoms were significantly related to mothers'

remoteness (e.g., remote, silent) and signs of depression (e.g., sad, low energy) during mother-infant interactions. On the contrary, in Muller Nix et al. (2004), no differences were found between pre-term and full-term mothers on maternal unresponsiveness. Furthermore, PTS symptoms were not related to maternal unresponsiveness at six or 18 months postpartum.

Ionio & Di Blasio (2014) identified that PTS at two months postpartum predicted indicators of maternal unavailability, characterised by looking away from their children's face during both the play and reunion phase of the Still Face Paradigm. Furthermore, correlations in Feeley et al. (2011) indicated that, at six months corrected-age, mothers of low birthweight infants who reported greater PTS symptoms were significantly less effective at structuring interactions with their child. Regardless, both papers included extremely small sample sizes.

2.3.5 Child behaviours

Two of the papers utilised the mothers' object relation scale (MORS; Oates & Gervai, 2003) (Davies et al., 2008; Mcdonald et al., 2011), which assesses how invasive and warm a child is perceived to be by their mother. Three papers observed and analysed children's behaviour towards their parent within parent-child interactions (Ionio et al., 2017; Ionio & Di Blasio, 2014; Muller Nix et al., 2004).

In Davies et al. (2008), higher levels of total birth-related PTS, as well as higher scores on the PTS clusters (re-experiencing, avoidance, increased arousal) were associated with mothers reporting infants to be less warm and more invasive, though correlations were relatively weak (r=-.14 to r=-.22 for warmth, r= .21 to r=.37 for invasion). Group comparisons between mothers based on their reported PTS symptoms also highlighted fully or partially symptomatic mothers perceived their infants to be significantly less warm and more invasive than non-symptomatic mothers. After PPD was considered, only a significant effect remained for perceived warmth.

McDonald et al. (2011) adopted a longitudinal design, whereby participants completed two measures of birth-related PTS (the PTSD questionnaire and the Impact of Events Scale) at three time points: six weeks, three months and two years postpartum. Parent-child relationship outcomes (including the MORS) were collected at two years postpartum. At two years postpartum, small but significant positive correlations were found between concurrent reports of both PTS measures and the invasion subscale of the MORS. Examining the longitudinal association between PTS and the MORS at two years postpartum, birth-related PTS symptoms assessed at three months were not significantly associated with either of the MORS subscales. Only the PTSD questionnaire reported at six weeks demonstrated a weak, but significant, correlation with the invasion subscale of the MORS at two years postpartum. However, once maternal depression and

anxiety at two years postpartum were adjusted for, earlier PTS symptoms (at six weeks and three months postpartum) did not significantly predict outcomes on either of the MORS subscales.

Ionio et al. (2017) found that, for mothers of pre-term infants, greater birth-related 'intrusive' PTS symptoms in the first two weeks following birth were significantly correlated with greater infant distress at three months postpartum. Father's PTS symptoms were not significantly associated with any of the infant behaviours. Similarly, Ionio & Di Blasio (2014) identified that, at three months postpartum, during the play phase of the Still Face Paradigm, infants of mothers with greater PTS symptoms were more likely to display behaviours indicating their distress, including crying and disorganised behaviours. During the still episode, greater PTS was associated with infants looking away and withdrawing from their mother.

Muller Nix et al (2004) assessed infants at two time points: six and 18 months postpartum. Here, there were no significant differences in infants' interactional behaviour in the three parental groups (full-term/low stress pre-term/high stress pre-term) at six months. Interestingly, at 18 months, observed passivity was higher for infants within the low stress group, compared to high stress and full-term dyads.

2.4 Discussion

To our knowledge, this is the first review to systematically summarise the evidence for the association between birth-related PTS symptoms and parent-child relationship quality. There is, albeit limited, some evidence that suggests birth-related PTS may be related to indicators of poorer parent-child relationship quality, including greater bonding difficulties, less positive affect, less optimal maternal behaviours towards the child (e.g., lower sensitivity, greater, hostility, intrusiveness, anxiety and unavailability) and child behaviours towards the parent (e.g., greater invasiveness and distress, less warmth). However, there is substantial variability and inconsistency in the findings across these constructs. It is therefore important to further explore why such differential results were found across the papers.

A key theme across the literature is that the relationship between parental experiences of PTS related to childbirth and their relationship with their child is influenced by a complex interplay of pre- and post-childbirth factors. In particular, many of the papers that assessed PPD found that, once this was included, birth-related PTS was not an important factor related to the parent-child relationship. Nonetheless, many of the reviewed papers found close associations between birth-related PTS, PPD and parent-child relationship quality. There is a high comorbidity between childbirth-related PTSD and PPD (White et al., 2006), and both PTSD and PPD are reported to present almost identically in the early postpartum period (Dekel et al., 2020). Given the robust

literature demonstrating a negative influence of PPD on parent-child relationships, if PTS following childbirth may be involved in the onset of PPD, this will be important for clinicians to be aware of, and to ensure that appropriate intervention is provided.

Another potential explanation for the mixed findings in the review could relate to the variability in methodology used across the reviewed papers. For instance, sample sizes varied between 19 (Ionio & Di Blasio, 2014) and 2802 (Kjerulff et al., 2021). Only a small number of the papers (n = 4) stated whether they were sufficiently powered to identify a significant effect. It is possible that the studies with smaller sample sizes may not have been sufficiently powered to identify a significant relationship between birth-related PTS and parent-child relationship quality.

There is some indication that the relationship between birth-related PTS and parent-child relationship quality may be more prominent in the first few weeks following childbirth. For instance, some data indicates that associations between birth-related PTS and the mother's behaviour towards the child (e.g., greater infant-related anxiety, less sensitivity) was weaker when the mother-child relationship is assessed later (i.e., months after birth). Nonetheless, this trend in findings was not consistent across the reviewed papers, and indeed the opposite was found in other studies. It is therefore not possible at this point to make firm conclusions on whether associations between birth-related PTS and the parent-child relationship varies according to the age of the child.

When considering the time point at which birth-related PTS was assessed, findings continue to be contradictory. For instance, Petit et al. (2016) found no significant correlations between concurrently reported PTS and indicators parent-child relationship quality at 12 months postpartum, whereas McDonald et al. (2011) found significant correlations when measures were concurrently measured at two years postpartum. Interestingly, Camicasa et al. (2017) found stronger associations between certain PTS symptoms and parent-child relationship quality when this relationship was assessed concurrently at 17 months postpartum, than between earlier reported PTS (i.e., in the first days and earlier months) and later parent-child relationship quality. Thus, although the evidence is not conclusive at present, it does suggest that further attention needs to be paid in the literature to parents who continue to experience birth-related PTS in the later months and years following birth.

Interestingly, no consistent patterns in the relationship between birth-related PTS and parent-child relationship quality were found relating to the type of measure used to assess the parent-child relationship (i.e., self-report vs observational). It has been argued that self-report questionnaires may be subject to bias. Firstly, parents may report socially desirable answers which present their behaviours towards their child, or their relationship with their child, in a more

positive light (Schwarz et al., 1985). Conversely, parents experiencing psychological distress may report a more negative perception of both parent and child behaviours, as seen in mothers with depression (Chi & Henshaw, 2002). However, within this review, mixed findings were established in studies which used self-report and/or observational measures. Additionally, the research design did not appear to influence patterns of findings across the reviewed papers. That is, evidence was inconsistent both in studies which used cross-sectional, and in studies which employed longitudinal designs.

It is also important to note that some of the papers (n = 5) measured PTS very shortly after the birth, and in some cases, before participants had left the hospital. Although the inclusion criteria for this review meant that participants did not need to fulfil the criteria for PTSD, it could be argued that measuring stress responses to birth in the days after the event may reflect an acute stress response rather than PTS (American Psychiatric Association, 1994). Nonetheless, three of these papers included measures of PTS at later time points, and the two that reported differences in PTS across the time points found symptoms to remain relatively stable over time (Camicasa et al., 2017; Ionio & Di Blasio, 2014).

2.4.1 Limitations

Methodological issues must be considered. The heterogeneity of the reviewed papers, including study design (cross-sectional, longitudinal, and case control), the type of measure used to assess parent-child relationship quality (observational and self-report), sample characteristics, and the time at which birth-related PTS and parent-child relationship quality were assessed, limits the comparisons available. Furthermore, many of the studies recruited participants from one hospital**,** which may limit how widely results can be generalised. Furthermore, nearly all studies relied on self-report questionnaires to assess birth-related PTS, and many to assess parent-child relationship quality, which may be subject to bias, as noted above. Finally, not all papers accounted for possible confounding variables (e.g., demographic variables, pre and postnatal parental mental health), consequently reducing the value of their results in enhancing understanding of whether a unique influence of parental birth-related PTS on parent-child relationships exists.

2.4.2 Conclusions and future research

In recent years, increased attention has been paid in the literature to birth-related PTS and parent-child relationships. While there is some evidence which suggests that parental birth-related PTS may have a negative impact on their relationship with their child during infancy, this

has not been consistently found across the literature, and some findings are contradictory. Given that birth-related PTS is a relatively novel area of literature, further research is necessary before substantial conclusions and recommendations can be made. Moreover, research assessing birth-related PTS in partners present at the birth is currently extremely limited; investigating how this may influence parent-child relationships is also warranted. Additionally, it will be important that future research examines specific PTS symptom clusters (e.g., intrusion, avoidance, increased arousal) with specific indicators of parent-child relationship quality (e.g., parental warmth, hostility), as this may offer better insight into how parental PTS may influence parent-child relationship quality, and whether certain PTS symptoms are more strongly related to the parent-child relationship.

Additionally, relatively few of the studies specifically focused on 'high-risk' birth groups, and those that did tended to be pre-term births. Further research into high-risk birth populations is warranted, given their increased risk of birth-related PTS (Yildiz et al., 2017). This will need to include infants who spend time in the Neonatal Intensive Care Unit (NICU), such as infants who experience neonatal hypoxic-ischemic encephalopathy. This will be important to aid understanding of whether birth-related PTS places high-risk birth populations at increased risk of difficulties within the parent-child relationship.

Finally, there appears to be a complex interplay of PTS with pre-childbirth variables, the childbirth experience and post-childbirth variables (Ayers, 2004). This includes comorbidity with other parental psychopathology such as postpartum depression and anxiety, which was found within the reviewed papers to impact upon the relationship between birth-related PTS and the parent-child relationship. While the assessment of risk factors was not within the scope of this review, factors such as parental education, parental distress during birth, pre-existing parental psychopathology, birth type, and infant birth complications, were all identified as possible confounders among the reviewed papers. Future research must consider these potential confounders to better understand the role that childbirth-related PTS plays within this, and its relative influence on the parent-child relationship. More detailed and rigorous exploration of whether, and how, parental birth-related PTS affects their relationship with their child, alongside factors that may buffer against any potential negative consequences, will be crucial to inform required support for parents. This will include developing a greater understanding of whether the parent-child relationship is impacted when parents continue to experience birth-related PTS at more distal time points (i.e., years after the birth).

2.4.3 Implications

Although this systematic review has not definitively evidenced a consistent relationship between birth-related PTS and parent-child relationships, there is some indication that increased parental birth-related PTS is related to poorer parent-child relationship quality. To contribute to the evidence base, EPs and health professionals are well placed to conduct further research and disseminate this to parents and educational staff. Alongside this, it may be particularly helpful to make others aware of theory and research which highlight the importance of early child-caregiver relationships for child development, such as Attachment theory (Bowlby, 1969).

Furthermore, it will be important that health professionals are aware of the possibility for childbirth-related PTS to have an impact on parent-child relationship quality, so that they can identify parents experiencing such distress and signpost to appropriate support.

Chapter 3 Maternal birth-related stress and school-age emotional and behavioural outcomes of children with hypoxic-ischemic encephalopathy

3.1 Introduction

3.1.1 Hypoxic-ischemic encephalopathy

Neonatal hypoxic-ischemic encephalopathy (HIE) is a consequence of inadequate blood flow and oxygen to an infant's brain, usually during labour and delivery (Kurinczuk et al., 2010). HIE affects approximately 2.96 per 1000 live births in England and Wales (Shipley et al., 2021) and poses a risk for infant mortality and poorer developmental outcomes, particularly in relation to children's cognitive and motor development (deVries & Jongmans, 2010; Perez et al, 2013; Pierrat et al., 2005). These serious complications at birth are often unexpected, with parents anticipating the birth of a healthy infant (Allen & Kelley, 2016; Heinghaus et al., 2013).

3.1.2 HIE and the birth experience

Medical advances have improved the survival rates of infants following HIE. Therapeutic hypothermia (TH) is recommended treatment for moderate to severe HIE (NICE, 2010) to reduce the risk of death or severe disability (Jacobs et al., 2013). TH involves immediate care in the neonatal intensive care unit (NICU) and cooling of the infant for 72 hours (Long & Brandon, 2007). During this time, parents are physically separated from their child, infants may be sedated, and physical manipulation of the infant (including contact with the parent) is limited (Long & Brandon, 2007). Furthermore, it is difficult to predict the outcome for the infant during the first days of TH and the subsequent neonatal period (Liu et al., 2020). As such, the birth experience and postnatal time can be particularly distressing for parents.

Qualitative research into parental experiences of infants who receive treatment in the NICU express feelings of guilt, powerlessness, sadness, hope and despair (Bäcke et al., 2021; Nassef et al., 2013; Wigert et al., 2006). More specifically to parents of children with HIE, the birth experience and ensuing postnatal period can be emotionally turbulent, particularly after anticipating the birth of a healthy child (Heringhaus et al.., 2013); parents note feelings of loss and

grief (Lemmon et al., 2017). Parents "face an unexpected, rapidly changing scenario involving separation, anxiety and need to have unexpected complex discussions about mortality and long-term morbidity" (Thyagarajan et al., 2018, p.2531). Even in the years after the birth of a child with HIE, it appears that parents continue to be impacted by the birth events, including encountering difficulties remembering the birth and expressing feelings of distress regarding visiting the hospital (Heringhaus et al., 2013). Given the above, it is plausible to suggest that mothers who give birth to children with HIE are at-risk of sustaining elevated levels of PTS associated with childbirth.

3.1.3 Emotional and behavioural development of children with HIE

Accumulating evidence suggests that HIE may result in poorer emotional and behavioural outcomes, even in those children who do not develop severe neuromotor impairment (e.g., cerebral palsy). In the literature, emotional and behavioural difficulties tend to be conceptualised as internalising problems (e.g., fearfulness, sad mood, social withdrawal, somatic complaints) or externalising problems (e.g., poor impulse control, noncompliance, aggression) (Achenbach & Edelbrock, 1978; Campbell, 1995).

Maladaptive emotional and behavioural development in childhood places children at greater risk of poorer psychological and social outcomes, with negative consequences persisting into adolescence and adulthood (Bevilacqua et al., 2018; Pihlakoski et al., 2006; Scott et al., 2001). For instance, evidence has shown internalising and externalising problems at the age of eight to respectively predict internalising disorders (e.g., anxiety disorders and depression) (Goodwin, Fergusson & Horwood, 2004) and criminality and substance abuse (Fergusson et al., 2005; Huesmann et al., 2003) in adulthood. Moreover, both have been found the be related to poor educational attainment (Riglin et al., 2014; Timmermans et al., 2009). Thus, it is crucial to understand populations who may be at risk for experiencing such difficulties, so that prevention and intervention can be established.

When considering the evidence regarding the impact of HIE on children's emotional and behavioural development, findings thus far are relatively limited and inconclusive. Lee-Kelland et al., (2020) identified from parental reports that, at school-age (i.e., six to eight years of age), compared to typically developing peers, children within the HIE group demonstrated significantly more emotional difficulties as reported by the Strengths and Difficulties Questionnaire (SDQ; Goodman, 2001). Van Schie et al. (2015) assessed the emotional and behavioural difficulties of children with HIE who had not received TH using the total score from the Child Behavior Checklist (CBCL; Achenbach & Rescorla, 2001). Within this study, CBCL scores for children with HIE were

comparable to general population samples. Nonetheless, the small sample sizes in both studies limit the generalisability of such findings. Reviews of the literature have found that children with HIE and without cerebral palsy are at greater risk of experiencing cognitive and behavioural difficulties (de Vries & Jongmans, 2010; Schregelmann et al., 2020). Yet, both reviews note that current evidence concerning school-age emotional and behavioural outcomes among HIE children is extremely scarce.

In a recent pilot study, Edmonds et al. (2022) identified that, in comparison to a typically developing control group, children with HIE who received TH were reported by parents to have greater emotional and behavioural problems on the SDQ. Although no significant differences were found between the groups on standard scores for internalising and externalising difficulties on the CBCL, a much larger proportion of scores fell into the 'borderline' or 'clinical' range for children in the HIE group. In addition, teachers completed the SDQ. Here, emotional, but not behavioural, difficulties, were significantly increased, suggesting that perhaps children's behaviour may be different across contexts.

3.1.4 Predictors of emotional and behavioural development

In order to promote adaptive emotional and behavioural development in childhood that is crucial for later outcomes, it is important to develop an understanding of the factors that may hinder such development. For children with a history of HIE and TH, there are various possible factors that may help to explain the increased risk of emotional and behavioural difficulties in this population. Ecological and developmental theories emphasise the influence of several contexts on children's development, including factors at the individual, family, and environmental level (Belsky, 1984; Bronfenbrenner, 1979). This includes several variables, such as child temperament (Caspi et al., 1995) and socioeconomic status (Dodge & Petit, 2003).

Nonetheless, reviews of the literature suggest that family factors involving parenting practices and parent-child relationship quality are amongst one of the most important and significant predictors of children's externalising difficulties (Loeber & Stouthamer-Loeber, 1986). Furthermore, Sameroff and Seifer (1985) suggest that both the environment and parents have the ability to moderate the effects of biological risk factors on child development; thus, parents can potentially buffer against poorer outcomes in children who are at increased risk. For instance, Forcada-Geux et al. (2006) identified more optimal maternal behaviours (e.g., greater maternal sensitivity) to be related to better behavioural outcomes in pre-term infants.

For children who experience neonatal HIE and subsequent TH, there may be biological and neurological factors that increase the risk of later developmental difficulties, such as possible

alterations to brain structure (Annink et al., 2021; Spencer et al., 2021; Thrarmapoopathy et al., 2020). Nonetheless, environmental factors that may further increase the risk of emotional and behavioural difficulties in children following HIE and TH must also be considered. One such factor that is important to examine in this population is possible increased levels of childbirth-related psychological distress in parents.

3.1.5 Postpartum maternal mental health and child outcomes

The association between maternal postnatal mental health difficulties and less optimal child outcomes have been well demonstrated within empirical research, including associations with poorer cognitive, social, emotional, and behavioural outcomes across childhood and adolescence (Halligan et al. 2007; Kingston & Tough, 2014; Murray et al., 2010; Murray et al., 2011). Moreover, even when considering the impact of paternal mental health, maternal psychological distress has been found to be an independent predictor of children's internalizing symptoms (Vänskä et al., 2017). Nonetheless, research in this area has predominantly been informed by the impact of maternal postnatal depression on children's development.

3.1.5.1 Maternal childbirth-related post-traumatic stress disorder (PTSD)

The influence of maternal post-traumatic stress disorder (PTSD) following childbirth on children's behavioural and emotional outcomes is of increasing interest. Prevalence rates of childbirth-related postpartum PTSD widely vary. Perinatal PTSD affects between 4% to 6% of women in general population samples (Yildiz et al., 2017). More specifically, childbirth-induced PTSD has been found to affect between 5% to 8% of women in the first one to three months after childbirth (Dekel et al., 2017). Although one of the key factors involved in the onset of PTSD following childbirth is one's perception of childbirth as a traumatic event (Dekel et al., 2017; Garthus-Niegel et al., 2013), there are certain variables which place parents at greater risk for experiencing such distress. This can include those who experience severe pregnancy or birth complications. In these high-risk samples, prevalence rates of perinatal PTSD increase to around 18.5% (Yildiz et al., 2017). However, these reported prevalence rates do not include parents who experience post-traumatic stress (PTS) symptoms that impact upon their functioning, but who do not fulfil the diagnostic criteria for PTSD (McKenzie-McHarg et al., 2015).

While it remains a more novel area of literature, longitudinal evidence suggests that parental childbirth-related PTS may impact upon child development. For instance, in a longitudinal study by Garthus-Niegel et al. (2017), it was identified that maternal PTS at eight weeks following childbirth was prospectively associated with poorer child socio-emotional development at two years of age, even when other indicators of maternal mental health and child

temperament were accounted for. Within Garthus-Niegel et al. (2017), stronger associations were found among boys and among children with a difficult temperament as reported by the mother. Enlow et al. (2011) also found that, even after controlling for possible confounding variables (depressive symptoms and infant exposure to traumatic events), maternal PTS symptoms at six months postpartum were predictive of poorer infant emotion regulation, internalising and externalising symptoms at 13 months of age. However, postpartum PTS was assessed more broadly rather than specifically to the childbirth in Enlow et al. (2011). Presently, the majority of the literature focuses on the impact of childbirth-related PTS on child outcomes in infancy; little data is available on outcomes in later childhood and adolescence. Additionally, little is known about maternal birth-related PTS and its relationship with children's emotional and behavioural outcomes in 'high-risk' birth populations, including HIE.

3.1.5.2 Maternal postpartum mental health and the mother-child relationship

A significant factor that is considered to help explain the possible trajectories from maternal postpartum mental health to child outcomes is the mother-child relationship. Belsky (1984) proposed that, while there are multiple determinants of parenting behaviours, the psychological resources of the parent are of utmost importance. Both theory and research have implicated mother-child relationship quality to be related to children's socio-emotional development. Attachment theory (Bowlby, 1969, 1982) emphasises the importance of the early mother-child relationship for child development. A secure attachment is formed through sensitive parenting; that is, the mother's ability to recognise, understand, and appropriately respond to the needs of their child (Ainsworth et al., 1978). Furthermore, attachment theory proposes that the early parent-child attachment relationship results in an internal working model (IWM). This refers to a set of expectations and beliefs that an individual maintains about themselves, others, and their relationships (Bowlby, 1973), and is believed to guide behaviour (Ainsworth, 1990).

The quality of the mother-infant attachment relationship can have profound implications for children's psychological development. Associations between the mother-child attachment relationship and emotional and behavioural development have been empirically supported by a large body of literature. Meta-analyses have identified insecure attachments to increase the risk of both internalising symptoms (Groh et al., 2012; Madigan et al., 2013) and externalising symptoms (Fearon et al., 2010) across childhood.

Despite the importance of sensitive and responsive parenting that is central to promoting a secure attachment, postpartum mental health difficulties can indeed reduce a mother's capacity to provide such care. For example, Stein et al.'s (2014) systematic review of the literature found

parental postpartum psychological distress to be related to the parent's reduced ability to respond to their child's cues in a sensitive and consistent manner.

Less research has specifically investigated the impact of maternal birth-related PTS on the mother-child relationship. There is indication that it has the potential negatively impact upon mother-child relationship quality, including reduced mother-to-infant emotional attachment, less sensitivity, and greater hostility towards the infant (Dekel et al., 2019; Feeley et al., 2011; Davies et al., 2008). However, other studies have failed to find such associations (Ayers et al., 2007; Feeley et al., 2011; Forcada-Geux et al., 2011) or found initial associations to no longer remain after accounting for possible confounding variables, such as maternal anxiety and depression (Stuijfzan et al., 2020). Inconsistencies between study methodologies and findings make it challenging to draw any firm conclusions at present (see Chapter 2).

Nonetheless, it is theoretically plausible that the experience of childbirth-related PTS could hinder the availability of the parent to attend and respond to their child (Erikson et al., 2019). Possible avoidance of reminders of the traumatic event could lead to avoidant behaviours towards their child (Lyons-Ruth & Block, 1996). Qualitative evidence suggests that some parents with birth-related PTS following a traumatic birth may adopt an avoidant and rejecting style, expressing a lack of desire to go near their child, or a delayed onset of an emotional attachment to their child, while still 'acting' out the mother role (Nicholls & Ayers, 2007). However, Nicholls and Ayers (2007) also found some parents to adopt an overprotective and anxious style of parenting, where mothers shared feelings of not wanting their baby to be out of their sight (Nicholls & Ayers, 2007). An overprotective parenting style may be associated with ongoing concerns about the child's health following the birth and following neonatal period (Thomasgard & Metz, 1993).

Although there is little conclusive evidence to date, it is possible that maternal birth-related PTS is related to less optimal emotional and behavioural development in children, perhaps due to the impact of PTS on the parent-child relationship.

3.1.6 The Current Study

In summary, the review of the current literature on the development of children following HIE lacks data in several ways. Firstly, there is little research investigating long-term outcomes of children with neonatal HIE and TH and without major neuro-disability, particularly in relation to their emotional and behavioural development. Secondly, qualitative data suggest that parents of children with HIE are at elevated risk to experience birth-related stress. However, to our knowledge, there is currently no quantitative evidence specifically investigating the level of birth-

related distress reported by parents of children with neonatal HIE, nor comparing this with parents of low-risk births. Thirdly, to our knowledge, there is currently no published data which examines the association between parental reported birth-related stress following neonatal HIE and children's emotional and behavioural difficulties.

The present study, therefore, has several aims. Firstly, it will examine the levels of emotional and behavioural difficulties at school-age (i.e., six to eight years of age) in children with neonatal HIE who received TH and have no major neuro-developmental disability compared to typically developing children. Secondly, this study will compare the rates of birth-related stress reported by parents of children with neonatal HIE compared with parents of typically developing children. Thirdly, the present study will examine the association between birth-related stress and emotional and behavioural difficulties. Finally, the study will examine whether parental reported birth-related stress mediates the relationship between neonatal HIE and emotional and behavioural difficulties in childhood. Improving understanding of the relationship between environmental factors (i.e., parental birth-related distress) and school-age emotional and behavioural difficulties is crucial to help guide prevention and intervention.

It was hypothesised that parents within the HIE group would report greater birth-related stress, and that children within the HIE group would demonstrate greater internalising and externalising difficulties. Furthermore, it was hypothesised that increased emotional and behavioural difficulties in children with HIE would be mediated through increased parental birth-related stress.

3.2 Method

3.2.1 Procedure

The study is part of a larger ongoing study aimed at understanding the school-age outcomes (i.e., at six to eight years of age) of children with neonatal HIE and TH (NENAH). The larger study assesses data from children, parents, and teachers on a wide range of variables (e.g., cognitive, behavioural and brain structure). The wider study was approved by the NHS Research Ethics Committee – Liverpool and North Central (IRAS Number: 263965). The present study specifically examines data relating to maternal birth-related stress and children's internalising and externalising difficulties, as described in the measures below. Accordingly, data were collected from parents and teacher questionnaires. The design and procedure for the current study was approved by the Ethics and Research Governance Online (ERGO) system at the University of Southampton (ERGO Number: 62791; Appendix D).

For the HIE sample, children were recruited from a clinical database; all children had received TH following HIE at the same hospital. Eligible participants were approached by the researchers and provided with an invitation letter if they were interested in taking part (Appendix E). Participants for the control sample were recruited using opportunity sampling. Schools in the same area as HIE participants were contacted to see if they were willing to advertise the study. Schools were provided a study advert (Appendix F), formal letter (Appendix G) and invitation letter for parents (Appendix H). Additionally, control participants were recruited using a friends, family, and staff approach. Following agreement to take part in the study, information sheets were provided to parents of HIE (Appendix I) and control (Appendix J) children.

After parental informed consent (Appendix K) was obtained, questionnaire data from parents was collected and teachers were requested to complete questionnaires. All assessment data was gathered when children were aged between six and eight years of age. Parents and teachers either completed paper questionnaires by hand or online via Microsoft Forms. After the data was completed, all data was then anonymised and combined into SPSS for analysis.

3.2.2 Design

In the present study, a cross-sectional design was employed, whereby participants were required to complete questionnaire measures when children were aged between six and eight years of age. Parents and teachers completed either paper or online questionnaires. The independent variable in the current study was HIE status (i.e., HIE or control group). Parental birth-related stress was both a dependent variable as well as a possible mediator of the effects of HIE on children's emotional and behavioural difficulties (i.e., internalising and externalising problems). Children's emotional and behavioural difficulties (assessed through both parent and teacher reports) was a dependent variable.

3.2.3 Participants

Participants in the current study were mothers and teachers of children who were aged between six and eight years of age. Other inclusion criteria were: children born >36+6 weeks gestational age. For the HIE group, children were required to have received HT. Exclusion criteria were: children with major neuro-developmental disability (i.e., cerebral palsy and a gross motor function of level >2 on the Gross Motor Function Classification System), children with medical conditions that could affect their development (e.g., underlying genetic syndrome, endocrinologic disease, neuromuscular disease), and parents whose command of English was not adequate enough to complete the questionnaires. Within the HIE group, the final sample of participants

included mothers of 45 children (28 females, 17 males). Typically developing children were recruited for the control group. Participants comprised mothers of 28 children (14 females, 14 males). The HIE and control group were aimed to be matched as close as possible on child sex and age. Additionally, teachers completed questionnaire measures (n=28 for the HIE group, 17 females, 11 males; n=21 for the control group, eight females, 13 males).

3.2.4 Measures

3.2.4.1 Maternal birth-related PTS.

The Modified Perinatal Post Traumatic Stress Disorders Questionnaire (PPQ; Callahan et al., 2006) is a 14-item measure of PTS related to childbirth and the ensuing postnatal period, originally developed for mothers of high-risk infants (DeMier et al., 1996). Items are rated on a 0-4 scale: *not at all* (0), *once or twice* (1), *sometimes* (2), *often but less than once a month* (3), *often more than once a month* (4). Scores can therefore range between 0 and 56. In the present study, mothers were asked to report on their current distress related to the childbirth experience. Though it is not a diagnostic tool, it measures three components of PTSD: intrusion, avoidance and increased arousal. A score of 19 or above is considered an indicator of clinically significant distress (Callahan et al., 2006). It has been found to have good validity (Callahan et al., 2006; Quinnell & Hynan, 1999) and internal consistency ($\alpha = .90$) (Callahan et al., 2006), even in mothers whose birth experiences were more distant (Zerach et al., 2015). The present study found high internal consistency, with a Chronbach's alpha of 0.95.

3.2.4.2 Child emotional and behavioural functioning

Parents completed the school-age Child Behavior Checklist (CBCL; Achenbach & Rescorla, 2001). This is a widely used measure of emotional and behavioural difficulties in children and young people aged between six and 18 years, used in both clinical and research settings (Ebesutani et al., 2010). The parent version comprises 113 items which are rated on a 0-2 scale: *not true* (0), *somewhat true* (1) or *very true* (2). Parents are asked to report on their child's behaviour in the past six months. In the present study, raw scores from the internalising problems subscale (sum of rating for items comprising the withdrawn, anxious/depressed and somatic complaints subscales, 32 items in total) and the externalising problems subscale (sum of rating for items comprising the rule-breaking and aggressive behaviours subscales, 35 items in total) were used for analysis. A higher score is representative of greater difficulties. Raw scores on the internalising and externalising subscales can be converted to *T* scores to be categorised as: normal (*T* score below 60), borderline clinical (high enough to be of concern; *T* score of 60-63) and clinical (clinically significant scores; *T* scores above 63). The CBCL has been found to have good reliability

and validity for use across cultures (Crijnen et al., 1999; Ivanova et al., 2007). In the current study, the internalising and externalising subscales showed good internal consistency (α = 0.90, α = 0.93 respectively).

The Teacher's Report Form (TRF; Achenbach, 1991) is the teacher version of the CBCL. 113 items are scored on a 0-2 scale (as above). Teachers are asked to report on the child's behaviour within the past two months. Raw scores for the internalising (35 items in total) and externalising (32 items in total) subscales were used as above. In the current sample, the internal consistency of the internalising and externalising subscales was 0.89 and 0.91 respectively (Chronach's alpha).

3.3 Results

3.3.1 Data Analysis Plan

Statistical analysis was completed using IBM SPSS Statistics (Version 28). Preliminary analyses were carried on the data separately according to group status (HIE or control) for each total or subscale score on questionnaire measures to screen for violations of assumptions for parametric tests. Normality of the variables was checked by visual inspections of histograms (Appendix L). Descriptive statistics were computed to generate the mean and standard deviation of the all the variables. Descriptive statistics were also computed to generate the percentage of children within the normal, borderline clinical and clinical ranges on the CBCL and TRF.

Analyses were carried out to explore any sex differences on the variables. No significant differences were found on any of the measures according to child sex which warranted further exploration.

Analyses explored differences between the HIE and control group on parental birth-related stress and emotional and behavioural outcomes using independent t-tests. Because of the non-normal distribution of the PPQ, CBCL and TRF scores, bootstrapping was used to obtain bias corrected accelerated 95% confidence intervals. In all cases where homogeneity of variance was violated, a t-test statistic with adjusted degrees of freedom is reported that takes this into account. All t-tests were confirmed by non-parametric Mann-Whitney U tests; as such, only parametric results are reported. As data were not normally distributed, Spearman's rank correlations were computed to explore the associations between the variables.

To investigate whether HIE exerts an effect on emotional and behavioural difficulties directly and/or indirectly through maternal PTS, mediation analysis as described by Hayes (2018) using PROCESS (Model 4) was applied. The method assesses: the total effect of a given predictor

(X) on an outcome (Y) without the mediator (M); the direct effect of X on Y while including M; and the indirect effect of X to Y through M. In our study, group status (i.e., HIE – coded as '*1*' vs. typically developing – coded as '*2*') was the predictor (X) for the internalising and externalising difficulties (as reported by parents) with the maternal PPQ score as the possible mediator. As there were no significant group differences on teacher reported internalising and externalising difficulties, and no trend in correlations were found between PPQ scores with teacher reported internalising and externalising difficulties, mediational analysis was not performed for TRF scores. Therefore, a total of two mediational analyses were conducted (i.e., separate mediational analyses for parent reported internalising and externalising difficulties as the outcome variable). For all analyses, thresholds for statistical significance were set at $p<.05$.

3.3.2 Sample characteristics

The original group of HIE participants included parents of 50 children, whereas the control group comprised 32. For five HIE and four control participants, at least one parental questionnaire measure was not available; these participants were excluded from further analyses. Therefore, the final sample included 45 HIE and 28 control participants. Of the final sample, 28 teachers completed the TRF for the HIE group, and 21 teachers completed the TRF in the control group. Characteristics of the HIE and control children are shown in Table 1.

Table 1 *Child Sample Characteristics*

	HIE (n=45)	Control (n=28)
	M (SD) or N (%)	M (SD) or N (%)
Child sex		
Male	17 (37.8%)	14 (50%)
Female	28 (62.2%)	14 (50%)
Child age	6.66 (.74)	7.39 (.73)
Child ethnicity		
White British	32 (71.1%)	22 (78.6%)
Mixed Ethnicity	1 (2.2%)	3 (10.7%)
Asian	1 (2.2%)	0 (0%)
Data not available	11 (24.4%)	3 (10.7%)

3.3.3 Descriptive statistics

Descriptive statistics highlighted that, in comparison to control children, a larger proportion of children in the HIE group had CBCL and TRF scores on the internalising and externalising scales that fell in the borderline or clinical range (Appendix M). For PPQ scores, 16 mothers within the HIE group (35.56% of the HIE sample) reported symptoms which indicated clinically significant birth-related stress (i.e., a total score of 19 or above). Only one mother (3.57% of the control sample) in the control group reported clinically significant levels of birth-related stress.

Descriptive statistics also demonstrated that, across both study groups, there was wide variation in individual scores on all measures (PPQ, CBCL, TRF). However, the maximum reported scores across the measures were consistently higher in the HIE group (Appendix M). Mean scores on the CBCL and TRF highlighted that, on average, teachers rated both HIE and control children lower on internalising and externalising difficulties than parents (see Tables 2 and 3).

3.3.4 Group differences in birth-related PTS and in emotional and behavioural difficulties

Significant differences were found in PPQ scores and certain subscales of the CBCL and TRF. Non-parametric testing produced similar results. In particular, Table 2 shows that mothers of HIE children reported significantly greater birth-related PTS (M = 15.82, SE = 2.13) than mothers of typically developing children (M = 3.36, SE = 1.04). This difference, 12.47, BCa 95% CI [7.75, 17.23], was significant $t(62.15) = 5.31$, $p<.001$. The effect size for this analysis ($d = 1.07$) was found to exceed Cohen's (1988) convention for a large effect ($d = .80$).

Furthermore, parents reported children with HIE to display greater internalising difficulties (M=9.40, SE = 1.04) than control children (M = 5.57, SE = 1.09). The difference between the two groups, 3.83, BCa 95% CI [0.61, 7.37, was significant $t(71) = 2.08$, $p=0.03$. Parents also reported children with HIE to display greater externalising difficulties (M = 8.89, SE = 1.32) than control children (M = 4.14, SE = 0.95), and this difference, 4.75, BCa 95% CI [1.66, 7.82] was significant, $t(70.62) = 2.87$, $p=.007$. The effect sizes for parental reported internalising and externalising difficulties ($d = .50$ and $d = .61$ respectively) represented Cohen's convention for a medium effect ($d = .50$).

Table 3 shows that, although teachers reported greater internalising and externalising difficulties for children with HIE than control children, these differences were not statistically significant. However, a medium effect was found for both internalising and externalising difficulties ($d = .46$ and $d = .52$ respectively).

Table 2 *Comparison Between HIE and Control Sample on Parent Reported Birth-related Stress and Internalising and Externalising Difficulties.*

	HIE (n=45)		Control (n=28)		Difference (95% CI)	*p*-value*
	M	SE	M	SE		
PPQ	15.82	2.13	3.36	1.04	12.47 [7.75, 17.23]	>.001
CBCL-I	9.40	1.04	5.57	1.09	3.83 [0.61, 7.37]	.030
CBCL-E	8.89	1.32	4.14	0.95	4.75 [1.66, 7.82]	.007

Note. PPQ: Perinatal Post-traumatic Stress Disorders Questionnaire total score; CBCL-PI: Child Behavior Checklist internalising difficulties total raw score; CBCL-E: Child Behavior Checklist externalising difficulties total raw score; M: mean; SE = standard error. **p*-values calculated by independent t-tests.

Table 3 *Comparison Between HIE and Control Sample on Teacher Reported Internalising and Externalising Difficulties.*

	HIE (n=28)		Control (n=21)		Difference (95% CI)	*p*-value*
	M	SE	M	SE		
TRF-I	4.75	1.19	2.38	0.63	2.37 [-.14, 5.32]	.137
TRF-E	3.61	1.17	1.05	0.41	2.56 [.35, 5.11]	.076

Note. TRF-I: Teacher Report Form internalising difficulties total raw score; TRF-E: Teacher Report Form externalising difficulties total raw score; M: mean; SD: standard deviation. **p*-values calculated by independent t-tests.

3.3.5 Correlations between measures

Spearman's rank correlations were used to explore the significance and direction of the relationship between the study variables (maternal PTS symptoms, child emotional and behavioural difficulties). Correlations are presented in Table 4. The data indicates that maternal birth-related PTS were significantly positively correlated to parent reported internalising difficulties (r = .504, p<.001) and externalising difficulties (r = .512, p<.001). As expected, children of mothers who reported greater birth-related distress were reported by parents to have greater internalising and externalising difficulties. However, birth-related PTS was not significantly correlated with teacher reports of internalising (r = -.021, p = .86) or externalising difficulties (r = .022, p =.88).

Interestingly, while parent reported internalising difficulties were not significantly correlated with teacher reported internalising difficulties (r = -.134, p = .36), externalising difficulties reported by teachers and parents were significantly positively correlated (r = -.440, p = .002).

Spearman's correlations were also conducted between the measures according to group status (HIE and control). Patterns of associations and the strength of these correlations were generally comparable to the correlations presented in Table 4, however there were some differences related to the statistical significance of the correlations, likely due to the smaller sample size in the control group (see Appendix N for separate correlation tables for the HIE and control group).

Table 4 *Spearman's Rank Correlations for Maternal Birth-related Stress, Parent Reported Internalising and Externalising Difficulties and Teacher Reported Internalising and Externalising Difficulties.*

Variable	1.	2.	3.	4.	5.
PPQ	-				
CBCL-I	.504***	-			
CBCL-E	.512***	.593***	-		
TRF-I	-.021	.134	.126	-	
TRF-E	.022	.145	.440**	.621***	-

Note. *p<.05; **p<.01; ***p<.001

3.3.6 Mediation analysis

In order to test whether maternal birth-related PTS mediated the effects of group status (i.e., HIE or typically developing) on children's emotional and behavioural outcomes (CBCL-I, CBCL-E, TRF-I, TRF-E), mediational analyses were performed.

3.3.6.1 Parent reported internalising difficulties

Figure 1 shows the results of the mediation analysis for CBCL internalising and externalising scores. The total effect of HIE group status on parental reported internalising difficulties was significant, b = -3.83, SE = 1.84, 95% CI [-7.50, -0.16], indicating that HIE was related to greater parental reported internalising difficulties. There was a significant relationship between group

status and birth-related PTS, *b* = -12.47, *SE* = 2.80, 95% CI [-18.05, -6.89]; suggesting that mothers within the HIE group reported greater birth-related PTS. There was also a significant positive relationship between birth-related PTS and parental reported internalising difficulties, with greater birth-related PTS being associated with a higher number of internalising difficulties in children, *b* = 0.22, *SE* = 0.07, 95% CI [0.07, 0.37]. When accounting for the mediator (birth-related PTS), the relationship between HIE group status on parental reported internalising difficulties was not significant, *b* = -1.12, *SE* = 1.98, 95% CI [-5.06, 2.82]. However, the indirect effect was significant, suggesting that HIE group status can affect children's parental reported internalising difficulties via parental birth-related PTS, *b* = -2.71, *SE* = 1.24, 95% CI [-5.43, 0.65]. Parents within the HIE group reported higher birth-related PTS, which predicted greater internalising difficulties as reported by parents.

3.3.6.2 Parent reported externalising difficulties

The total effect of HIE group status on parental reported externalising difficulties was significant, *b* = -4.75, *SE* = 1.87, 95% CI [-8.47, -1.03], indicating HIE was related to greater parental reported externalising difficulties. There was a significant relationship between group status and birth-related PTS, as reported above. However, the relationship between birth-related PTS and parental reported externalising difficulties was not significant, *b* = 0.14, *SE* = 0.08, 95% CI [-.020, 0.29]. When accounting for birth-related PTS, the relationship between HIE group status on parental reported externalising difficulties was not significant, *b* = 3.06, *SE* = 2.08, 95% CI [-7.21, 1.09]. Furthermore, the indirect effect was not significant, suggesting that HIE status does not affect children's parental reported externalising difficulties via parental birth-related PTS, *b* = -1.69, *SE* = 1.33, 95% CI [-4.66, 0.53].

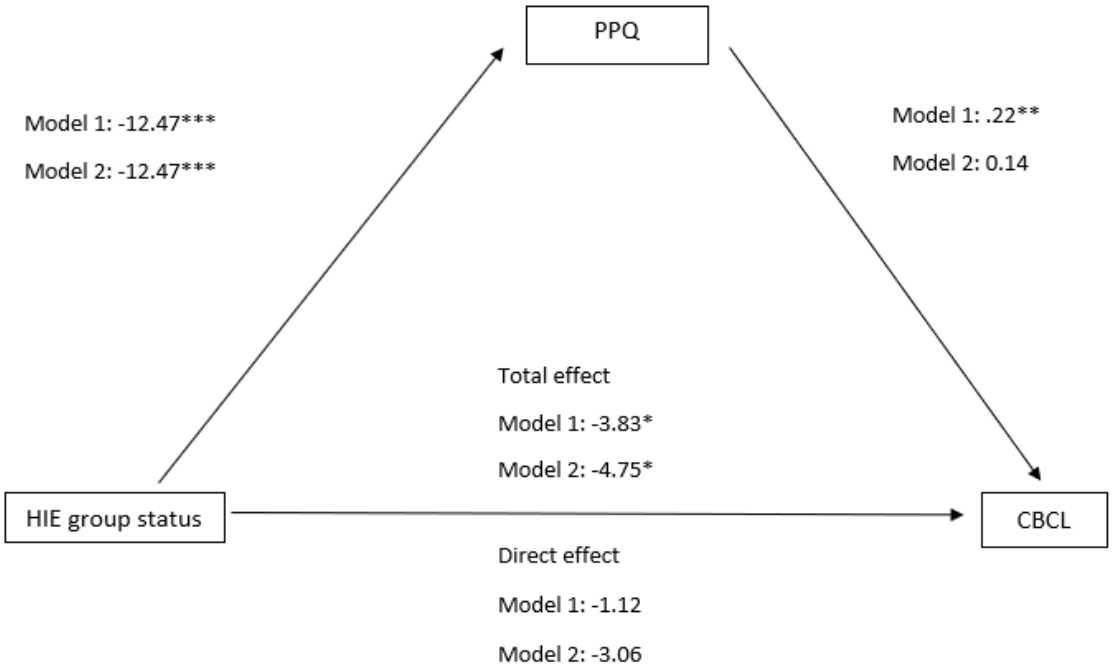


Figure 2 *Mediation Analysis Showing the Effect of HIE on Parental Reported Internalising and Externalising Difficulties via Maternal Birth-related Stress. Model 1 Refers to Mediation Analysis with CBCL-I as the Outcome Variable. Model 2 Refers to Mediation Analysis with CBCL-E as the Oucome Variable.*

Indirect Effect via:

Model 1, b = 2.71, 95% CI [-5.43, -.65]

Model 2, b = 1.69, 95% CI [-4.66, .53]

*Note. *p<.05; **p<.01; ***p<.001.*

3.4 Discussion

In this study, we explored the associations between HIE with TH, maternal birth-related PTS and children's internalising and externalising difficulties at school-age. Secondly, we tested the mediating effect of maternal birth-related PTS on the association between HIE and emotional and behavioural difficulties.

Firstly, as hypothesised, we found that, between six and eight years postpartum, mothers whose babies had neonatal HIE and received TH self-reported significantly higher levels of birth-related PTS symptoms. These findings are consistent with prior qualitative literature which suggests that giving birth to a child who has perinatal asphyxia with subsequent HIE is a stressful experience for parents (Bäcke et al., 2021; Nassef et al., 2013), and that feelings distress related to the birth can continue beyond the postpartum period (Heringhaus et al., 2013). It is also

consistent with prevalence data that highlights women who experience adverse events during pregnancy or labour to be at increased risk for experiencing birth-related PTS (Yildiz et al., 2017). It appears that, even many years following the birth, parents of children with HIE continue to experience high levels of birth-related distress. When considering previous literature using the PPQ, average scores within the HIE group are greater than general population samples (Hairston et al., 2018), and slightly elevated in comparison to other high-risk samples, such as pre-term and extremely low birthweight infants (Greene et al., 2015; Gondwe et al., 2020). The average levels of birth-related PTS reported appear to be comparable to those reported by mothers of extremely low birthweight infants with additional developmental difficulties (Zerach et al., 2015). Furthermore, 35.56% of the mothers in the HIE sample reported birth-related PTS symptoms that indicated clinically significant levels of distress. The reported rate of clinically significant birth-related PTS in the HIE sample appears to be much greater than estimated prevalence rates of perinatal PTSD in the general population and high-risk samples (Dekel et al., 2017; Yildiz et al., 2017). Thus, our results suggest that, even many years following the birth experience, giving birth to a child who experiences neonatal HIE and receives subsequent TH may be related to significant PTS symptoms in mothers which may warrant further support.

Secondly, the findings provide evidence that parents of children with a history of neonatal HIE and TH report significantly increased levels of internalising and externalising difficulties at school-age compared to the levels reported by parents of typically developing children. For the teacher reports, the mean scores for internalising and externalising difficulties were found to be greater in the HIE group than the control group, but these differences were not found to be statistically significant. Of note is that the effect sizes for group differences on teacher reports of internalising and externalising difficulties were comparable to group differences on parent reports. It is therefore possible that the smaller sample size in the control group meant that it lacked statistical power to demonstrate a statistically significant effect.

While previous literature has produced inconsistent findings, the results of the current study are in line with the majority of the existing literature, which highlights that children with HIE who received TH are at increased risk of emotional and behavioural difficulties at school-age (Edmonds et al., 2022; Lee Kelland et al., 2020). Furthermore, the reported rates of internalising and externalising difficulties in the current HIE sample appear to be greater than other high-risk birth population samples. For instance, in the HIE sample, percentages of CBCL scores that fell in the borderline clinical and clinically significant ranges ranged between 11.1% to 28.89%. This is much greater than rates of borderline and clinically significant internalising and externalising difficulties in previous research which studied pre-term and very low birthweight children, where rates vary between 7% to 13.5% (Jin et al., 2020; Potijk et al., 2012; Reijneveld et al., 2006).

On average, across the sample, parents tended to report greater internalising and externalising difficulties than teachers. This discrepancy between parents and teacher report is not surprising, given previous research which found parents to report greater emotional and behavioural difficulties than teachers (Barnhill et al., 2000; Woo et al., 2007), particularly on internalising difficulties (Berg-Nielsen et al., 2012). There are various possible explanations for this finding. Firstly, children's behaviour is situation and context dependent (i.e., situational specificity; Achenbach et al., 1987). Moreover, the school and home environment are likely to influence children's behaviour in different ways. Importantly, internalising difficulties are less likely to be reported by teachers (Molins & Clopton, 2002), likely due to the challenges in identifying such difficulties in the school and classroom environment in comparison to externalising problems (Papandrea & Winefield, 2011). This may further help to explain why significant differences between the study groups were not found on teacher reported internalising and externalising difficulties. This also fits with our finding that parent and teacher reports of externalising, but not internalising, were significantly correlated. Secondly, it could be argued that mothers experiencing psychological distress may over-report child behaviour difficulties, as seen in mothers with anxiety and depression (Berg-Nielsen et al., 2003; Najman et al., 2001). Nonetheless, research has found increased parental ratings of emotional and behavioural difficulties to be stronger predictors of the child's later functioning than teacher reports (Ferdinand et al., 2007). Therefore, our findings are noteworthy.

Importantly, our results suggest that increased emotional and behavioural difficulties in children with HIE may partly be explained by mothers' birth-related PTS. Findings revealed increased maternal birth-related PTS as an important mechanism in the link between HIE and increased internalising difficulties as reported by parents. Conversely, although HIE was associated with increased parental reported externalising difficulties, this relationship was not explained by birth-related PTS. It is therefore apparent that, other mechanisms, that were not assessed in the current study, may underpin the association between HIE and externalising difficulties. Of relevance are findings from other studies on other outcomes for children with HIE. For instance, language (expressive and receptive) and executive function difficulties have also been identified in children with HIE (Edmonds et al., 2021; Schreglmann et al., 2020). Given their close associations with behavioural problems (Chow & Wehby, 2018; Dai et al., 2021), cognitive and language difficulties may be additional factors which place children with HIE at increased risk for externalising difficulties.

3.4.1 Strengths, Limitation and Future Directions

A key strength of this study is that children's emotional and behavioural outcomes were collected from both parents and teachers. This therefore allowed the influence of HIE and maternal birth-related PTS to be considered across two different and important contexts during childhood.

However, there are a number of limitations that must be considered when interpreting the findings, beyond those already considered (e.g., reporter bias). A key limitation is that the research did not include possible confounding variables that may affect maternal mental health and/or children's emotional and behavioural development, such as factors at the level of the individual (e.g., executive functioning; Dai et al., 2021), family (e.g., maternal depression and anxiety, mother-child relationship quality; Barker et al., 2011; Fihrer et al., 2009; Kok et al., 2013) and community (e.g., socioeconomic deprivation; Ban et al., 2012; Piotrowska et al., 2015). Variables related to the quality of the mother-child relationship are of particular relevance for inclusion in future studies, given their potential to mediate the association between maternal postpartum mental health and child psychosocial development. We are therefore unable to conclude whether the observed associations are influenced by these factors. It will be crucial for future research to consider such variables, to better understand the relevance and interplay of a range of biological and environmental factors on the development of children with HIE.

Secondly, only mother's birth-related distress was assessed. The existing qualitative literature which has included fathers of children with birth asphyxia and who receive TH suggests that they also experience distress related to the birth experience (Heringhaus et al., 2013; Thyagarajan et al., 2013). It would therefore be useful for future research to include measures of father's and partner's birth-related PTS. Previous research highlights fathers of infants in the NICU to experience stress, especially those of high-risk birth populations (e.g., low birthweight or pre-term infants; Prouhet et al., 2018).

Thirdly, the size of the groups varied considerably. Compared to the sample of HIE children (n=45), the sample size of typically developing children was relatively small (n=28). It is important to note that, due to the ongoing nature of the wider NENAH project, the findings reported in the present study are based on the current available data, rather than the final sample. Although the current study utilises a slightly larger sample than previous research exploring behavioural outcomes of children with HIE (Lee-Kelland et al., 2020; van Schie et al., 2015), employing a larger sample size will be important in future research so that findings have the potential to be more widely generalised.

Finally, the cross-sectional nature of the study means that causality cannot be determined. In the future, longitudinal research examining measures from birth to school-age will be beneficial in better understanding developmental trajectories for children with HIE.

3.4.2 Conclusions

The long-term impact of neonatal HIE and TH is a fairly recent and developing area of research. The current study makes a significant contribution to our understanding of the impact of HIE with TH on both parent and child outcomes when children are of school-age. Our results suggest that mothers of children with HIE experience significantly greater birth-related PTS, six to eight years after the birth. Furthermore, while medical advances have substantially improved outcomes for children with HIE, our findings suggest that children remain at increased risk of experiencing emotional and behavioural difficulties at school-age, particularly within the home environment. While maternal birth-related PTS helps to explain parental reported internalising behaviours in children with HIE, other factors beyond parental birth-related stress are also important for the development of HIE children, particularly in relation to parental reported externalising behaviours. Possible influences, including but not limited to, cognitive and language factors (Shreglmann et al., 2020), will be important to consider in future research. Additionally, our findings need to be confirmed by further studies, as this is one of very few studies to assess both maternal birth-related PTS and school-age emotional and behavioural outcomes in children with HIE.

3.4.3 Implications

The findings suggest several implications. Firstly, this group of parents and children have so far received little attention within the literature to date, particularly in relation to longer-term outcomes. The current study highlighted the potential long-term impact of neonatal HIE and TH for children and mothers, in relation to both child psychosocial development and parental mental health. As a result, it is crucial to raise awareness of the possible vulnerabilities associated with neonatal HIE and TH, for both parents and children. This will be important within both health and educational practice. While it may be difficult to change the birth experience itself, identifying mothers experiencing substantial birth-related distress and providing appropriate support at the early postnatal stage will be important to help reduce the risk of ongoing distress and related emotional and behavioural difficulties in children. It will also be important to investigate whether later intervention may be required to reduce long-term birth-related stress in mothers of children with HIE.

In terms of implications for Educational Psychology practice, a key implication will be to raise awareness and understanding among educational professionals of the potential vulnerabilities within parents and children following neonatal HIE and TH. This will be important for EPs and school staff to feel sufficiently equipped to support these children in school. For instance, it will be imperative that there is recognition among educational staff regarding the increased risk of emotional and behavioural difficulties for children with HIE, so that additional support and intervention can be put in place as appropriate. However, it will also be important to recognise that the nature of the needs of children with HIE is likely to widely vary and required support will need to be based on the individual needs of the child. EPs are well placed to support school staff in understanding and meeting the individual emotional and behavioural needs of children with HIE. There is also a need for further research to improve our understanding of the school-age needs of children with HIE.

Furthermore, the current study has highlighted the impact of traumatic birth experiences on both maternal mental health and child outcomes, and the relationship between the two. EPs have a role to play within schools in promoting an understanding of the influence of children's early life experiences (including pregnancy and birth) on their development, such as through training and within consultations. This will be important so that key stakeholders supporting children (e.g., school staff and families) understand the child's behaviour within the context of their experiences (Bronfenbrenner, 1979). An important aim of providing such training to schools and modelling the importance of speaking to families about children's early life experiences, would be that school staff begin to feel confident and competent to independently gather this information.

Appendix A Data Extraction Table

Study code	Author & year	Country	Sample characteristics	Methodology	Key Findings	Limitations
1	Ayers, Wright & Wells (2007)	United Kingdom	n = 64 couples (mean age = 32.4). **Inclusion criteria for parents:** couples were cohabiting/married or in a long-term relationship together, the male was at birth **Inclusion criteria for infants:** the baby was born between 6	Cross-sectional – questionnaire measures completed 9 weeks after birth. **Measures:** *Parent-child relationships:* self-report version of the Bethlehem Mother-Infant interaction scale (adapted to measure parent-baby bond) *Birth-related post-traumatic stress:* IES	• Men and women did not differ on mean symptoms of intrusion and avoidance. • Compared to men, women reported fewer positive emotions in birth, more support in birth, more self-blame and a poorer parent-baby bond. • PTSD symptoms were not associated with the parent-baby bond.	• Low response rate (31%) • Recruited from one London hospital – limits generalisability.

			and 12 weeks before contact.			
2	Camicasa et al (2017)	Italy	n = 41 mother-infant dyads Data from a previous study – participants recruited from obstetric departments in two hospitals in Northern Italy. **Inclusion criteria for parents:** no psychiatric diagnosis, 18 years and older. **Inclusion criteria for infants:** healthy	Longitudinal - questionnaire and observational data: • First days after birth (PTS) • 3 months (PTS). • 17 months (mother-infant interactions and PTS). **Measures:** *Parent-child relationships:* PSI-short form and maternal mind-mindedness measured via observation that was videotaped and coded using the mind-mindedness manual (during a 20-minute free play session). PSI – parent-child dysfunctional interaction	• PTS scores: mean of the total symptoms did not significantly change between 87 hours and 17 months postpartum (p =.23). • Correlations: PTS at both 87 hours and 3 months were not significantly associated with PCDI on the PSI or any of the mind-mindedness variables at 17 months. • PTS at 17 months: hyper-arousal symptoms were correlated with PCDI (r=.31) and negatively correlated with maternal mind-mindedness (r=-.36). • Mediation: maternal mind-mindedness fully mediated the association between hyper-arousal PTS symptoms and PCDI (p<.001).	• Small sample size – limits generalisability

			baby with Apgar score >7 at 5 minutes after birth.	*Birth-related post-traumatic stress:* PPQ		
3	Davies, Slade, Wright & Stewart (2008)	United Kingdom	n = 211 women. **Inclusion criteria for parents:** over 16 years of age, women must have delivered a healthy infant. **Exclusion criteria for parents:** if women had experienced a specific adverse clinical event during labour and/or delivery or if their infants had required cardiopulmonary resuscitation following delivery,	The relationship between birth-related PTS and parent-child relationships was cross-sectional – questionnaires. Measures completed at 6 weeks postpartum. **Measures:** *Parent-child relationships:* MPAS and the MORS-SF. *Birth-related post-traumatic stress:* Post-traumatic stress disorder questionnaire (PTSDQ) within the context of childbirth and IES.	• Mothers categorised based on PTS symptoms: fully symptomatic, partially symptomatic, non-symptomatic. Group comparisons: • Compared to non-symptomatic, mothers with full or partial post-traumatic stress symptoms viewed their infants as being less warm towards them (p<.001) and more invasive (p<.0001). • They also perceived their attachment to their infants to be of lower quality (p<.0001), including greater infant-direct hostility (p<.0001). Correlations: • All PTSDQ subscales (re-experiencing total, avoidance total, arousal total, total stress) were significantly correlated (either at .05 or .01) with: • decreased warmth and increased arousal on the MORS • Lower quality of attachment, more infant-directed hostility and less pleasure in interaction (except for re-experiencing). • When depressive symptomatology was added as a covariate, the only significant main effect remaining was for the MORS warmth scale (p<.034).	• Self-report • Recruited from one postpartum inpatients ward at a maternity hospital – limits generalisability.

			women with significant social problems and/or a history of mental health problems. **Exclusion criteria for infants:** if the infant had been admitted to a SCBU,			
4	Dekel et al (2019)	Global (North America – 66%)	n = 685 women (mean age = 31) **Inclusion criteria for parents:** women were over 18 years, women gave birth in the last 6 months	Cross-sectional – questionnaire. **Measures:** *Parent-child relationships:* Maternal attachment inventory *Birth-related post-traumatic stress:* PTSD checklist for DSM-V, with 'most recent childbirth' as index event.	• Significantly lower attachment levels in postpartum PTSD than no PTSD or general PTSD (p<.001). Multiple regression (pre-childbirth variable, post-childbirth attachment-related variables and postpartum PTSD and general PTSD): • Variables accounted for 20.9% of the variance in predicting maternal attachment (p<.001). • Mother's distress during birth and complication in the infant: each had a significant contribution of 7% of the variance (greater distress, lower attachment). Postpartum PTSD (but not general PTSD) added a significant contribution above and beyond pre-morbid factors.	• Self-report

5	Ertan et al (2021)	France	n = 916 mothers and 64 partners. **Exclusion criteria for parents:** women/partners of women who gave birth less than 1 month or more than 1 year ago, persons under guardianship or curatorship	Cross-sectional online self-report questionnaire (1 to 12 months postpartum). **Measures:** Parents were asked: "Have you experienced particularly upsetting or traumatic events during childbirth?" *Parent-child relationships:* Mother-to-Infant Bonding Scale (MIBS) – validated in French. Partners completed perceived mother-child bond (MIB). *Birth-related post-traumatic stress:* City Birth Trauma Scale (CBTS) – a non-validated French version.	Women: • Significant positive correlations between perceived mother-child bond and the CBTS birth-related symptoms subscale (.345), general symptoms subscale (.366) and total symptoms (.412). Correlations significant at the 0.01 level. Partners: • Positive correlation between MIB and CBTS general symptoms (.378) and total symptoms (.338). Correlations significant at the 0.01 level. • No significant correlation between MIB and CBTS birth-related symptoms (.133).	• Cross-sectional – cannot determine causality. • French version of the CBTS was not validated. • Unclear whether partners were present at the birth or not.

6	Feeley et al (2011)	Canada	n = 21 mothers of ***very low birthweight*** infants (mean age = 30.9). **Inclusion criteria for infants:** if the infant was hospitalised in the NICU and weighed less than 1,500g at birth. .	Cross-sectional – measures completed at 6 months corrected age. **Measures:** *Parent-child relationships:* Observation of 15-minutes of mother-infant play, coded with the Emotional Availability Scales. *Birth-related post-traumatic stress:* PPQ.	• Mothers who reported more PTSD symptoms were observed to be less sensitive (-.49, p<.05) and less effective at structuring interactions with their infant (-.47, p<.05). • Correlations between PTSD symptoms with non-intrusiveness and non-hostility were non-significant.	• Prebirth PTSD symptoms of participants were not measured. • Small sample (recruited from one hospital in Canada) – limits generalisability.
7	Forcada-Geux et al (2011)	Switzerland	n = 47 mother-infant dyads of ***pre-term infants***, 25 mother-infant dyads of full-term infants. **Exclusion criteria for pre-term parents:**	Longitudinal • 6 months corrected age: mother-child relationship (10-minute recorded play session). • 18-months corrected age: birth-related stress. **Measures:**	• Multivariate test of mother-infant relationship according to groups (low-stress premature, high-stress premature, full-term): • 6 months: percentage of sensitive mother and cooperative infant dyads was significantly lower in pre-term dyads (high and low PTS) compared with full-term dyads. Maternal control-compliant infant was significantly higher in high stress compared to low stress and full-term dyads. • Mothers of pre-term infants (regardless of PTS status) showed significantly less balanced	• PTS measured months after the mother-child relationship was assessed.

			parents' psychiatric illness and/or drug abuse, difficulties speaking French. **Exclusion criteria for pre-term infants:** infant malformation, chromosomic abnormality and fetopathy. **Exclusion criteria for control parents:** difficulties during pregnancy or delivery, somatic abnormalities, parents' psychiatric problems and language difficulties.	*Parent-child relationships:* Working Model of the Child Interview and mother-infant interaction play session coded according to the third revision of the Care Index. *Birth-related post-traumatic stress:* PPQ Perinatal Risk Inventory used to describe the severity of the infant's perinatal problems – considered high-risk/low-risk.	representations of their infants than mothers of full-term.	
8	Hairston et al (2018)	Israel	n = 114 mothers	Cross-sectional online questionnaires.	Correlations:	• Cross-sectional – cannot determine causality.

			Infant age = 4 to 12 weeks. Recruited from a delivery ward in Israel, internet advertisements and snowballing. **Exclusion criteria for parents:** birth before or at 32 weeks. **Exclusion criteria for infants:** infants with a chronic illness.	**Measures:** *Parent-child relationships:* the Hebrew translation of the PBQ. *Birth-related post-traumatic stress:* Hebrew translation of the modified PPQ.	• PPQ significantly correlated with PBQ general (.415, p<.029), PBQ anger (.426, p<.029) and PBQ anxiety (.372, p<.029). Mediation analysis: • PPQ was a significant mediator between maternal attachment styles and bonding, specifically: • PPQ significantly mediated the relationship between mother's avoidant style and PBQ anger. The direct effect of PPQ on PBQ anger was significant (p<.05). PBQ did not significantly mediate the relationship between mother's anxious/ambivalent style and PBQ anxiety. The direct effect of PPQ on PBQ anxiety was not significant (p=.547).	• Self-report.
9	Handelzalts et al (2019)	Israel	n = 504 mothers (mean age = 30.9) Infants aged 0 to 13 months.	Cross-sectional online questionnaire. **Measures:** *Parent-child relationships:*	Correlations: • PBQ significantly correlated with CBTS total (.38, p<.01) and CBTS general symptoms (.47, p<.01), but not CBTS birth symptoms (.06) Mediation: • Demographic variables that were significantly correlated with bonding (the number of	• Cross-sectional – cannot determine causality. • Did not control for prenatal depression/PTSD. • Self-report.

			Inclusion criteria fir parents: mothers older than 18 years, singleton pregnancy within the previous 13 months.	PBQ *Birth-related post-traumatic stress:* City Birth Trauma Scale Other variables explored: Postnatal depression, resilience (dispositional optimism) and neuroticism.	children and education level) were included within the model. • No significant direct relationship between personality traits and bonding, but significant indirect paths were found: both resilience and neuroticism affected PBQ through postnatal depression, but not through CBTS total symptoms. • No significant relationship was found between CBTS total symptoms and PBQ. CBTS subscales – significant indirect paths: • Resilience and PBQ through CBTS general symptoms (p=.019). • Neuroticism and PBQ through CBTS birth symptoms (p=.013) and CBTS general symptoms (p=.001). The relationship between CBTS birth symptoms and bonding was negative (-.12, p<.01; greater PTSD symptoms = fewer bonding difficulties)	
10	Handelzalts et al (2021)	Israel	n = 210 women (mean age = 32.14)	Longitudinal online self-report questionnaire: • 1-4 days postpartum (demographic information) • two months postpartum (online completion of CBTS) • six months postpartum (online completion of	• Correlations: birth-related PTSD symptoms significantly correlated with bonding (.36, p<.01), general symptoms significantly correlated with bonding (.52, p<.01). Mediation model: • Controlled for significant covariates (university education and birth type). • The relationship between adult attachment styles and postpartum bonding was fully mediated by postpartum psychopathology	• Participants recruited from one hospital – limits. generalisability. • All self-report measures. • High drop-out rate (only 34.54% of original sample at final stage).

				postpartum bonding questionnaire - PBQ) **Measures:** *Parent-child relationships:* Hebrew version of the PBQ *Birth-related post-traumatic stress:* the validated Hebrew version of the CBTS – birth-related symptoms and general symptoms. The study also measured adult attachment styles and postpartum depression.	(general PTSD symptoms and postpartum depression, not birth-related PTSD). Once insecure attachment styles, PPD and general related symptoms were included in the model, postpartum PTSD birth-related symptoms were not significantly related to poorer bonding. However postpartum PTSD general symptoms were (p<.01).	
11	Ionio & Di Blasio (2014)	Italy	n = 58 pregnant women. 19 women attended all four stages of the study.	Longitudinal – questionnaire and observational data: • seventh month of pregnancy • two days postpartum (PPQ) • two months postpartum (PPQ) • three months postpartum (recording of mother-infant interaction).	• PPQ symptoms 2 days vs 2 months: no significant differences. Correlations among PTSD symptoms at 2 months and IRSS variables: • PTSD positively correlated with: • Crying (.537, p<.05), arch position (.537, p<.05) and disorganised behaviour during the play phase (.547, p<.05) • Looking away during the still phase (.491, p<.05).	• High attrition rate. • Small sample size – limits generalisability.

			Exclusion criteria for parents: women with pregnancy-related disturbances and/or personality disorders.	**Measures:** *Parent-child relationships:* Still Face paradigm – three 2-minute phases (free play, still face, resume normal interaction). Recorded and coded using the Infant Regulatory Scoring System (IRSS) and Maternal Regulatory Scoring System (MRSS). *Birth-related post-traumatic stress:* PPQ	• No significant correlations during the reunion phase. Linear regression between PTSD 2 months and IRSS/MRSS: IRSS: • Play episode: mothers with many total stress symptoms had children that put themselves in arch position (p=.02) and show a large number of disorganised behaviours with strong displays of distress and uncontrollable and inconsolable crying (p = .018). • Still episode: looked away from mother (p=0.03) and turned aside and away from their mother (p = .05). • Reunion episode: children put themselves in the arch position (p = 0.05). MRSS: • Mothers with a high number of PTSD symptoms during the play session did not directly look at their child (p= .01), described their child's status in a negative way (p = .05) made sounds with their mouth to catch the child's attention (p = .01), sang (p = .04) and kept on touching their child (p=.01). • Reunion episode: did not keep a middle distance from their child (p=.012), did not look directly at the children's face (p=.02), did not sing (p=.04) but made sounds with their mouth to catch the children's attention (p=.02).	

12	Ionio et al (2017)	Italy	n = 45 mothers and fathers of ***pre-term infants***, 36 mothers and fathers of full-term infants. **Exclusion criteria for infants:** presence of congenital anomalies, major sensory impairment, severe brain injuries and other neurological complications.	Longitudinal/case-control Birth-related stress completed within 7 to 14 days from delivery. Parenting stress index and observations of mother-infant interactions (5 minute free-play) at 3 months. **Measures:** *Parent-child relationships:* PSI short-form, mother-infant interactions observed, videotaped and coded by the Global Rating Scales (four maternal dimensions – sensitivity, intrusiveness, remoteness, signs of depression, three infant dimensions – communicative, inert, distressed).	• Parent- child relationships: Mothers of pre-term infants had significantly higher levels of intrusiveness (p=.029) and remoteness (p=.015). • PTS: No significant differences were found between mothers' and fathers' IES-R scores or 'parent-child dysfunctional interaction' subscale of the PSI. Pre-term sample correlations: • Mothers: significant correlations were found between the hyperarousal subscale of the IES-R and 'distressed' on the infant GRS (-.466, p<.01) • Fathers: avoidance subscale of the IES-R was correlated with mother's remoteness (-.611, p<.01) and signs of depression (-.529, p<.01). Linear regression (mother-child interactions in the pre-term sample): • Father's avoidance on IES significantly predicted mother's remoteness (p<.01) and signs of depression (p<.05).	• Limited sample size – from one hospital.

				The parenting stress index – short form (Italian version) which includes the 'parent-child dysfunctional interaction' subscale – did not look at the relationship between the PCDI and birth-related PTS. *Birth-related post-traumatic stress:* Impact of event sale revised (IES-R) – Italian version. Includes three clusters: avoidance, intrusion and hyperarousal		
13	Kjerulff et al (2021)	United States	n = 3006 women completed the 1-month postpartum interviews. 6 months: n = 2909, 12 months: n = 2802. Recruited from childbirth education	Prospective cohort study. Telephone interviews: • 1-month postpartum (PTS and parent-child relationships). • 6 months postpartum (parent-child relationships)	• Women who reported one or more CR-PTSD symptoms were approximately twice as likely to score in the bottom third on the postpartum bonding measure in comparison to women who did not report CR-PTSD, after controlling for maternal age, education, race, marital status, postpartum depression, stress and social support. • aORs and 95% CIs to measure the associations between CR-PTSD and maternal-infant bonding across the three time points (1, 6 and 12 months) were similar, indicating a persistent and stable association between CR-PTSD and maternal-infant bonding.	• Did not use a measure specifically designed to measure childbirth-related PTSD.

			classes, hospital tours, low-income clinics, private clinician offices, ultrasound centres, hospital intranet postings, newspaper adverts and targeted mailings. **Inclusion criteria for parents:** 18-35 years at recruitment, singleton pregnancy, planning to deliver in a hospital in Pennsylvania. **Exclusion criteria for parents:** prior pregnancy of 20	• 12 months postpartum (parent-child relationships). **Measures:** *Parent-child relationships:* PBQ – modified and shortened version. *Birth-related post-traumatic stress:* adapted version of the Trauma Screening Questionnaire (TSQ).		

			weeks gestation or longer, planning to deliver at home or in a birthing centre not associated with a hospital, delivering before 34 weeks gestation.			
14	Mayopoulos et al (2021)	Global – majority US (86%)	Women who gave birth in the last 6 months (average 2 months postpartum). n = 637 women who ***gave birth during COVID-19*** and 637 women who gave birth before COVID-19.	Cross-sectional online self-report questionnaire. **Measures:** *Parent-child relationships:* MIBS and the Maternal Attachment Inventory (MAI) *Birth-related post-traumatic stress:* Acute stress response to childbirth - Peritraumatic Distress Inventory (PDI)	• Women delivering during COVID-19 had significantly higher stress response to childbirth on PDI than matched controls p=.008. • Mediation: Acute stress to childbirth (PDI) significantly mediated the paths between study group and CB-PTSD and maternal bonding (initial bonding problems – MIBS and general bonding problems- MAI). • The COVID-19 group had a higher acute stress response, which was in turn associated with more CB-PTSD symptoms (p<.001) and more problems with maternal bonding (p<.001).	• Cross-sectional – cannot determine causality. • Self-report.

			Matched groups were based on similar demographic characteristics.	CB-PTSD – post-traumatic checklist for DSM-V (PCL-5)		
15	McDonald et al (2011)	United Kingdom	n = 79 women (from a previous study) Used data from a previous study – participants recruited from a postnatal ward in a hospital. **Inclusion criteria for parents:** over 16 years, married or cohabiting.	Longitudinal – postal questionnaires. • 6 weeks postpartum (PTS) • 3 months postpartum (PTS) • 2-years postpartum (PTS and parent-child relationships) **Measures:** *Parent-child relationships:* PSI-short form (parent-child dysfunctional interaction subscale) and mothers object relation scale – short form (MORS)	2 years postpartum: • PTSDQ total scores were moderately correlated with parent-child dysfunctional interactions (r=.37). • IES total scores were moderately correlated with PCDI (r-.41). • Small but significant correlations were found between the MORS-SF invasion scores and both PTSDQ total score (r=.30, p=.006) and IES total score (r=.23, p=.041). PTS symptoms at 6 weeks and 3 months: • PTSDQ at 3 months was significantly correlated with PCDI (r=.27). • IES at 3 months was significantly correlated with PCDI (r=.27). • PTSDQ at 6 weeks was significantly correlated with the invasion subscale of the MORS-SF at 2 years (r=.27). • Hierarchical multiple regression (controlling for scores on the hospital anxiety and depression scale): no significant predictions were found for PCDI or the MORS subscales.	• Self-report data.

			Exclusion criteria: if the baby was on the special care baby unit for over 24 hours, women were in situations of known domestic violence, women had insufficient English to complete the measures	*Birth-related post-traumatic stress:* post-traumatic stress disorder questionnaire (PTSDQ considering labour and birth) and IES		
16	Muller-Nix et al (2004)	Switzerland	45 mother-infant dyads of ***pre-term infants*** and 25 mother-infant dyads of full-term babies.	Longitudinal - data collected at 2 times points: • 6 months of corrected age (parent-child relationship) • 18 months of corrected age (PTS and parent-child relationship). **Measures:**	• PPQ – lower in mothers of high-risk pre-term children (and low-risk pre-term) than full-term babies. 6 months, according to maternal stress (full-term, high stress, low stress): • Maternal sensitivity – significantly lower in high stress than full-term (p=.03). • Maternal control – significantly higher in high stress dyads than full-term and low stress dyads (p<.001). • No significant differences in infant's interactional behaviour between the three groups. 18 months:	• Small sample • Did not control for possible confounders of other mental health difficulties

				Parent-child relationships: A mother-child play interaction (10 minutes) was videotaped and coded according to the third revision of the Care Index. *Birth-related post-traumatic stress:* PPQ.	• No significant differences in maternal interactional behaviour. • Infant's compliance-compulsivity – significantly higher in high stress dyads than full-term and low stress (p=.03). • Infant's passivity – higher in low stress than high stress and full-term dyads. Partial correlations (at 6 months): • After controlling for the PPQ: perinatal risk score (PERI) did not correlate significantly with any of the three maternal characteristics. • After controlling for the PERI: PPQ was close to the threshold of significance for sensitivity (p=0.09) and control (p=.06), suggesting the impact of the traumatic experience could be stronger than the infant's severity of risk.	
17	Nacik Rados et al (2020)	Croatia	n = 603 mothers (mean age = 31). Infants aged 1 to 12 months.	Cross-sectional online questionnaire. **Measures:** *Parent-child relationships:* PBQ.	• Of women who fulfilled the PTSD criteria, 39.4% had bonding difficulties. Younger infants (1-6 months): • Birth related PTSD symptoms had a moderate correlation with bonding difficulties (.30, p<.01). • General PTSD symptoms had a positive correlation with bonding difficulties (.50, p<.01). • Multigroup analysis: birth related PTSD symptoms did not have a significant direct	•

				Birth-related post-traumatic stress: translated and validated Croatian version of the CBTS.	effect on bonding (p=.34). General PTSD symptoms had a significant direct and indirect (via depressive symptoms) on bonding. Older infants (7-12 months): • Birth-related PTSD symptoms had a moderate correlation with bonding (.29, p<.01). • General PTSD symptoms has a positive correlation with bonding difficulties (.58, p<.01). • Multigroup analysis: birth related PTSD symptoms did not have a significant direct effect on bonding (p=.58). • General PTSD symptoms had a significant direct and indirect (via depressive symptoms) on bonding.	
18	Parfitt & Ayers (2009)	United Kingdom	126 women and 26 men (mean age = 32.58) recruited through a convenience sample. Final sample at 12 months = 62 mother-infant dyads of pre-term infants.	Cross- sectional - online questionnaire. **Measures:** *Parent-child relationships:* PBQ *Birth-related post-traumatic stress:* Post-traumatic Stress Diagnostic Scale – modified in relation to childbirth	Correlations: • PTSD and parent-baby bond: 0.36, p<.001. • Structural equation modelling: PTSD has a direct effect on the parent-baby bond but effect size was small (r = .20)	• Small sample for men. • Sample included a high proportion of people with obstetric intervention and PTSD or depression.

			Infants' age = 1 to 24 months old. **Inclusion criteria for parents:** over 18 years. Men must have attended the birth.			
19	Petit et al (2016)	France	Initial sample: 100 pre-term mother-infant dyads. Final sample at 12 months: 62 ***pre-term*** mother-infant dyads. **Exclusion criteria for parents:** evident psychiatric illness, drug abuse, age under 18.	Longitudinal – questionnaire and observational data. • V2: before hospital discharge (mean = 7.2 weeks after birth) (PPQ) • V3: 6 months postpartum (PPQ) • V4: 12 months postpartum (PPQ and PIPE) **Measures:** *Parent-child relationships:* Pediatric Infant Parent Exam (PIPE) to evaluate the mother-infant	• Correlations between PPQ and PIPE score at 12 months was only significant at V3 (6 months after birth - .034, p=0.008). • Correlations were not significant between PIPE score at 12 months and PPQ at discharge (.22, p=.10) and 12 months (.21, p=.11). • PPQ at 6 months was positively correlated with delivery conditions, anxio-depression state of the mother at V1 (inclusion), V2 and V3. PPQ score was highly correlated. It was also correlated with some of the baby's clinical characteristics (intrauterine growth restriction and perinatal risk inventory score at inclusion). • PPQ score at 12 months was correlated with delivery conditions and scores of depression/anxiety at all assessment times.	• Lack of control group (full-term dyads). • Sample fairly limited – only from 3 hospitals. • High attrition rate at 12 months – 38%.

			Exclusion criteria for infants: unfavourable vital prognosis evaluated with the perinatal risk inventory, malformation and/or generic anomaly diagnosed.	interaction during a short play session. *Birth-related post-traumatic stress:* modified PPQ – adapted to parents of perinatal high-risk children.		
20	Ponti et al (2020)	Italy	n = 103 women (mean age = 35.05) **Inclusion criteria for parents:** over 18 years, no previous psychopathological diagnosis, singleton and no risk pregnancy, no previous spontaneous or	Longitudinal - data collected at two time points: • During hospitalisation (postpartum stress – measured 2 days postpartum). • Three months after childbirth (parent-child relationship). **Measures:** *Parent-child relationships:* Maternal Postnatal Attachment Scale (MPAS).	• No significant differences in the PPQ and MPAS emerged in relation to the socio-demographical variables considered (work status, marital status, planned/unplanned, primiparous/multiparous pregnancy). Correlations: • Postnatal attachment (MPAS) was negatively correlated with the level of postpartum distress symptoms (PPQ): -.66, p<.001. Mediation: • High levels of postpartum distress symptoms had a significant positive effect on postpartum depression and significant and negative effects on postnatal attachment. The relationship between postpartum distress symptoms and postnatal attachment was directly and indirectly mediated by the level of postnatal depression (p<.001).	• Only recruited from the maternity ward of one hospital in Pisa.

			induced termination of pregnancy.	*Birth-related post-traumatic stress:* the Italian version of the Perinatal PTSD questionnaire (PPQ). **Other variables explored:** Edinburgh Postnatal Depression Scale		
21	Smorti et al (2021)	Italy	n = 105 women (M age = 34.97). **Inclusion criteria for parents:** native Italian women, above 18 years, no previous psychopathological diagnosis, singleton and no risk pregnancy.	Longitudinal for the wider study, however the measures of parent-child relationship and PTS were correlational (3 months after childbirth) **Measures:** *Parent-child relationships:* Maternal Postnatal Attachment Scale *Birth-related post-traumatic stress:* PPQ	• Postnatal attachment was negatively correlated with the childbirth as a traumatic event (-0.38, p<.01). • Mediation: prenatal attachment has a significant and positive effect on the level of postnatal attachment, both directly (p<.001) and indirectly through the childbirth experience as a traumatic event (the relationship between PPQ and MPAS was significant - p<.01)	• Cannot be applied to high-risk groups. • Only used self-report questionnaires.

				Other measures: prenatal attachment (week 31-32 gestation)		
22	Stuijfzand, Garthus-Neigel & Horsh (2020)	Switzerland	n = 210 mothers (mean age = 32.55) n = 91 fathers (mean age = 34.04). n = 216 children. **Inclusion criteria for parents:** partners must have been present at birth. **Exclusion criteria for parents:** mothers and partners of twins or multiple births.	Prospective cohort study – questionnaires. • 1 month postpartum (PTS) • 3 months postpartum (parent-child relationships) **Measures:** *Parent-child relationships:* MIBS – validated French version. *Birth-related post-traumatic stress:* post-traumatic diagnostic scale – French version related to childbirth (PSD-F). Potential confounders: Hospitalized Anxiety and Depression Scale, Medical	Correlations: • PTSD at 1 month significantly correlated with mother-infant bonding at 3 months (.21, p<.01) and father-infant bonding at 3 months (.27, p<.05). Structural equation models: • Mothers: Higher maternal birth-related PTSD symptoms at 1 month postpartum were prospectively associated with worse mother-infant bonding (p<.05). • Fathers: Birth-related PTSD symptoms were not prospectively associated with father-infant bonding. The only significant pathway in the model was between antenatal social support and bonding (p<.05). Cross associations for partners (n=77 couples): • No correlation found between maternal PTSD-CB symptoms and father infant bonding (r =-.03, p=.789). • There was a significant association between paternal PTSD-CB symptoms and mother-infant bonding (r=.27, p=.01).	• Self-report questionnaires

				Outcome Study Social Support Scale, Obstetric History, Birth-Related information.	Mediation (PTSD at 1 month and parent-infant relationships at 3 months adjusting for psychological distress at 1 month): • Mothers: PTSD-CB symptoms were no longer predictive of bonding when psychological distress included. • Fathers: neither PTSD-CB or psychological distress were predictive. Mediation via psychological distress for mothers: • No significant direct or indirect pathway was found. Mediation using PTSD subscales (PTSD at 1 months and parent-child relationships at 3 months via psychological distress at 1 month): • Mothers: A significant indirect effect was found between PTSD-CB and bonding via psychological distress for the intrusion and hyperarousal subscales. No significant direct effects were found.	
23	Suetsugu, Haruna & Kamibeppu (2020)	Japan	n = 130 mothers. **Inclusion criteria for parents:** women over 20 years old.	Longitudinal survey: • T1: 1-month postpartum • T4: 4 months postpartum **Measures:**	Bonding failure at T1: • IES-R (T1) – total score (.417, p<.001), impaired bonding (.465, p<.001), anxiety about care (.212, p<.01). • Rejection and anger/risk of abuse were non-significantly associated with the IES-R. Bonding failure at T2:	• Self-report measures.

			Inclusion criteria for infants: infants with a normal gestational period and a birth weight of 2500-4000g. **Exclusion criteria for parents:** women who had given birth to twins or had medical problems, or who had experienced a traumatic life event within 6 months.	*Parent-child relationships:* PBQ – Japanese version. *Birth-related post-traumatic stress:* IES-revised – Japanese version. Other outcomes assessed: depression	• IES-R at T2 was significantly associated with total score (.184, p<.05) but none of the subscales. • IES-R at T1 was not a significant predictor.	
24	Suttora et al (2021)	Italy	n = 64 mother-infant dyads: 32 ***pre-term infants***, 32 born full-term. Mother-infant dyads were recorded for 10-to-15-minute play sessions.	Cross-sectional – observation and self-report at 6 months.	• No significant differences in the measures due to birth condition (pre-term/full-term). Correlations: • No significant correlations between PPQ and the use of attuned (-.16, p = .195) and non-attuned (.03, p = .791) mind related comments.	• Cross-sectional – cannot determine causality. • Pre-term sample only from one neonatal intensive care unit.

			Mean age of mothers = 36.72 (pre-term), 34.48 (full term). **Exclusion criteria for infants:** presence of genetic abnormalities, severe neurofunctional impairment and/or neurosensory disabilities.	Mothers completed questionnaires following the play session. **Measures:** *Parent-child relationships:* maternal utterances transcribed and coded video sessions using the maternal mind-mindedness coding manual (mind-related, attuned or non-attuned). *Birth-related post-traumatic stress:* Perinatal PTSD Questionnaire (PPQ) – modified version.	Moderation: No significant results, indicating no significant interactions between the level of post-traumatic stress on maternal mind-mindedness.	

Appendix B STROBE Statement – checklist of items that should be included in reports of observational studies

	Item No	Recommendation
Title and abstract	1	(*a*) Indicate the study's design with a commonly used term in the title or the abstract
		(*b*) Provide in the abstract an informative and balanced summary of what was done and what was found
Introduction		
Background/rationale	2	Explain the scientific background and rationale for the investigation being reported
Objectives	3	State specific objectives, including any prespecified hypotheses
Methods		
Study design	4	Present key elements of study design early in the paper
Setting	5	Describe the setting, locations, and relevant dates, including periods of recruitment, exposure, follow-up, and data collection
Participants	6	(*a*) *Cohort study*—Give the eligibility criteria, and the sources and methods of selection of participants. Describe methods of follow-up *Case-control study*—Give the eligibility criteria, and the sources and methods of case ascertainment and control selection. Give the rationale for the choice of cases and controls *Cross-sectional study*—Give the eligibility criteria, and the sources and methods of selection of participants
		(*b*) *Cohort study*—For matched studies, give matching criteria and number of exposed and unexposed *Case-control study*—For matched studies, give matching criteria and the number of controls per case

Variables	7	Clearly define all outcomes, exposures, predictors, potential confounders, and effect modifiers. Give diagnostic criteria, if applicable
Data sources/ measurement	8*	For each variable of interest, give sources of data and details of methods of assessment (measurement). Describe comparability of assessment methods if there is more than one group
Bias	9	Describe any efforts to address potential sources of bias
Study size	10	Explain how the study size was arrived at
Quantitative variables	11	Explain how quantitative variables were handled in the analyses. If applicable, describe which groupings were chosen and why
Statistical methods	12	(*a*) Describe all statistical methods, including those used to control for confounding
		(*b*) Describe any methods used to examine subgroups and interactions
		(*c*) Explain how missing data were addressed
		(*d*) *Cohort study*—If applicable, explain how loss to follow-up was addressed *Case-control study*—If applicable, explain how matching of cases and controls was addressed *Cross-sectional study*—If applicable, describe analytical methods taking account of sampling strategy
		(*e*) Describe any sensitivity analyses

Appendix C Quality Assessment

Appendix C

Study	Title and Abstract (1)	Introduction (2, 3)	Methods (4-12)	Results (13-17)	Discussion (18-21)	Funding (22)
1)Ayers, Wright & Wells (2007)	1a) Yes 1b) Yes	2) Yes 3) Yes – direction of hypotheses not stated	4) Yes 5) Yes 6a) Yes – inclusion criteria 6b) n/a 7) Yes 8) Yes – reliability not reported 9) No 10) Yes 11) Yes 12a) Yes 12b) Yes – men/women 12c) No 12d) No 12e) No	13a) Yes 13b) Yes 13c) No 14a) No 14b) No 14c) n/a 15) No 16a) No 16b) n/a 16c) n/a 17) Yes – subgroups of PTS symptoms	18) Yes 19) Yes 20) Yes but limited reference to other research 21) Yes	22) Not stated
2)Camicasa et al (2017)	1a) No 1b) Yes	2) Yes 3) Yes - directions of hypotheses stated	4) Yes 5) Limited 6a) Yes	13a) Yes 13b) Yes – contacted those	18) Yes 19) Yes 20) Yes	22) Not stated

			6b) n/a 7) Yes 8) Yes – reliability reported 9) Yes – use of a second coder 10) Yes – consented from a previous study 11) Yes 12a) Yes 12b) Yes 12c) No 12d) Limited 12e) No	who agreed from previous study 13c) No 14a) Yes 14b) No 14c) Yes 15) Yes 16a) Yes 16b) n/a 16c) n/a 17) Yes – mediation	21) Yes	
3)Davies et al (2008)	1a) Limited 1b) Yes	2) Yes 3) Partially – aims stated but no hypotheses	4) Yes 5) Yes 6a) Yes – inclusion criteria 6b) n/a 7) Yes 8) Yes – reliability reported	13a) Yes 13b) n/a 13c) No 14a) Yes 14b) No 14c) n/a 15) Yes	18) Yes 19) Yes 20) Yes 21) Yes	22) Not stated

			9) Yes – measured previous traumatic experiences 10) Yes 11) Partially 12a) Yes 12b) Yes 12c) No 12d) No 12e) No	16a) Yes – depression 16b) Yes – fully/partially/non-symptomatic 16c) n/a 17) Yes		
4)Dekel et al (2018)	1a) No 1b) Yes	2) Yes 3) Yes – hypotheses stated	4) Limited 5) Yes 6a) Yes 6b) n/a 7) Yes 8) Yes – reliability reported 9) Yes – potential confounders considered 10) No 11) Yes 12a) Yes	13a) No 13b) n/a 13c) No 14a) Yes 14b) No – only overall amount missing 14c) n/a 15) No 16a) Yes	18) Yes 19) Yes 20) Yes but limited reference to other research 21) No	22) Not stated

			12b) Yes 12c) Yes 12d) No 12e) No	16b) Yes – for PP-PTSD 16c) n/a 17) Yes		
5)Ertan et al (2021)	1a) Yes 1b) Yes	2) Yes 3) Yes, although direction of hypotheses not described	4) Yes 5) Yes 6a) Yes – clear exclusion criteria b) n/a 7) Yes – potential confounders are investigated 8) Yes – reliability of the measures included 9) Yes – any incomplete questionnaires were removed 10) No 11) Yes 12a) Yes 12b) Yes	13a) Yes 13b) n/a 13c) Not used 14a) Yes – reported and analysed 14b) n/a – responses with missing answers were eliminated 14c) n/a 15) Yes 16a) Yes 16b) n/a 16c) n/a 17) Yes	18) Yes 19) Yes e.g., validity of the French version of the CBTS 20) Yes – linked to previous research 21) Yes, this is mentioned	22) Yes – no funding

			12c) Yes – those who did not fully complete the questionnaires were not included 12d) No 12e) No			
6)Feeley et al	1a) Partially 1b) Yes	2) Yes 3) Yes – pilot study, direction of hypotheses not stated	4) Yes 5) Yes 6a) Yes – inclusion criteria 6b) n/a 7) Yes 8) Yes – reliability reported 9) Partially – two scorers for questionnaire but not observation 10) No 11) Yes 12a) Yes 12b) n/a – no subgroups	13a) No 13b) n/a 13c) No 14a) Yes 14b) No 14c) n/a 15) No 16a) No 16b) n/a 16c) n/a 17) No	18) Yes 19) Yes but limited 20) Limited 21) No	22) Yes

			12c) No 12d) No 12e) No			
7)Forcada-Geux et al (2011)	1a) No 1b) Yes	2) Yes 3) Yes – hypotheses stated	4) Yes 5) Yes 6a) Yes – clear exclusion criteria 6b) Partially - recruited from same hospital, demographic variables compared but not matched 7) Yes 8) Yes – reliability not reported 9) Yes – two coders for interviews 10) Partially 11) Yes 12a) Yes 12b) Yes – high/low PTS/full-term	13a) Yes 13b) Yes 13c) No 14a) Yes 14b) Yes – participants with missing data removed 14c) Yes 15) Yes 16a) No 16b) Yes – high/low PTS 16c) n/a 17) Yes	18) Yes 19) Yes 20) Yes 21) Yes/partially	22) Not stated

			12c) Yes - participants with missing data removed, no missing data for control group 12d) No 12e) No			
8)Hairston et al (2018)	1a) No 1b) Yes	2) Yes 3) Yes – hypotheses stated	4) Yes 5) Yes 6a) Yes – clear exclusion criteria 6b) n/a 7) Yes 8) – Yes – reliability reported 9) Yes – demographic variables considered 10) Yes 11) Yes 12a) Yes 12b) Yes – mediation 12c) Yes – removed if large gaps	13a) Yes 13b) n/a – cross-sectional 13c) No 14a) Yes 14b) Yes 14c) n/a 15) Yes 16a) Yes 16b) n/a 16c) n/a 17) Yes - mediation	18) Yes 19) Yes 20) Yes 21) Yes	22) Yes – Academic College of Tel Aviv-Yafo

			12d) No 12e) No			
9)Handelzalts et al (2019)	1a) Yes 1b) Yes	2) Yes 3) Yes – hypotheses stated	4) Yes 5) Yes 6a) Yes 6b) n/a 7)Yes 8) Yes 9) Yes – clear inclusion criteria, controlled for demographic variables that correlated with bonding 10) No 11) Yes 12a) Yes – demographic variables considered 12b) Yes – mediation 12c) Not stated 12d) No	13a) No 13b) n/a 13) No 14a) Yes 14b) No 14c) n/a 15) Yes 16a) Yes – significant confounders included in mediation 16b) n/a 16c) n/a 17) Yes – interactions (mediation)	18) Yes 19) Yes 20) Yes 21) Yes	22) Not stated

			12e) No			
10)Handelzalts et al (2021)	1a) Yes 1b) Yes	2) Yes 3) Yes	4) Yes 5) Yes 6a) Yes 6b) n/a 7) Yes 8) Yes – reliability of measures reported 9) Yes – assessed dropout characteristics, demographic characteristics that had a significant or close to significant relationship with bonding were considered as covariates. 10) Yes 11) Yes 12a) Yes 12b) Yes 12c) Yes	13a) Yes 13b) Limited 13c) Yes 14a) Yes 14b) No – details not specified 14c) Yes 15) Yes 16a) Yes 16b) n/a 16c) *** 17) Yes	18) Yes 19) Yes 20) Yes 21) Yes	22) Yes – not funded

			12d) Yes 12e) No			
11)Ionio & Di Blasio (2014)	1a) Limited 1b) Yes	2) Yes 3) Yes – directions of hypotheses stated	4) Yes 5) Yes 6a) Yes – clear exclusion criteria 6b) n/a 7) Yes 8) Yes – no reliability reported 9) Yes – compared variables of total sample to final sample, two independent coders for observation 10) Yes 11) Yes 12a) Yes 12b) Yes 12c) No 12d) Yes – compared total sample to final sample	13a) Yes 13b) Partially 13c) No 14a) Yes 14b) No 14c) Yes 15) Yes 16a) Yes 16b) n/a 16c) n/a 17) Yes	18) Yes 19) Yes 20) Yes – some reference to other research 21) Yes	22) Not stated

			12e) No			
12)Ionio et al (2017)	1a) No 1b) Yes	2) Yes 3) Yes	4) Yes 5) Yes 6a) Yes 6b) Yes – recruited from same hospital 7) Yes 8) Yes 9) Yes – coded by two researchers who were blind to group 10) Yes – number from the hospital that agreed 11) Yes 12a) Yes – groups compared on demographic characteristics 12b) Yes 12c) Not stated 12d) No	13a) Yes for recruitment, not stated how many dropped out during second part 13b) Yes (at recruitment) 13c) No 14a) Yes 14b) No 14c) Yes 15) No 16a) No 16b) n/a 16c) n/a 17) Yes – group comparisons	18) Yes 19) Yes 20) Yes 21) Yes	22) Not stated

			12e) No			
13)Kjerulff et al (2021)	1a) Yes 1b) Yes	2) Yes 3) Yes – direction of hypotheses not stated	4) Yes 5) Yes 6a) Yes – clear inclusion and exclusion criteria 6b) n/a 7) Yes 8) Yes 9) Yes – demographic variables 10) Partially 11) Yes 12a) Yes 12b) Yes 12c) No 12d) No 12e) No	13a) Partially 13b) No 13c) No 14a) Yes 14b) No 14c) Yes 15) Yes 16a) Yes 16b) Yes 16c) n/a 17) Yes	18) Yes 19) Yes 20) Yes 21) Yes	22) Yes – National Institute of Health

14)Mayopoulos et al (2021)	1a) Yes 1b) Yes	2) Yes 3) Yes – in introduction	4) Yes 5) Yes 6a) Yes 6b) Yes 7) Yes 8) Yes – reliability of measures reported 9) Yes – efforts taken to ensure groups were matched 10) Yes 11) Yes 12a) Yes 12b) Yes 12c) Yes – the FIML procedure 12d) Yes 12e) No	13a) Yes 13b) n/a 13c) No 14a) Yes 14b) No 14c) n/a 15) Yes 16a) Yes – CIs included 16b) n/a 16c) n/a 17) Yes	18) Yes 19) Yes – briefly 20) Limited reference to previous research 21) Limited	22) Yes
15)McDonald et al (2011)	1a) Limited 1b) Yes	2) Yes	4) Yes but limited 5) Limited	13a) Yes 13b) Yes	18) Yes 19) Yes, but limited	22) Not stated.

		3) Yes – clear aims and hypotheses	6a) No inclusion/exclusion criteria stated 6b) n/a 7) Yes 8) Yes – no reliability reported 9) Yes – demographic variables considered 10) Yes 11) Yes 12a) Yes 12b) Yes 12c) Yes – incomplete data removed (2) 12d) No 12e) No	13c) Yes 14a) Yes 14b) No 14c) Yes 15) Yes 16a) Yes 16b) n/a 16c) n/a 17) Yes	20) Yes 21) No	
16)Muller-Nix et al (2014)	1a) Limited 1b) Yes	2) Yes 3) Yes – hypotheses stated	4) Yes 5) Yes 6a) Yes – clear inclusion criteria	13a) Yes 13b) Yes 13c) No 14a) Yes	18) Yes 19) Yes but limited 20) Yes 21) No	22) Not stated

			6b) Yes – recruited from same hospital 7) Yes 8) Yes – reliability not reported 9) Yes – two coders for observations 10) Yes 11) Yes 12a) Yes 12b) Yes 12c) Yes – incomplete data removed 12d) No 12e) No	14b) Yes 14c) Yes 15) Yes 16a) Yes 16b) Yes – low/high stress 16c) n/a 17) Yes		
17)Nakic Rados et al (2020)	1a) Yes 1b) Yes	2) Yes 3) Yes – hypotheses stated	4) In abstract 5) Yes 6a) No eligibility criteria stated 6b) n/a	13a) Yes 13b) n/a 13c) No 14a) Yes 14b) No	18) Yes 19) Yes – to a limited extent 20) Yes 21) No	22) Yes – University of Croatia

			7) Yes 8) Yes – reliability reported 9) No 10) No 11) Yes 12a) Yes 12b) Yes – younger/older infants considered separately 12c) No 12d) No 12e) No	14c) n/a 15) Yes 16a) Yes 16b) Yes – age of infant 16c) n/a 17) Yes – older/younger infants		
18)Parfitt & Ayers (2009)	1a) Yes 1b) Yes	2) Yes 3) Yes – hypotheses stated	4) Yes 5) Yes 6a) Yes 6b) n/a 7) Yes 8) Yes – reliability reported 9) Yes – depression considered	13a) No 13b) n/a 13c) No 14a) Yes 14b) No 14c) n/a 15) Yes 16a) Yes	18) Yes 19) Yes 20) Yes 21) Yes	22) Not stated

			10) No 11) Yes 12a) Yes 12b) Yes 12c) No 12d) No 12e) No	16b) Yes – PTSD/no PTSD 16c) n/a 17) Yes		
19)Petit et al (2016)	1a) No 1b) Yes	2) Yes 3) Yes	4) Yes 5) Reported elsewhere 6a) Yes – clear exclusion criteria 6b) n/a 7) Yes 8) Yes – no reliability of scales reported 9) No 10) Yes – flow diagram 11) Yes 12a) Yes 12b) Yes	13a) Yes 13b) Yes 13c) Yes 14a) Yes 14b) No 14c) Yes 15) Yes 16a) Yes 16b) n/a 16c) n/a 17) Yes	18) Yes 19) Yes 20) Yes 21) No	22) Yes – Hospital Research Program from French Ministry of Health

			12c) No 12d) Yes – compared characteristics (no statistically significant differences) 12e) No			
20)Ponti et al (2020)	1a) Yes 1b) Yes	2) Yes 3) Yes – hypotheses stated	4) Yes 5) Yes 6a) Yes 6b) n/a 7) Yes 8) Yes – reliability reported 9) Yes – clear inclusion criteria removing potential confounders 10) No 11) Yes 12a) Yes – demographic variables considered 12b) Yes	13a) No 13b) No 13c) No 14a) Yes 14b) No 14c) Yes 15) No 16a) No 16b) n/a 16c) n/a 17) Yes – interactions (mediation)	18) Yes 19) Yes 20) Yes 21) Yes	22) Funding not reported but no conflict of interest noted

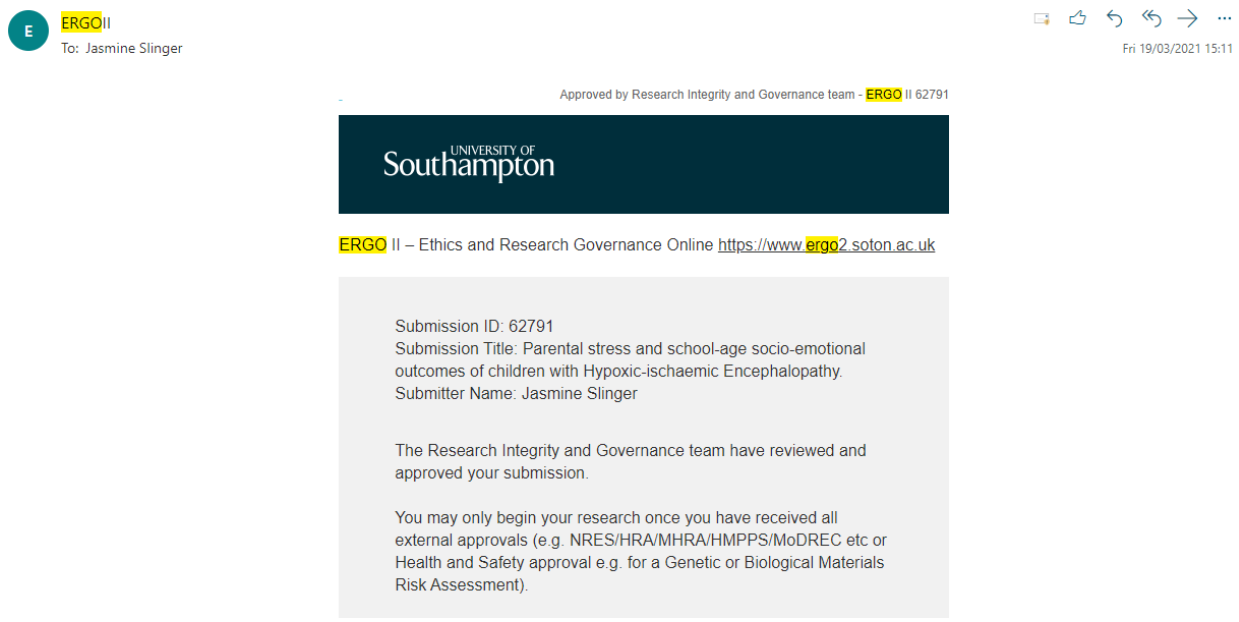
			12c) Missing data not reported 12d) Not reported 12e) No			
21)Smorti et al	1a) Yes 1b) Yes	2) Yes 3) Yes – hypotheses stated	4) Yes 5) Yes 6a) Yes – inclusion criteria 6b) n/a 7) Yes 8) Yes – reliability reported 9) Yes – demographic variables and potential confounder (prenatal attachment) included 10) Yes 11) Yes 12a) Yes 12b) Yes - mediation 12c) Yes	13a) Yes 13b) Yes – no reasons provided 13c) No 14a) Yes 14b) No 14c) Yes 15) No 16a) No 16b) n/a 16c) n/a 17) Yes	18) Yes 19) Yes 20) Yes 21) Limited	22) Yes – no funding

			12d) Yes 12e) No			
22)Stuijfzand et al	1a) Yes 1b) Yes	2) Yes 3) Yes – direction of hypotheses stated	4) Yes 5) Yes 6a) Yes – inclusion criteria 6b) n/a 7) Yes 8) Yes – reliability reported 9) Yes – potential confounders measured 10) Yes 11) Yes 12a) Yes 12b) Yes 12c) Yes – using full information maximum likelihood. 12d) Yes – as above 12e) No	13a) Yes 13b) Yes 13c) Yes 14a) Yes, however sociodemographic not reported 14b) No 14c) No 15) No - only for demographic data 16a) Yes 16b) n/a 16c) n/a 17) Yes	18) Yes 19) Yes 20) Yes 21) Yes	22) Yes – no funding
23)Suetsugu et al	1a) Yes	2) Yes	4) Yes	13a) Yes	18) Yes	22) Yes

	1b) Yes	3) Yes	5) Yes 6a) Yes – clear inclusion criteria 6b) n/a 7) Yes 8) Yes – reliability reported 9) Yes – demographic and obstetric variables measured 10) Yes 11) Yes 12a) Yes 12b) Yes 12c) Yes – incomplete data removed 12d) No 12e) No	13b) No 13c) No 14a) Yes 14b) Yes – incomplete data removed (6) 14c) Yes 15) Yes 16a) No 16b) Yes 16c) n/a 17) Yes	19) Yes 20) Yes – limited reference to other research 21) Yes	
24)Suttora et al (2021)	1a) Yes 1b) Yes	2) Yes 3) Yes – hypotheses stated	4) Yes 5) Yes 6a) Yes 6b) Yes	13a) Yes 13b) n/a 13c) No 14a) Yes	18) Yes 19) Yes 20) Yes 21) Yes - limited	22) No

			7) Yes 8) Yes – reliability reported 9) Yes – use of a second coder 10) No 11) Yes 12a) Yes 12b) Yes 12c) No 12d) No – differences between the groups were noted 12e) No	14b) Yes – only hospitalisation data missing for one participant 14c) n/a 15) Yes 16a) Yes – CIs reported 16b) n/a 16c) n/a 17) Yes		

Appendix D Ethics approval



E ERGOII
To: Jasmine Slinger

Fri 19/03/2021 15:11

Approved by Research Integrity and Governance team - ERGO II 62791

UNIVERSITY OF Southampton

ERGO II – Ethics and Research Governance Online https://www.ergo2.soton.ac.uk

Submission ID: 62791
Submission Title: Parental stress and school-age socio-emotional outcomes of children with Hypoxic-ischaemic Encephalopathy.
Submitter Name: Jasmine Slinger

The Research Integrity and Governance team have reviewed and approved your submission.

You may only begin your research once you have received all external approvals (e.g. NRES/HRA/MHRA/HMPPS/MoDREC etc or Health and Safety approval e.g. for a Genetic or Biological Materials Risk Assessment).

Appendix E Invitation letter to parents of children with HIE

Study Title: Neurodevelopmental trajectories and neural correlates in children with Neonatal Hypoxic-Ischaemic Encephalopathy (HIE)

Dear Parent/Guardian

We are writing to you to ask you if you and <name of child> would like to participate in a study that we are currently conducting at Southampton University Hospitals NHS Trust / University of Southampton.

We want to investigate the long term outcome of children who received hypothermia treatment ("brain cooling") after being starved of oxygen around the time of birth.

We have enclosed an information leaflet about our research to help you decide if you want to take part in the study. If you would like to participate in our research, we will arrange a date for you and <name of child> to come to the hospital and take part in the research study.

If you would like to discuss this study further or have any questions regarding the questionnaires and how this might apply to your child, please find our contact details in the information leaflet.

We would like to thank you in advance for your interest in this study.

Yours sincerely,

Dr. Brigitte Vollmer

Associate Professor in Paediatric Neurology

Consultant in Paediatric Neurology

Appendix F Poster advert for control participants

WOULD YOU LIKE TO BE PART OF AN EXCITING NEW RESEARCH STUDY CALLED NENAH

At the University of Southampton and Southampton Children's Hospital, we are conducting a study looking at the experiences of children who were poorly after they were born, and received a therapy called "brain cooling".

supported by action medical research for children
Charity reg. nos 208701 and SC039284

We would like to study how these children are doing at school and home.

We also would like to see how their brain has developed after the difficult time they had as newborns. For this, we will take pictures of the brain using an "MRI scanner".

We are looking for children aged 6 to 8 years old who did not have any problems when they were newborns. We want to see if there are any differences with learning, and how they feel about things compared to the children who had 'brain cooling'. We would love to hear from you if you want to

take part in this research, or if you simply want to learn more about this research. You can email us, telephone us, and you can also get some information by looking at this webpage

https://action.org.uk/research/birth-asphyxia-predicting-long-term-effects,

AND

If you would like more information about taking part in the study, we would love to hear from you

You can email us at HTstudy@soton.ac.uk OR contact us via telephone 023 8120 3036

Scan to see information about this study and get in touch with us.

This study has been reviewed by the Liverpool and North Central Ethics Committee.

(IRAS ID: 263965; REC Ref N: 19/NW/0478)

Appendix G Letter for schools

Study Title: Neurodevelopmental trajectories and neural correlates in children with Neonatal Hypoxic-Ischaemic Encephalopathy (HIE) - NENAH

Dear <Name of School>,

We would like to ask you if you would support us with a research study we are currently conducting at Southampton University Hospitals NHS Trust / University of Southampton.

We want to investigate the long term outcome of children who received hypothermia treatment ("brain cooling") after being starved of oxygen around the time of birth. In particular, we want to examine whether children who received brain cooling differ from typically developing children on measures of general health, and on thinking and behavioural measures that are important for school readiness and school success.

This therapy has now become standard care in the majority of neonatal centres, as it has been shown to reduce severe disability and mortality at toddler age. However, there is very little long-term outcome data beyond toddler age, and the available data are partly inconclusive.

The aim of the study is to improve understanding of school-age outcomes in children who underwent hypothermia treatment and survived without major neurodevelopmental disabilities. Specifically, we aim to investigate behavioural and cognitive outcomes compare to typically developing children at school-age. In addition, we would like to explore how brain MRI imagining structure compare to typically developing children and how neonatal MRI and early neurodevelopmental assessment predict cognitive and behavioural outcomes at school age.

The proposed project aims to address some important gaps in the existing research on long-term effects of neonatal brain cooling. As well as providing important information on long term outcomes that are important for everyday functioning and school success, it will raise awareness of behavioural and educational difficulties that a large proportion of these children are likely to encounter.

As part of this study, we would like to recruit a group of typically developing children to act as control group. We would like to ask your assistance in recruiting healthy children. If you are happy

to assist us with recruitment, we would like to ask your support by advertising the project in the school newsletter and within the school office with the attached poster.

If you would like to know more about the project or if you have any questions please do not hesitate to contact us and we will be very pleased to speak to you about the study.

We would like to thank you in advance for your support.

Yours sincerely,

Dr Brigitte Vollmer – Principal Investigator – b.vollmer@soton.ac.uk

Dr Rina Cianfaglione – Psychology Research Fellow – R.Cianfaglione@soton.ac.uk

Appendix H Invitation letter for parents of control children

Study Title: **Neurodevelopmental trajectories and neural correlates in children with Neonatal Hypoxic-Ischaemic Encephalopathy (HIE) - NENAH**

Dear Parent/Guardian

We would like to ask if you and your child would like to take part in a research study.

What is the purpose of the study?

We are investigating the long-term outcomes and experiences of children who received a brain cooling treatment after they were born because they had been starved of oxygen around the time of their birth.

Why is my child being asked?

Your child did not have brain cooling treatment, but we want to compare children who were born without experiencing any difficulties, and who are developing normally with the children who had brain cooling to see if there are any differences in learning. We will assess the children who had the brain cooling treatment and the "comparison" group of children who did not experience difficulties at birth, using the same tests.

What does it involve?

We will spend three to four hours assessing your child at a time suitable for you, but will spread the assessment over a few days. Depending on your and your child's preference, the assessment would be done either at home or at Southampton General Hospital. During these sessions a range of your child's abilities will be assessed such as their general intellectual ability, academic performance, memory and problem solving skills. They will also complete a neurological and motor development assessment which will assess their ability to move around.

We will also ask you to come to Southampton General Hospital for a brain MRI scan. The scan will take about 30 minutes and the appointment will approximately take 1 hours.

If you would like, when we have the results of the assessment we will send you a report that will tell you how your child has performed. This is optional, and you can choose not to have the report.

What now?

Please complete the slip below to let us know if you are interested in hearing a bit more about the study. Please return the slip to us in the prepaid envelope even if you don't want to take part, so we know not to contact you again in the future. .

If you are interested, we will send you a more detailed information sheet and one of us will phone you to discuss the study with you and answer any questions that you might have. You can then decide if you would like your child to participate or not.

Thank you for taking the time to read this and consider your child's participation.

Yours sincerely,

Dr Brigitte Vollmer

Principal Investigator

Dr Rina Cianfaglione

Psychology Research Fellow

Appendix I Information sheet for parents of HIE children

Parent/Guardian Information Sheet – Children with history of neonatal HIE

Study Title: **Neurodevelopmental trajectories and neural correlates in children with Neonatal Hypoxic-Ischaemic Encephalopathy (HIE) - NENAH**

You and your child are being invited to take part in a research study. Before you decide to take part in the study it is important to understand why the research is being done and what it will involve for you and your child. Please take time to read the following information and don't hesitate to ask us if there is anything you are unsure of or would like more information about.

What is the purpose of the study?

We want to investigate the long term outcome of children who received "brain cooling" (hypothermia treatment) after being starved of oxygen around the time of birth.

Babies who are born under such difficult conditions are at high risk for brain injury and long term problems. Brain cooling" has been shown to reduce severe disability at toddler age but there is very little information on how the affected children do at school age. We want to examine whether "brain cooled" children differ from children who were not starved of oxygen as a baby and did not need "brain cooling" in measures of general health, thinking and behaviour, all of which are important for school success and relationship with peers.

Why has my child been chosen?

Your child underwent brain cooling as a baby at Princess Anne Hospital in Southampton and is now 6 to 8 years old.

What will happen to my child if they take part?

If you decide to take part, we will invite you and your child to attend 2 sessions at Southampton General Hospital (SGH). During the first session your child's ability will be assessed on a range

of measures including their general intellectual ability, academic performance (reading, language and mathematics ability), problem solving, and motor skills. This session will be held at the Wellcome Research Centre Facilities at SGH. However, if you prefer or if travelling to SGH is difficult for you, this can also be arranged either at your child's school or your home.

These assessments will take about four hours in total, but they can be split in two or three sessions if you prefer, and your child can take regular breaks. We may ask you if we can video record some of the assessments with your child, so another researcher can make sure the assessments were carried out and scored correctly. We need to do this for about 10% of the children that participate in our study and we will randomly select children from those that agreed to be filmed. Of course, you can let us know if you are not happy for your child's assessment to be recorded and we will then not record the assessment. If you agree for the assessment to be videotaped, we will store the video in a folder protected by a secure password on the University of Southampton password protected server. The video will only be accessed and watched by the research team for scoring purposes. Nobody else has access to the video. The video will be deleted immediately when the study is finished (end of February 2022).

We will also invite you and your child to a second session at SGH. At this session, we would like to look at how your child's brain has developed since birth. The way we will do this is by using a Magnetic Resonance Imaging (MRI) camera to do a MRI brain scan. The MRI will be done in a playful setting and we will invite you and your child for a visit to get familiar with the MRI camera and ask any questions you or your child may have, before we arrange for the visit to do the MRI. The MRI will be completed while your child is awake. Your child will be able to listen to music or watch a movie during the scan.

When you attend a session at Southampton General Hospital, we will reimburse any travel expenses you may encounter, and we will provide a lunch voucher to use in the hospital restaurant.

To compensate for your and your child's time we will offer a £10 voucher at the end of both sessions.

We would like your permission to use some of the findings from the routine 2-year neurodevelopmental assessment as well as findings from brain imaging from the time when your child was on the neonatal unit so that we can relate this information to the findings at 6 - 8 years of age.

Do I have to take part?

It is up to you to decide whether or not you want to include your child in this study. Involving your child in this research study is entirely voluntary, and you and your child are free to withdraw at any time and without giving a reason. A decision to withdraw at any time, or a decision not to take part, will not affect the standard of care your child receives.

What do I have to do?

If you decide to take part, you will be asked to sign a consent form (a copy of which you will keep). We will arrange a date for you and your child to come to the hospital and take part in the research study.

We will ask you to fill in some questionnaires about your child's behaviour and whether their health has impacted your or your family's life since they were discharged. You will be able to complete these questionnaires while your child is taking part in the study assessments.

With your permission, we will ask your child's teacher to fill in some of these questionnaires too.

What are the possible benefits of taking part?

There is no direct benefit to your child from taking part in this study. However, the study will provide essential information about any difficulties in thinking and problem solving that children who received 'brain cooling' might experience when they are at school. We hope that this research will eventually improve information and support for parents and also alert teachers to potential difficulties that these children may experience at school.

What will happen after the assessments and brain scan have been completed?

Once all assessments have been completed, we will also ask you if you would like a report about your child's performance. We will not send you a report if you don't want it; however, If any specific problems are detected for your child as part of this study, this will be first discussed with you, and then a referrals to any relevant specialists will be made by the research team within 2 weeks from the completion of the assessments.

What if something goes wrong?

Although we think it is very unlikely that anything will go wrong, if your child is harmed by taking part in this research study, there are no special compensation arrangements. If your child is harmed through someone's negligence, then you will have grounds for legal action, but you may have to pay for it.

Regardless of this, if you wish to complain, or have any concerns about any aspect of the way you have been approached or your child treated during the course of this study, the normal National Health Service complaints mechanisms should be available to you (Patient Advice and Liaison Service).

PALS

C Level Centre Block

Mailpoint 81

Southampton General Hospital

Tremona Road

Email: PALS@suht.swest.nhs.uk

Tel: 023 8079 8498

Will my taking part in this study be kept confidential?

All information collected about you and your child during the course of the research will be kept strictly confidential. Any study information about your child which leaves the hospital will have their name and address removed so they cannot be recognised from it.

With your consent, your GP will be notified of your child's participation in the study and any relevant medical information obtained about your child as part of the study. We will also ask the GP surgery if they could advertise our study by displaying a study information poster in the surgery so we can recruit children who did not have cooling treatment as a baby to act as control.

What will happen to the results of the research study?

The results of the study, once analysed, will be published in appropriate professional journals, shared with medical and nursing professionals locally, nationally and internationally in reports, at conferences and through network groups.

Also, at the end of the study, we will organise an event for families of children who have been treated at Princess Anne Hospital and non-medical professionals who work with the children (e.g. teachers, educational psychologists, community paediatricians), at which we will feed back the study findings. All data will be anonymous -neither you or your child will not be identifiable.

A summary of the research findings can be sent to you, upon your request.

Who is organising and funding the research?
This study is funded by Action Medical Research. It has been approved by the <xxx> Research Ethics Committee and the local research governance committee.

If you would like more information on the study you can check out this link:
https://action.org.uk/research/birth-asphyxia-predicting-long-term-effects

University Hospital of Southampton (UHS) is the sponsor for this study based in the United Kingdom. We will be using information from from your child and/or your child's medical records in order to undertake this study and will act as the data controller for this study. This means that we are responsible for looking after your information and using it properly. University Hospital of Southampton will keep identifiable information about you for 30 years after the study has finished.

Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that we have already obtained. To safeguard your rights, we will use the minimum personally-identifiable information possible.

You can find out more about how we use your information
https://www.hra.nhs.uk/information-about-patients/.

The research team will collect information from your child's medical records for this research study in accordance with our instructions.

The research team will keep your and your child's name your child's NHS number and contact details confidential.. The research team will use this information as needed, to contact you about the research study, and make sure that relevant information about the study is recorded for your care, and to oversee the quality of the study. Certain

individuals from UHS and regulatory organisations may look at your medical and research records to check the accuracy of the research study. UHS will only receive information without any identifying information. The people who analyse the information will not be able to identify you and will not be able to find out you and your child's name, NHS number or contact details.

The research team will keep identifiable information about you from this study for 30 years after the study has finished.

When you agree to take part in a research study, the information about your child's health and care may be provided to researchers running other research studies in this organisation and in other organisations. These organisations may be universities, NHS organisations or companies involved in health and care research in this country or abroad. Your child's information will only be used by organisations and researchers to conduct research in accordance with the UK Policy Framework for Health and Social Care Research.

This information will not identify you or your child's and will not be combined with other information in a way that could identify you or your child. The information will only be used for the purpose of health and care research, and cannot be used to contact you or to affect your care or that of your child. It will not be used to make decisions about future services available to your child, such as insurance.

Contact for further information

- Study email address - HTstudy@soton.ac.uk

- Dr Brigitte Vollmer – Principal Investigator

- Dr Rina Cianfaglione– Psychology Research Fellow

Tel: 023 8120 3036

Appendix J Information sheet for parents of control children

Parent/Guardian Information Sheet – Children without history of neonatal HIE

Study Title: **Neurodevelopmental trajectories and neural correlates in children with Neonatal Hypoxic-Ischaemic Encephalopathy (HIE) - NENAH**

You told us that you would be interested in you and your child taking part in our research study. Before you decide to take part in the study it is important to understand why the research is being done and what it will involve for you and your child. Please take time to read the following information and don't hesitate to ask us if there is anything you are unsure of or would like more information about. We will telephone you shortly to discuss any questions that you might have about the research study.

What is the purpose of the study?

We want to investigate the long term outcome of children who received "brain cooling" (hypothermia treatment) after being starved of oxygen around the time of birth.

Babies who are born under such difficult conditions are at high risk for brain injury and long term problems. "Brain cooling" has been shown to reduce severe disability at toddler age but there is very little information on how the affected children do at school age . We want to examine whether "brain cooled" children differ from children who were not starved oxygen as a baby and did not need "brain cooling" in measures of general health, thinking and behaviour, all of which are important for school success and relationship with peers.

Why has my child been chosen?

Your child is 6 to 8 years old and did not undergo "brain cooling" as a baby. We want to compare children who were born without experiencing any difficulties with children who had 'brain cooling' to see if there are any differences.

Please note that if your child has any relevant medical condition that could affect their neurodevelopment and/or behaviour he/she is not eligible to take part in this study. In addition, we can only include children that are born around their due date (above 37 weeks of gestation).

We are happy to be contacted and talk to you about the study if you are not sure whether your child can be included in the study.

What will happen to my child if they take part?

If you decide to take part, we will invite you and your child to attend 2 sessions at Southampton General Hospital (SGH). During the first session your child's ability will be assessed on a range of measures including their general intellectual ability, academic performance (reading, language and mathematics ability), problem solving, and motor skills. This session will be held at the Wellcome Research Centre Facilities at SGH. However, if you prefer or if travelling to SGH is difficult for you, this can also be arranged either at your child's school or your home.

These assessments will take about four hours in total, but they can be split in two or three sessions if you prefer, and your child can take regular breaks. We may ask you if we can video record some of the assessments with your child, so another researcher can make sure the assessments were carried out and scored correctly. We need to do this for about 10% of the children that participate in our study and we will randomly select children from those that agreed to be filmed. Of course, you can let us know if you are not happy for your child's assessment to be recorded and we will then not record the assessment. If you agree for the assessment to be videotaped, we will store the video in a folder protected by a secure password on the University of Southampton password protected server. The video will only be accessed and watched by the research team for scoring purposes. Nobody else has access to the video. The video will be deleted immediately when the study is finished (end of February 2022).

We will also invite you and your child to a second session at SGH. At this session, we would like to look at how your child's brain has developed since birth. The way we will do this is by using a Magnetic Resonance Imaging (MRI) camera to do a MRI brain scan. The MRI will be done in a playful setting and we will invite you and your child for a visit to get familiar with the MRI camera and ask any questions you or your child may have, before we arrange for the visit to do the MRI. The MRI will be completed while your child is awake. Your child will be able to listen to music or watch a movie during the scan.

When you attend a session at Southampton General Hospital, we will reimburse any travel expenses you may encounter, and we will provide a lunch voucher to use in the hospital restaurant.

To compensate for your and your child's time we will offer a £10 voucher at the end of both sessions.

Do I have to take part?

It is up to you to decide whether or not you want to include your child in this study. Involving your child in this research study is entirely voluntary, and you and your child are free to withdraw at any time and without giving a reason. A decision to withdraw at any time, or a decision not to take part, will not affect the standard of care your child receives.

What do I have to do?

If you decide to take part, you will be asked to sign a consent form (a copy of which you will keep). We will arrange a date for you and your child to come to take part in the research study.

We will ask you to fill in some questionnaires about your child's behaviour and how their health has impacted on your or your family's life. With your permission, we will ask your child's teacher to fill in some of these questionnaires too.

What are the possible benefits of taking part?

There is no direct benefit to your child from taking part in this study. However the study will provide essential information about any difficulties in thinking and problem solving that children who received 'brain cooling' might experience when they are in school. We hope that this research will eventually improve information and support for parents and also alert teachers to potential difficulties that these children may experience at school.

What will happen after the assessments and brain scan have been completed?

Once all assessments have been completed, we will also ask you if you would like us to send you a report about your child's performance. We will not send you a report if you don't want it; however, If any specific problems are detected for your child as part of this study, this will be first discussed with you, and then a referral to any relevant specialists will be made by the research team within 2 weeks from the completion of the assessments.

What if something goes wrong?

Although we think it is very unlikely that anything will go wrong, if your child is harmed by taking part in this research study, there are no special compensation arrangements. If your child is harmed through someone's negligence, then you will have grounds for legal action, but you may have to pay for it.

Regardless of this, if you wish to complain, or have any concerns about any aspect of the way you have been approached or your child treated during the course of this study, the normal

National Health Service complaints mechanisms should be available to you (Patient Advice and Liaison Service).

PALS

C Level Centre Block **Email:** PALS@suht.swest.nhs.uk

Mailpoint 81

Southampton General Hospital

Tremona Road

Will my taking part in this study be kept confidential?

All information collected about you and your child during the course of the research will be kept strictly confidential. Any study information about your child which leaves the hospital will have their name and address removed so they cannot be recognised from it.

With your consent only, your GP will be informed of any relevant medical information obtained about your child as part of the study.

What will happen to the results of the research study?

The results of the study, once analysed, will be published in appropriate professional journals, shared with medical and nursing professionals locally, nationally and internationally in reports, at conferences and through network groups.

Also, at the end of the study, we will organise an event for families of children who have had "brain cooling" at Princess Anne Hospital and non-medical professionals who work with the children (e.g. teachers, educational psychologists, community paediatricians), at which we will feed back the study findings. All data will be anonymous – neither you nor your child will not be identifiable.

A summary of the research findings can be sent to you if you request one.

Who is organising and funding the research?

This study is funded by Action Medical Research. It has been approved by the <xxx> Research Ethics Committee and the local research governance committee.

If you would like more information about the study you can check out this link: https://action.org.uk/research/birth-asphyxia-predicting-long-term-effects

University Hospital of Southampton (UHS) is the sponsor for this study based in the United Kingdom. We will be using information from your child and/or your child's medical records in order to undertake this study and will act as the data controller for this study. This means that we are responsible for looking after your information and using it properly. University Hospital of Southampton will keep identifiable information about your child for 30 years after the study has finished.

Your rights to access, change or move your information are limited, as we need to manage your information in specific ways in order for the research to be reliable and accurate. If you withdraw from the study, we will keep the information about you that we have already obtained. To safeguard your rights, we will use the minimum personally-identifiable information possible.

You can find out more about how we use your information https://www.hra.nhs.uk/information-about-patients/.

The research team will collect information from your child's medical records for this research study in accordance with our instructions.

The research team will keep your and your child's name, your child's NHS number and contact details confidential. The research team will use this information as needed, to contact you about the research study, and make sure that relevant information about the study is recorded for your care, and to oversee the quality of the study. Certain individuals from UHS and regulatory organisations may look at your medical and research records to check the accuracy of the research study. UHS will only receive information without any identifying information. The people who analyse the information will not be able to identify you and will not be able to find out you and your child's name, NHS number or contact details.

The research team will keep identifiable information about you from this study for 30 years after the study has finished.

When you agree to take part in a research study, the information about your child's health and care may be provided to researchers running other research studies in this organisation and in other organisations. These organisations may be universities, NHS organisations or companies involved in health and care research in this country or abroad. Your information will only be used by organisations and researchers to conduct research in accordance with the UK Policy Framework for Health and Social Care Research.

This information will not identify you or your child and will not be combined with other information in a way that could identify you or your child. The information will only be used for the purpose of health and care research, and cannot be used to contact you or to affect your care or that of your child. It will not be used to make decisions about future services available to your child, such as insurance.

Contact for further information

- Study email address - HTstudy@soton.ac.uk

- Dr Brigitte Vollmer – Principal Investigator

- Dr Rina Cianfaglione– Psychology Research Fellow

Tel: 023 8120 3036

Appendix K Parent consent form

Tel: 023 8120 3036

Email:HTstudy@soton.ac.uk

Participant ID #

PARENT / PARTICIPANT CONSENT FORM

Study Title: Neurodevelopmental trajectories and neural correlates in children with Neonatal Hypoxic-Ischaemic Encephalopathy (HIE) – NENAH

Ethics reference: IRAS N 263965

Name of Researchers: Dr Brigitte Vollmer, Dr Rina Cianfaglione

Name of Participant:

Please initial the box(es) if you agree with the statement(s):

I have read and understood the information sheet dated September2019 (Version 2) for the above study and have had the opportunity to ask questions about the study.	
I understand that my/ my child's participation is voluntary and I/he/she may withdraw at any time, without giving reason and without his/her medical care or my/his/her legal rights being affected.	

I understand that sections of any of my child's medical notes may be looked at by responsible individuals from the research team or by regulatory authorities where it is relevant to my/my child's taking part in research. I give permission for these individuals to access my child's records and for data to be used for the purpose of this study.

I agree for my child's teacher to be contacted to provide information regarding my child's emotions and behaviour at school.

I give my permission for my child to be video recorded during the assessments.

I agree for my child's GP to be contacted about my child's participation in this study and to be informed about any relevant medical information obtained about my child as part of the study.

I agree that data gathered in this study may be stored anonymously and securely and may be used for future research.

I agree / for my child / to take part in the above study.

Name of Child (PRINT NAME)……

Name of Parent/Guardian (PRINT NAME): ……..

Date: ……

Signature of Parent/Guardian: ……

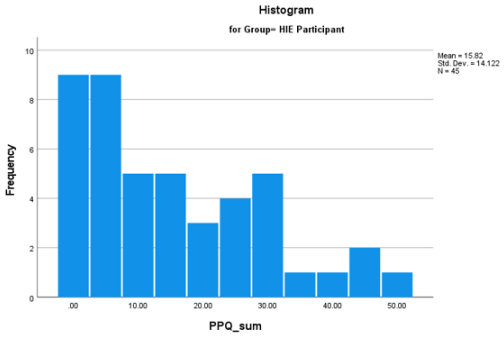
Name of Researcher (PRINT NAME): ……

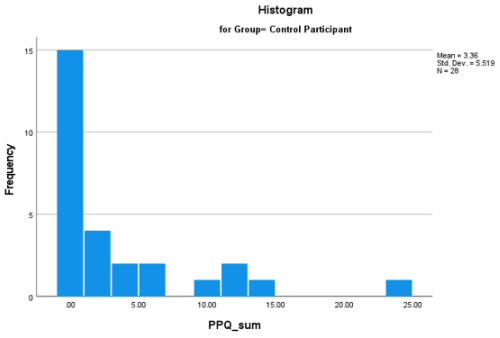
Date:……..

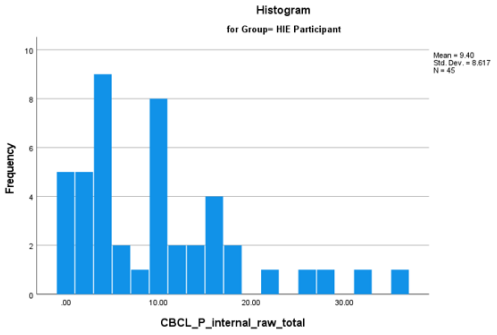
Signature of Researcher: ……

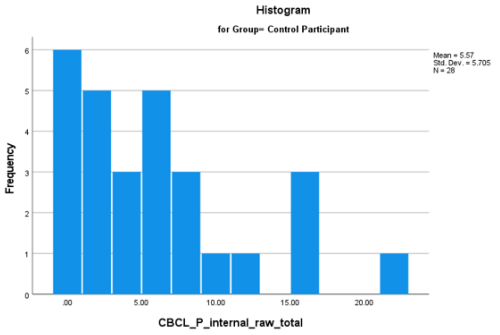
1 for participant; 1 for researcher; 1 for hospital notes

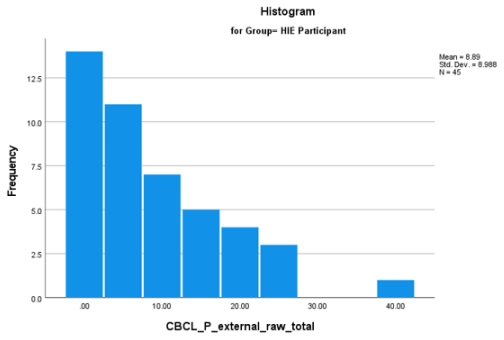
Appendix L Histograms

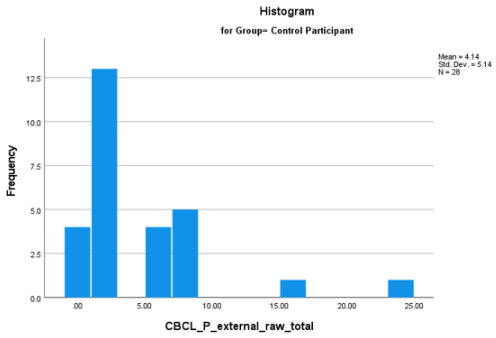


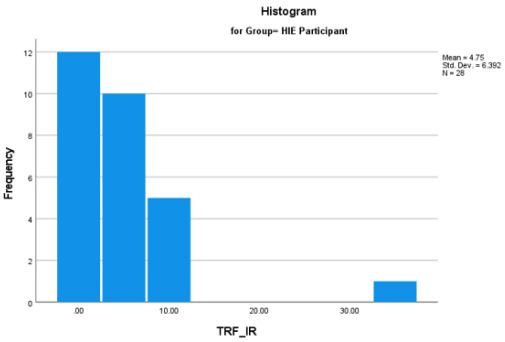


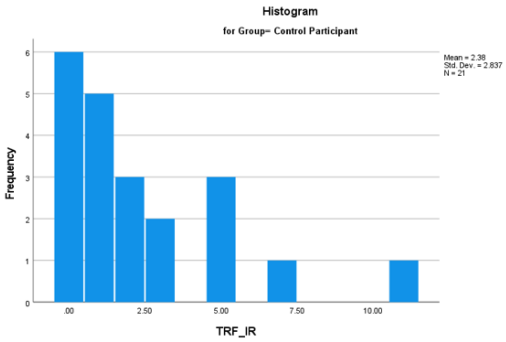


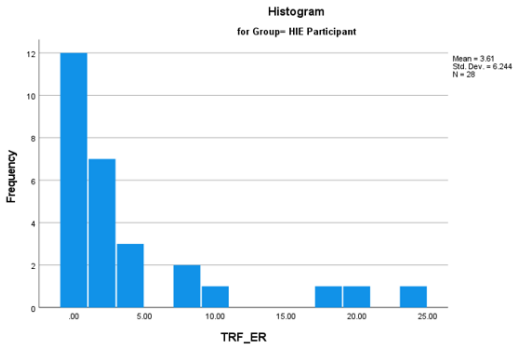


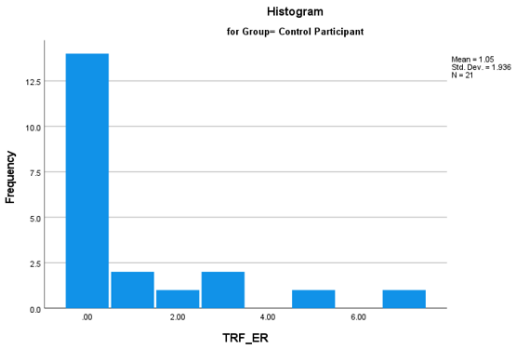





Histogram
for Group= HIE Participant
Mean = 4.75
Std. Dev. = 6.392
N = 28
Frequency
TRF_IR


Histogram
for Group= Control Participant
Mean = 2.38
Std. Dev. = 2.837
N = 21
Frequency
TRF_IR


Histogram
for Group= HIE Participant
Mean = 3.61
Std. Dev. = 6.244
N = 28
Frequency
TRF_ER


Histogram
for Group= Control Participant
Mean = 1.05
Std. Dev. = 1.936
N = 21
Frequency
TRF_ER

Appendix M Descriptive statistics

Table 5 *Frequency and Percentage of Children with HIE and Typically Developing Peers in CBCL Diagnostic Categories.*

	HIE (n=45)		Control (n=28)	
	Internalising	Externalising	Internalising	Externalising
	N (%)	N (%)	N (%)	N (%)
Normal	27 (60)	31 (68.89)	22 (78.57)	26 (92.86)
Borderline	5 (11.11)	6 (13.33)	1 (3.57)	1 (3.57)
Clinical	13 (28.89)	8 (17.78)	5 (17.86)	1 (3.57)

Table 6 *Frequency and Percentage of Children with HIE and Typically Developing Peers in TRF Diagnostic Categories.*

	HIE (n=28)		Control (n=21)	
	Internalising	Externalising	Internalising	Externalising
	N (%)	N (%)	N (%)	N (%)
Normal	24 (85.71)	22 (78.57)	20 (95.24)	20 (95.24)
Borderline	2 (7.14)	2 (7.14)	1 (4.76)	1 (4.76)
Clinical	2 (7.14)	4 (14.29)	0 (0)	0 (0)

Table 7 *Ranges in Scores on the PPQ, CBCL and TRF.*

	HIE		Control	
	Minimum	Maximum	Minimum	Maximum
PPQ	0	50	0	23
CBCL-I	0	35	0	21
CBCL-E	0	42	0	23
TRF-I	0	33	0	11
TRF-E	0	23	0	7

Appendix N Individual correlation tables

Table 8 *Spearman's Rank Correlations for Maternal Birth-related Stress, Parent Reported Internalising and Externalising Difficulties and Teacher Reported Internalising and Externalising Difficulties in the HIE Sample.*

Variable	1.	2.	3.	4.	5.
PPQ	-				
CBCL-I	.505**	-			
CBCL-E	.419*	.623***	-		
TRF-I	-.227	.133	.220	-	
TRF-E	-.260	.085	.471*	.680***	-

Note. *p<.05;**p<.01;***p<.001

Table 9 *Spearman's Rank Correlations for Maternal Birth-related Stress, Parent Reported Internalising and Externalising Difficulties and Teacher Reported Internalising and Externalising Difficulties in the Control Sample.*

Variable	1.	2.	3.	4.	5.
PPQ	-				
CBCL-I	.368	-			
CBCL-E	.590**	.509*	-		
TRF-I	-.261	.000	-.281	-	
TRF-E	.050	.160	.232	.418	-

Note. p<.05;**p<.01;***p<.001

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