

University of Southampton Research Repository

Copyright © and Moral Rights for this thesis and, where applicable, any accompanying data are retained by the author and/or other copyright owners. A copy can be downloaded for personal non-commercial research or study, without prior permission or charge. This thesis and the accompanying data cannot be reproduced or quoted extensively from without first obtaining permission in writing from the copyright holder/s. The content of the thesis and accompanying research data (where applicable) must not be changed in any way or sold commercially in any format or medium without the formal permission of the copyright holder/s.

When referring to this thesis and any accompanying data, full bibliographic details must be given, e.g.

Thesis: Author (Year of Submission) "Full thesis title", University of Southampton, name of the University Faculty or School or Department, PhD Thesis, pagination.

Data: Author (Year) Title. URI [dataset]

University of Southampton

Faculty of Environmental and Life Sciences

School of Psychology

**The Influence of Child Temperament and Parenting Behaviours on the
Development of Childhood Anxiety**

by

Lucy Ruffle BSc (Hons) MSc

ORCID ID: 0000-0002-2898-0800

Thesis for the degree of Doctorate in Clinical Psychology

May 2020

Word Count: 18 435

Abstract

Childhood anxiety disorders are the most prevalent type of mental health problem in children and they can have long-term detrimental impacts. It is therefore important to understand the risk factors involved in their development in order for effective prevention programmes to be developed. Observational and experimental research have implicated specific parenting behaviours, temperamental, and genetic factors in the development of childhood anxiety disorders.

The first chapter examines the temperamental factors implicated in the development of childhood anxiety disorders. Behavioural Inhibition (BI) is a temperamental style that can be identified from infancy, which is characterised by persistent demonstrations of fear, wariness, and reticence in unfamiliar situations. A systematic literature review and meta-analysis is presented, examining the association between BI and presentations of childhood anxiety. 55 studies reporting a quantitative outcome for the association between BI and childhood anxiety were included in the meta-analysis. The 55 studies, from 43 independent samples, presented 135 effect sizes for the association between BI and childhood anxiety. A positive significant association was found between BI and childhood anxiety. Meta-regressions examined factors moderating the association; stronger effect sizes were found when: anxiety was measured continuously, BI was measured by parent-report, and anxiety was measured by parent-report. Recommendations are made for further research to examine the interaction between temperamental and parenting factors in the development of childhood anxiety.

The second chapter presents an empirical study investigating the impact of two specific parenting behaviours on infant affect and behaviour. Research has shown that parental expression of anxiety can increase their child's anxiety, whereas demonstrating encouraging behaviour is associated with less anxiety in the child. The study examined whether it is possible to modify these parenting behaviours using video tuition to model the target parenting behaviours, and then examined if this had an impact on infants' affect and behaviour. Twenty-two mother-infant dyads completed two experimental social-referencing tasks in which they met with a stranger. In one, mothers expressed anxiety when interacting with a stranger, and in the other they expressed encouragement. Infant affect and behaviour during subsequent interaction with the strangers was recorded. Measures of maternal mood/anxiety and infant temperament were gathered to examine if these moderated any effects found. The video tuition was successful in modifying mothers' behaviours. Infant temperament had the strongest effect on infants' affect and behaviour, irrespective of the experimental condition. The study has demonstrated the utility of video tuition for modifying behaviours implicated in the development of childhood anxiety. Recommendations are made to replicate the study in a clinical sample and for further exploration of temperamental factors.

Keywords: childhood; infancy; anxiety; behavioural inhibition; temperament; parenting

Table of Contents

Abstract	2
Table of Contents	3
Table of Tables	5
Table of Figures	6
Research Thesis: Declaration of Authorship	7
Acknowledgements	8
Definitions and Abbreviations	9
Chapter 1: What is the association between Behavioural Inhibition (BI) and presentations of childhood anxiety?: A systematic review and meta-analysis	10
Introduction	11
Method	20
Results	27
Discussion	32
Chapter 2: Effects of modification of maternal expressed anxiety and encouragement on infant affect and behaviour, using a social referencing paradigm.	40
Introduction	41
Method	53
Results	60
Discussion	68

Appendix A – Summary of the 55 Studies Included in the Meta-Analysis	78
Appendix B – Summary of all excluded articles reviewed at full text stage	86
Appendix C - Graph to illustrate relationship between Fisher’s Z and Pearson’s r	90
Appendix D – Standard Quality Assessment Criteria.....	91
Appendix E – Forest Plot.....	98
Appendix F - Histograms for maternal mood and anxiety questionnaires	104
References	106

Table of Tables

Table	Title	Location
1	ANOVA to Explore if Order of the Experimental Conditions Influenced Infant Affect	Page 63
2	Summary of the 55 Studies Included in the Meta-Analysis	Appendix A
3	Assessment of Study Quality for the 55 Included Studies using the 11 Relevant Criteria from Kmet et al. (2004) Standard Quality Assessment Criteria for Quantitative Studies	Appendix D

Table of Figures

Figure	Title	Location
1	PRISMA flowchart	Page 23
2	Scatter plot illustrating the association between the number of years between the BI and anxiety measures being conducted and the size of the association (Z) between BI and anxiety	Page 28
3	Box plot examining the differences in effect size between social anxiety outcomes and other (non-social) anxiety outcomes	Page 30
4	Funnel plot to visually assess for publication bias	Page 31
5	Linear regression (black line) between infant negative temperament (measured by the IBQ-R VSF) and number of looks from infant to mother in part 2	Page 62
6	Linear regression (black line) between infant negative affect (measured by the IBQ-R VSF) and infant fearfulness	Page 64
7	Graph to illustrate relationship between Fisher's Z and Pearson's r	Appendix C
8	Forest Plot illustrating the effect size (Z) between BI and anxiety from 135 effect sizes from 55 studies	Appendix E
9	Histogram to illustrate responses to the Social Phobia Inventory (SPIN)	Appendix F
10	Histogram to illustrate responses to the Penn State Worry Questionnaire (PSWQ)	Appendix F
11	Histogram to illustrate responses to the Edinburgh Postnatal Depression Scale (EPDS)	Appendix F

Research Thesis: Declaration of Authorship

Print name:	Lucy Ruffle
-------------	-------------

Title of thesis:	The Influence of Child Temperament and Parenting Behaviours on the Development of Childhood Anxiety
------------------	---

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. None of this work has been published before submission.

Signature:		Date:	22.05.2020.
------------	--	-------	-------------

Acknowledgements

*“What’s wrong with knowing what you know now
and not knowing what you don’t know until later?”*

- A.A. Milne

Firstly, I’d like to say thank you to my two thesis supervisors: Dr Pete Lawrence and Dr Angharad Rudkin. Pete, I am grateful for your enthusiasm and vision in conceptualising these two projects, your impressive grasp of the subject, your dedication as a ‘stranger’ in the experimental study, and for your helpful feedback when reviewing the manuscripts. Angharad, I am grateful for your consistent and reliable support throughout these projects; I always felt reassured that you were there to support me whenever needed, and I am also grateful for your helpful feedback when you reviewed the manuscripts.

A very big thank you goes to all of the mums and babies who took part in our study. Without you putting your faith in us and giving up your time to help with this research endeavour, this project would not have been possible. Thank you to everyone who helped in spreading the word to their friends and family about our study. Thank you to Rich Mutimer, Erich Graf, and Richard Curtis for helping out in the ‘baby study’. This project took a lot of coordinating and I would like to say thank you to Paul Reynolds in the Psychology department for all his support in setting up the recording equipment, and to Manar and Amy for their work in coding the video recordings. I would also like to say thank you to all the wonderful admin team in the FOS office who were so friendly and welcoming to all our mums and babies as they arrived for the study.

Thank you to Michael Miles, our Voluntary Research Assistant, for your incredible work supporting these two projects. Your contribution to these projects has been invaluable and you’ve helped every step of the way, always going above and beyond in everything I asked for your help with. Thank you!

To my cohort of clinical psychology trainees – I have loved training with you since 2017. It’s been wonderful riding the highs and lows of training with you, and as much as we can’t wait to get to the end, I’m also going to miss the time we have together.

Finally, a special thank you to Tom, my parents - Jane and Ged, and my friends, for their unwavering support - not only with this thesis project, but all throughout the many years that I have been working towards training as a clinical psychologist. Tom, your bright, enquiring, scientific mind has helped me through countless statistics crises – but it is your patience and kindness that I am most grateful for.

Definitions and Abbreviations

BI	Behavioural Inhibition
BIS	Behavioural Inhibition System
CI	Confidence Interval
<i>d</i>	Cohen's <i>d</i> (effect size)
DSM	Diagnostic and Statistical Manual of Mental Disorders
ERGO	Ethics and Research Governance Online
EPDS	Edinburgh Postnatal Depression Scale
GAD	Generalised Anxiety Disorder
IBQ-R VSF	Infant Behaviour Questionnaire – Revised, very short form
κ	Kappa (statistic used to measure inter-rater reliability)
M	Mean
MASC	Multidimensional Anxiety Scale for Children
N	Number of Participants
n	Number of Studies
OR	Odds Ratio
PRISMA	Preferred Reporting Items for Systematic Reviews and Meta-Analyses
PROSPERO	International Prospective Register of Systematic Reviews
PSWQ	Penn State Worry Questionnaire
R	A language and environment for statistical computing and graphics
RCADS	Revised Child Anxiety and Depression Scale
SAD	Social Anxiety Disorder
SD	Standard Deviation
SPIN	Social Phobia Inventory
SR	Social Referencing
VIG	Video Interaction Guidance
VFT	Video Feedback Therapy
Z	Fisher's Z Transformation

**Chapter 1: What is the association between Behavioural Inhibition (BI) and presentations
of childhood anxiety?: A systematic review and meta-analysis**

Introduction

Anxiety disorders

Fears are common in childhood and in many cases the types of fears experienced are adaptive or associated with the child's level of psychological and social development, such as toddlers experiencing fears about imaginary creatures before they are able to distinguish between fantasy and reality, or adolescents fearing peer rejection when peer relationships become the source of self-esteem. However, for some children the fear they experience becomes developmentally atypical and maladaptive, resulting in the child being prevented from engaging in developmentally appropriate activities, such as going to school, making friends, or engaging with extracurricular activities. A child may be diagnosed with an anxiety disorder in cases where the frequency and severity of anxiety symptoms are causing distress and impairing their ability to engage in activities of daily living typical for their stage of development. Typical symptoms of anxiety disorders include cognitive symptoms (e.g. inaccurate appraisal of threat), affective symptoms (e.g. feelings of fearfulness), physiological symptoms (e.g. increased heart rate, shaking, butterflies in stomach) and behavioural symptoms (e.g. avoidance of feared situations or objects) (Ollendick, King, & Yule, 1994).

Anxiety disorders typically have an onset in middle childhood – a large observational study (N > 9000) in the USA reported the median age of onset for anxiety disorders was eleven years old (Kessler et al., 2005), although there is evidence to suggest that the onset can be much earlier in infancy or early childhood (Tandon, Cardeli, & Luby, 2009). Without intervention, anxiety disorders are often chronic in nature, with low levels of natural remission (Swinson, 1997). Childhood anxiety disorders are strong predictors of both co-morbid mental health problems and anxiety disorders in adulthood (Public Health England, 2016).

Prevalence of childhood anxiety disorders

Anxiety disorders are the most common mental disorders in childhood and adolescence. An England-wide survey of children's mental health in 2017 reported that one in eight (12.8%) of five to 19 year olds had a mental disorder and one in 18 (5.5%) preschool children met the diagnostic criteria for at least one mental disorder (NHS Digital, 2018). Previous surveys of the prevalence of mental disorders in children and young people across England have demonstrated a trend of increasing prevalence of mental disorders in five to 15 year olds in England over time, from 2004 to 2017 when the most recent study was conducted. Similar prevalence of mental disorders in childhood and adolescence have been found globally; a systematic review and meta-analysis study reported a worldwide prevalence of 13.8% for any mental disorder in young people up to 18 years, with anxiety disorders as the most common, with an estimated global prevalence rate of 6.5% (Polanczyk, Salum, Sugaya, Caye, & Rohde, 2015).

Impact on the child

Anxiety disorders in childhood can have detrimental and long-term impacts on children, their families, and the wider community. Childhood anxiety disorders have been found to be associated with school truanting, school drop-out, and poorer educational attainment (Public Health England, 2016). The impact of childhood anxiety has been shown to be long-standing, with childhood anxiety disorders being associated with lower earnings in adulthood (Public Health England, 2016) and increased social and health care costs (Creswell et al., 2015). Anxiety disorders during childhood and adolescence have been shown in a longitudinal study to be associated with a significant increase in the probability of having an anxiety disorder in early adulthood (Essau, Lewinsohn, Lim, Ho, & Rohde, 2018). Childhood anxiety disorders are also associated with increased risk of experiencing mood disorders, other anxiety disorders, and substance misuse (Public Health England, 2016).

Investigations into the long-term impacts of childhood anxiety can inadvertently overlook the present-day distress and suffering that childhood anxiety can cause. A cross-sectional study examining quality of life in a paediatric sample found that the severity of anxiety symptoms was significantly inversely related to quality of life. Additionally, this study reported that social anxiety and somatic-related anxiety were particularly associated with worse quality of life outcomes (Ramsawh & Chavira, 2016). Biedel, Turner, & Morris (1999) compared the experiences of 50 children with social phobia (as diagnosed by DSM-IV criteria) to a control group of children without an anxiety disorder and found higher levels of loneliness, tearfulness, and dysphoria in the children with social phobia, which highlights the current emotional suffering experienced by young people with social phobia.

Impact on the family

The impact on families of early childhood anxiety disorders (2 – 5 years) was investigated in a large scale cross-sectional study (N>900) (Towe-Goodman, Franz, Copeland, Angold, & Egger, 2014). Parents of children with early anxiety reported more severe worry, depression, and fatigue, as well as increased disruption to relationships and limitations in their ability to engage with social or personal activities, as compared to parents of children with early childhood disorders other than anxiety. The authors suggest that these family outcomes may serve to maintain a child's anxiety if the family becomes increasingly socially isolated and thus the child misses out on opportunities for engagement with activities important for their development, and the parent misses out on accessing potential sources of support. Lost parent productivity was also found to be a consequence of childhood anxiety disorders (Creswell et al., 2015).

Understanding the risk factors for childhood anxiety

It is important to understand the factors which contribute to the development of childhood anxiety disorders in order for effective preventative programmes to be developed. Finding effective

ways to prevent the short- and long-term suffering caused by anxiety disorders is crucial, given the personal and family distress that childhood anxiety can cause, along with the known detrimental educational, health, and social outcomes. Donovan and Spence (2000) present the case for the importance of developing preventative strategies for childhood anxiety disorders, in light of evidence that patterns of anxiety become more difficult to change the longer the duration of the anxiety.

Ashford, Smit, van Lier, Cuijpers, and Koot (2008) concluded from a nine-year longitudinal study investigating the risk factors associated with childhood internalising problems (including anxiety and depression) that up to 57% of cases of internalising problems in late childhood (age eleven) could be ameliorated effectively through preventative interventions addressing risk factors identified between two to five years of age.

Early intervention is desirable not only on a personal level, to relieve suffering, but also on a broader scale for social policy. Heckman (2000) reviewed research examining the economic impact of social policy across a broad range of areas, including investments into early childhood programmes, and concluded that, in general, compared to later interventions, early intervention costs less and is more effective.

Behavioural Inhibition as a risk factor

One risk factor for the development of childhood anxiety is the temperamental style of Behavioural Inhibition (BI). Kagan and colleagues first described the temperamental style of BI from behavioural observations of young children (Kagan, Reznick, Clarke, Snidman, & Garcia-Coll, 1984). Klein and Mumfer (2018) summarise the categorisation of BI as *“young children who exhibit fear, wariness, and reticence in unfamiliar situations and with unfamiliar people”* (p. 283). BI is typically assessed from the second year of life (i.e. from 12 months) and typical behavioural presentations in an infant or child with BI include seeking proximity to familiar and ‘safe’ figures, a reluctance to engage

in social interactions, and expressing distress in novel situations (Dodd, Hudson, & Rapee, 2017). Studies have also shown that BI identified through observational measures is also correlated with biological measures, such as increased heart rate, increased pupil dilation, and increased levels of salivary cortisol (Kagan, 2018).

Klein & Mumper (2018) highlight that BI differs from other temperament or personality traits (e.g. neuroticism) as BI is context-dependent, whereas other personality traits are considered to be evident across all contexts. It is important to note that the research projects which contributed to the development of the concept of BI (as a stable temperamental style) were conducted in a university laboratory setting, with the samples involved being predominantly Caucasian, from middle-class backgrounds, and described as 'secure' families (Kagan, 2018). Hence, BI is not a concept that can be broadly generalised outside of these parameters.

BI is typically assessed in later infancy, toddlerhood, or early childhood, via laboratory observation or parent-report measures, e.g. The Behavioural Inhibition Questionnaire (BIQ; Bishop, Spence & McDonald, 2003) and the Behavioral Inhibition Scale (BIS; Muris, Meesters, & Spinder, 2003). Kagan and colleagues originally described BI as a categorical construct, that is, a child was behaviourally inhibited or not. Later research has conceptualised BI as an extreme on a continuum (Lemery, Essex, & Smider, 2002). However, Kagan continues to advocate for the use of the original categorical classification of BI (Kagan, 2018).

Observational measures of BI are often conducted in a laboratory setting, with varying procedures and protocols being used by different research groups (Rothbart & Bates, 2006). Typically, observational BI measures will involve the child being exposed to novel stimuli (e.g. toys, unfamiliar adults, unfamiliar peers) and coding observed behaviour such as proximity to their caregiver, referencing to their caregiver, amount of interaction with the unfamiliar person or object, expressions of fearfulness, and time to approach the unfamiliar stimuli (Kim et al., 2011).

Empirical studies have reported a link between BI and both childhood anxiety symptoms and disorders. Pahl, Barrett, and Gullo (2012) presented findings from a large scale (N = 236) cross-sectional study, examining the risk factors from early childhood (4 – 6 years) for anxiety, using parent-report measures of BI and anxiety, as well as parent-report measures of parenting behaviours, and of child stress, anxiety, and depression. Their analyses, using structural equation modelling, found a positive association between BI and child anxiety at preschool age. BI also partially mediated the relationship between early childhood anxiety and the parent factors of maternal parenting stress and both maternal and paternal negative affectivity. The authors discuss the importance of developing preventative programmes that take into consideration the impact of these parent variables (e.g. parenting stress) that were found to influence the association between BI and childhood anxiety.

A longitudinal prospective study by Hudson and colleagues followed up behaviourally inhibited (N = 102) and behaviourally uninhibited (N = 100) children from pre-school (3 – 4 years) (Hudson, Dodd, & Bovopoulos, 2011) to middle childhood (9 years) (Hudson & Dodd, 2012) and to late childhood (12 years old) (Hudson, Murayama, Meteyard, Morris, & Dodd, 2019). BI was found to be a significant predictor of childhood anxiety disorders in middle childhood, as measured by the Anxiety Disorders Interview Schedule – this predictive association remained significant even when controlling for baseline anxiety. This prospective association between BI and childhood anxiety was also found to continue into late childhood (Hudson et al., 2019). This latest follow up also reported on the additive and interactive effects of parent factors influencing the relationship between BI and childhood anxiety; whereby, behaviourally inhibited pre-schoolers had higher anxiety symptoms by late childhood only when maternal over-involvement had been present when the child was age four.

Meta-analytic studies

Kostyrka-Allchorne, Wass, and Sonuga-Barke (2020) presented a systematic review and meta-analysis of studies examining the predictive association between parent ratings of infant temperament and later symptoms of psychopathology in childhood and adolescence. A significant positive association was found between BI and psychopathology in childhood and adolescence. Later psychopathology was also found to be significantly associated with parent reports of infant negative emotionality and activity levels. However, the authors commented on the lower than expected effect sizes for these findings and queried the predictive power of infant temperament assessed by questionnaire rather than observation. Hence, it is important to understand whether the strength of the association between BI and later psychopathology differs depending on how BI is ascertained.

Clauss & Blackford (2012) reported a meta-analysis examining the association between BI and the risk for children developing social anxiety disorder (SAD), in which BI was associated with a greater than sevenfold increase in odds of developing SAD. Seven prospective studies were included in this meta-analysis, which showed a strong and significant association between BI and risk for developing SAD (odds ratio (OR) = 7.59; 95% confidence interval (CI) = 3.03 – 19.00), even after controlling for methodological difference in the measure of BI, the type of control group used, as well as the age of both BI assessment and anxiety diagnosis. Parental psychopathology is a known risk factor for childhood anxiety disorders and the impact of this variable was also examined – BI was still associated with an increased risk for SAD in the subsample of studies in which children were not recruited on the basis of parental psychopathology.

However, as outlined above, the theoretical and empirical research suggests that BI is not only associated with SAD but with all presentations of childhood anxiety. Sandstrom, Uher, & Pavlova (2019) presented a meta-analysis examining the prospective association between childhood BI and anxiety, which included 27 studies with 20 unique samples. Sandstrom et al. (2019) reported findings

consistent with Clauss & Blackford (2012) albeit with a lower point prevalence of the association between BI and SAD (OR = 5.84; 95% CI = 3.38 – 10.09). Additionally, compared to its absence, BI in early childhood was associated with an almost threefold (OR = 2.80; 95% CI = 2.03 – 3.86) increase in the odds of later anxiety in general, as well as increased odds of later generalised anxiety disorder (GAD; OR = 2.04, 95% CI = 1.43 – 2.91) and specific phobia (OR = 1.49, 95% CI = 1.03 – 2.14).

The two meta-analyses examining the association between BI and anxiety disorders (Clauss & Blackford, 2012; Sandstrom et al., 2019) only included studies employing a prospective design investigating the relationship between BI and childhood anxiety, to the exclusion of retrospective and cross-sectional studies. Therefore, while meta-analyses have demonstrated significant prospective relationships between BI and anxiety disorders, there is no meta-analytic quantification of whether cross-sectional and prospective associations differ.

Aims of this review

This current systematic review and meta-analysis aims to complement the findings from Clauss & Blackford (2012) and Sandstrom et al. (2019) by examining the strength and significance of the association between BI and presentations of childhood anxiety across all study designs, including cross-sectional and retrospective designs as well as prospective designs. Specifically, we will produce the first meta-analytic quantification of whether cross-sectional associations of BI and child anxiety differ from their prospective association.

Inclusion of cross-sectional and retrospective studies, in addition to prospective studies, will capture a greater breadth of the literature examining BI and childhood anxiety. We are aware from Sandstrom et al. (2019) that seven studies were excluded on the basis of BI being measured retrospectively and four cross-sectional studies were excluded. We expect that the findings from this systematic review and meta-analysis will contribute to our understanding of how the concepts of BI

and childhood anxiety relate. Inclusion of a broad scope of the literature will also allow us to examine other design factors which might moderate the association between BI and anxiety. Namely, the moderation analysis will examine the relative influence of: study design (prospective, cross-sectional or retrospective), how BI and anxiety were defined (categorically or as continuous constructs), how BI and anxiety were measured (e.g. parent-report, clinical interview, clinical observation, self-report), and the amount of time between the conduction of the BI and anxiety measures.

Method

Protocol

The protocol for this systematic review and meta-analysis was registered on the International prospective register of systematic reviews (PROSPERO; registration number: CRD42019153829) prior to commencing the searches. This included details of the review questions, the databases to be used, the inclusion and exclusion criteria, and the planned method for analysis.

Eligibility criteria

The inclusion and exclusion criteria were drafted before commencing the searches and these criteria were piloted on a sample of 50 papers from the scoping searches. From this piloting exercise the inclusion and exclusion criteria were finalised.

Inclusion criteria:

1. Studies of human participants between 0 and 18 years of age.
2. Studies reporting a validated measure of behavioural inhibition (BI), and a validated measure of anxiety in childhood (0 - 18 years), for the same children. The BI measure can be conducted via observation or parent-report (not self-report); the anxiety measure can be from clinical interview or from clinician-, parent-, teacher- or child self-report.
3. Studies published in a peer-reviewed journal in English.

Exclusion criteria:

1. Studies which do not include a validated quantitative measure of both BI and childhood anxiety for the same child.

2. Intervention studies which do not include a baseline measure of BI or childhood anxiety symptoms (i.e., pre-intervention).
3. Papers with no extractable quantitative data, e.g. review papers, conference abstracts, theoretical discussions.
4. Studies with samples identified in light of a specific health condition (for example, children with neurodevelopmental conditions, such as Autism Spectrum Condition or Attention Deficit Hyperactivity Disorder, intellectual disabilities, specific physical health conditions, or diagnosed mental health conditions).

As the temperamental style of Behavioural Inhibition (BI) was defined by Kagan and colleagues in 1984, the search was restricted to studies from this year onwards. BI is operationalised as the expression of fear, wariness, or reticence in unfamiliar situations or with unfamiliar people, observed in young children. Only studies measuring BI as defined by Kagan were included. Studies which did not measure BI, but employed a measure of a feature of Gray's sensitivity to reinforcement model - the Behavioural Inhibition System (BIS) (Gray, 1970) - were not included.

Searches

The searches were conducted in October, 2019 using the electronic databases of CINAHL (via EBSCO), Embase (via Ovid), MEDLINE (via EBSCO) and PsycINFO (via EBSCO), using the following search terms:

anxi* OR phobi* OR wariness OR internalizing OR internalising

AND

(behavioral OR behavioural OR temperament*) N3 inhibit* OR BI

AND

'longitudinal' OR 'prospective' OR 'follow up' OR 'follow-up' OR 'followup' OR 'cohort' OR
'retrospect*' OR 'associat*' OR 'trajector*' OR 'predict*' OR 'relations* between' OR 'link*
between'

AND

infan* OR toddler* OR child* OR youth* OR young OR teen* OR adolescen* OR paediatric*
OR pediatric*

The PRISMA flow diagram (Figure 1) illustrates the review process and the number of records identified at each stage. 744 records were identified from the initial search (after removal of duplicates). Nine additional records were identified from hand-searching the reference list of previous relevant reviews.

Study selection

Two reviewers double-screened eligibility of all 753 abstracts. Articles were included in the next stage of full text review if either reviewer selected an abstract for inclusion. Any disagreements were discussed by the research team and an agreement was reached in all cases. Two reviewers assessed the 234 articles selected for full text review, to identify which studies fully met the inclusion and exclusion criteria for the review. Seventy articles met the eligibility criteria for inclusion; however, data appropriate for the meta-analysis was not available for 15 of these articles. In five cases the authors did not respond to e-mail requests for the required data, and in ten, the study design did not produce data appropriate for this meta-analysis, e.g. only children with BI were included with no comparison group. We retained a total of 55 records for inclusion in this meta-analysis. Appendix A

summarises the 55 studies included in the meta-analysis and Appendix B outlines all of the excluded studies with reasons for exclusion.

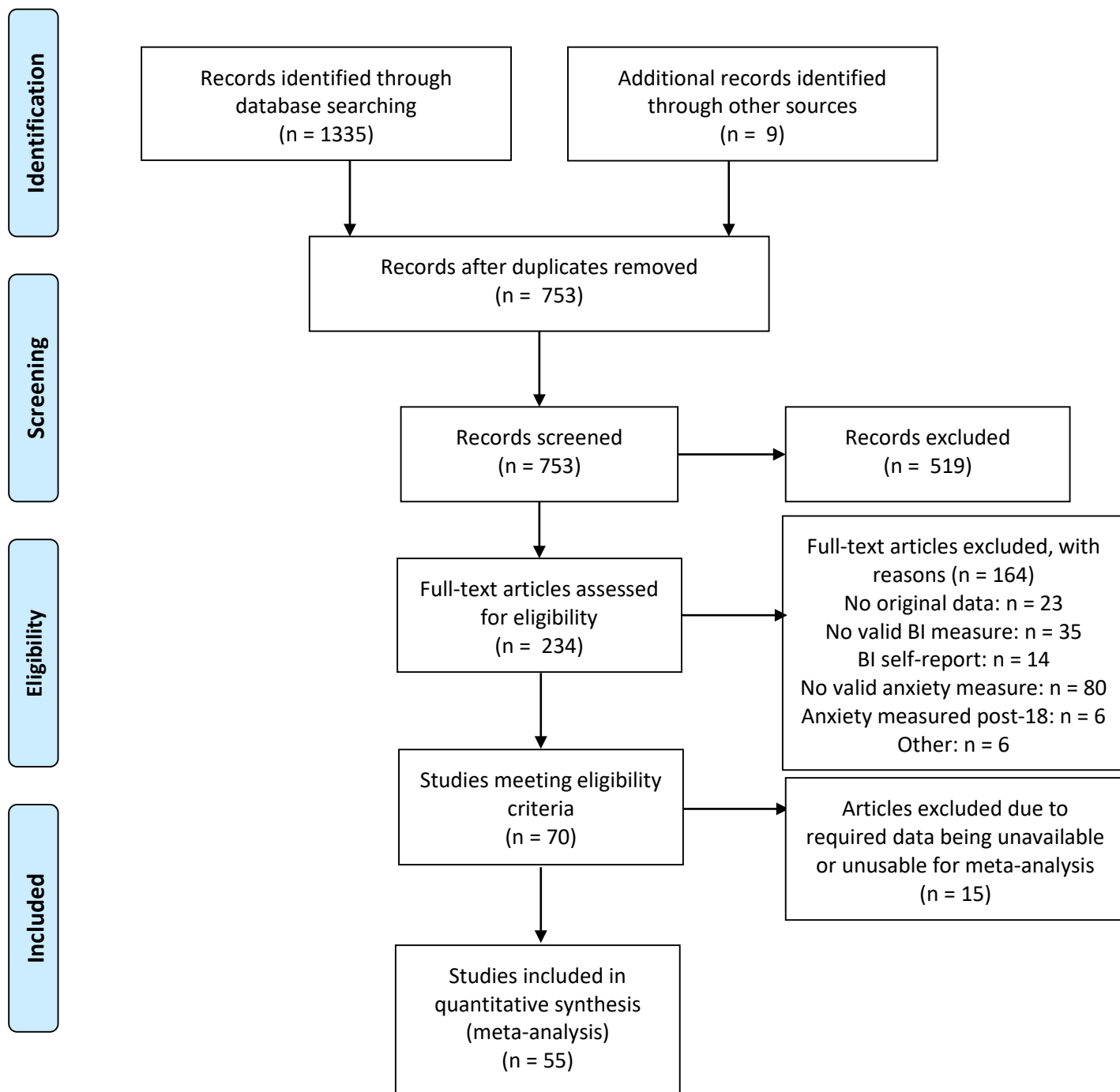


Figure 1. PRISMA flowchart

Data extraction

For each included study the following data was extracted: first author, year of publication, citation, sample size, study design, age of child when BI was measured (mean, SD, range), age of child when anxiety measured (mean, SD, range), percentage of female infants/children, all outcome measures of BI and all outcome measures of childhood anxiety symptoms (including the type of measure, e.g. parent-report, clinical observation and whether BI and anxiety were measured categorically or continuously) and anxiety outcome (e.g. any anxiety, social anxiety, phobia).

Synthesis of results

Meta-analyses (weighted, using random effects modelling) were carried out in the R software environment. Some studies had more than one effect size; for example two different measures of anxiety. This violates the assumption of independence of data in meta-analysis. We used robust variance estimation in order to examine all reported effects. This allows for dependent effect sizes, by correcting studies' standard errors to account for associations between effects within studies (Hedges, Tipton, & Johnson, 2010). We used the 'robumeta' package in the R software environment for primary and moderator analyses (Fisher & Tipton, 2015). To allow for all included studies to be incorporated in the data synthesis the outcomes were transformed into a common metric - Fisher's Z scores were used as an index of effect size of the association between the two main outcome measures of BI and childhood anxiety. Fisher's Z values are very closely aligned to Pearson's r values; they provide a confidence interval and, additionally, take into account skewed sampling distributions, which are a particular problem in small samples sizes (Cox, 2008). A figure to illustrate the relationship between Pearson's r and Fisher's Z can be found in Appendix C. Studies presenting categorical data for BI and anxiety (i.e. number of children classified as BI/non-BI and number of children diagnosed/not diagnosed with an anxiety disorder) were converted into odds ratios using the effect size calculator

from the Campbell Collaboration (Wilson, n.d.). Data in the form of odds ratios were then converted to Fisher's Z scores using the effect size calculator from the Campbell Collaboration (Wilson, n.d.). All other extracted data formats (i.e. Pearson's r, Chi squared) were converted to Fisher's Z scores in the 'compute.es' R package, for computing effect sizes. This package uses recommended formulas as described in The Handbook of Research Synthesis and Meta-Analysis (Cooper, Hedges, & Valentine, 2008). As noted above, 10 studies were excluded due to the data format being inappropriate for the meta-analysis. It was not possible to compute a Fisher's Z score for studies using categorical measures of BI and anxiety which only included children categorised as BI, i.e. no comparison group, nor was it possible for Beta-coefficients to be converted into Fisher's Z scores when continuous measures were used. The impact of heterogeneity between studies was assessed using the I^2 statistic.

Meta-regressions were conducted for the secondary research questions, examining the impact of moderators. These analyses included assessment of moderation of the association between BI and anxiety by continuous variables (e.g. years between the conduct of the BI and anxiety assessments) and by categorical variables (e.g. method of BI assessment, method of anxiety assessment, study design).

Typically the analyses in a meta-analysis would be performed using the transformed values (Fisher's Z) and then converted back to the original metric to present the data (Borenstein, Hedges, Higgins, & Rothstein, 2009). However, here we present the findings using the transformed values (Fisher's Z) given that the original data came from multiple metrics, including Pearson's r, odds ratios and Chi squared, and hence conversion back to original metrics is not possible.

Study quality and Publication bias

Study quality of the 55 included studies was assessed using the Standard Quality Assessment Criteria for Evaluating Primary Research Papers from a Variety of Fields (Kmet et al. 2004), which can

be used to assess research quality across a broad range of study designs. The quality of quantitative research papers is rated across domains, including clarity of research question/objective, measures employed to minimise bias such as random allocation and blinding, and reporting of estimates of variance. Each relevant domain was rated on a 0 – 2 scale to indicate if the quality standard domain was not met, partially met, or met. An overall summary score (between 0 and 1) for each study was calculated (sum of the total scores for each relevant item divided by the total possible score), to allow for quality comparisons between studies. See Appendix D for the quality ratings for all 55 included studies. Prior to conducting the quality rating it was considered that it may be necessary to restrict the meta-analysis to include only studies which were of high quality, or to perform a sensitivity analysis to examine the effect of study quality. However, the quality ratings for all 55 included studies was high (range from 0.77 – 1.00) and thus all 55 studies were included at all stages of analysis.

The 'metafor' R package was used for the assessment of publication bias, in order to conduct the Egger test and produce a funnel plot (Viechtbauer, 2010). To conduct publication bias assessments a weighted mean effect size for each included independent sample was calculated. This was required because these assessments require statistical independence and they are not validated for meta-analysis, such as this one, which is using robust variance estimation.

Results

Included studies

The meta-analysis included 55 studies published in peer-reviewed journals between 1993 and 2019 (Appendix A summarises all of the 55 studies included in the meta-analysis.). The studies were comprised of 43 independent samples and 135 effect sizes quantifying the association between BI and anxiety. BI was measured categorically (i.e. presence or absence of BI) in 15 of the studies and measured continuously in 40 of the studies. BI was measured by parent-report in 26 studies, by observation in 12 studies, and by a combination of observation and self-report in 17 studies. Anxiety was measured categorically (i.e. presence or absence of anxiety disorder diagnosis) in 16 of the studies and measured continuously in 39 of the studies. The anxiety outcomes reported on were: any anxiety (n = 61), any non-social anxiety (n = 7), social anxiety (n = 39), generalised anxiety (n = 7), separation anxiety (n = 12), phobia (n = 7), panic (n = 2) and selective mutism (n = 1) (NB. one study presented a compound measure of separation and generalised anxiety).

Sensitivity Analysis

Sensitivity analyses were conducted, which showed that the effect sizes and Tau^2 did not differ with the size of ρ and hence suggests that these values are robust to different values of p (Fisher & Tipton, 2015). Therefore, for all analyses ρ was kept at 0.8.

Association between BI and childhood anxiety

First, the meta-analysis examined the overall association between BI and anxiety, pooling all of the 135 effect sizes from the 55 studies – a small but significant association between BI and anxiety was found. The overall intercept model (not accounting for any moderators) had an overall point-

estimate of $Z = 0.259$ (95% Confidence Interval (CI) = 0.187 – 0.332), $p < .001$). Heterogeneity was high: $I^2 = 94.15\%$. A forest plot to visually represent this analysis can be found in Appendix E.

Moderation by study design features

We conducted meta-regressions to examine whether variables relating to the study design were associated with effect size differences.

Study design

The association between BI and anxiety was significant for both prospective ($Z = 0.207$, 95% CI = 0.138 – 0.275) and cross-sectional designs ($Z = 0.325$, 95% CI = 0.184 – 0.466). The association was not significant for retrospective designs ($Z = 0.247$, 95% CI = -0.162 – 0.656). There were no statistically significant differences in the strength of the association between BI and anxiety when comparing prospective, cross-sectional, and retrospective study designs. Within the prospective studies, the amount of time between the BI and anxiety measures being administered made a significant difference on the overall effects ($p = .008$). As the time between BI and anxiety measures increases the strength of the association between BI and anxiety decreases, as shown in Figure 2.

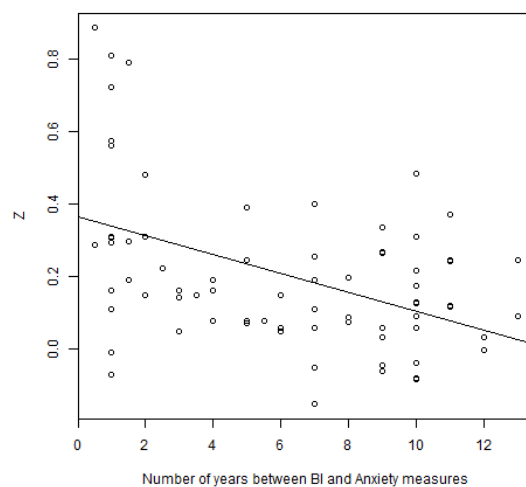


Figure 2. Scatter plot illustrating the association between the number of years between the BI and anxiety measures being conducted and the size of the association (Z) between BI and anxiety.

BI measures

Overall effects did not differ significantly as a function of how BI was categorised (i.e. as a categorical or continuous measure): $t(23, 2) = 0.074, p = .34$.

The association between BI and anxiety was significant when BI was measured by parent-report ($Z = 0.435, 95\% \text{ CI} = 0.340 - 0.530$) or with a composite measure of observation and parent-report ($Z = 0.157, \text{ CI} = 0.085 - 0.229$) but the association was not significant when BI was measured by observation alone ($Z = 0.057, 95\% \text{ CI} = -0.102 - 0.215$).

These differences in the overall effect as a function of how BI was measured were significant: there was a significant difference in the effect size when BI was measured by parent-report when compared to BI being measured by observation ($t(24, 7) = 4.375, p < .001$). Similarly, there was a significant difference in effect size when comparing BI as measured by parent-report to BI as measured using a composite measure of observation and parent report: ($t(34, 2) = -4.85, p < .001$).

Anxiety measures

The association between BI and anxiety was significant when anxiety was categorised either categorically ($Z = 0.153, 95\% \text{ CI} = 0.068 - 0.238$) or continuously ($Z = 0.306, 95\% \text{ CI} = 0.211 - 0.401$). However, overall effects differed significantly as a function of how anxiety was categorised. Effects were significantly larger for outcomes which had measured anxiety continuously than when anxiety was measured categorically: $t(29, 9) = -2.48, p = .02$.

The association between BI and anxiety was significant when anxiety was measured by parent-report ($Z = 0.313, 95\% \text{ CI} = 0.200 - 0.436$) and when anxiety was measured by clinical interview ($Z = 0.221, 95\% \text{ CI} = 0.112 - 0.330$). Whereas, the association between BI and anxiety was not significant when anxiety was measured by self-report ($Z = 0.087, 95\% \text{ CI} = -0.068 - 0.243$) or from combined parent- and child-reports ($Z = 0.161, 95\% \text{ CI} = -0.067 - 0.388$).

These differences in the overall effect as a function of how anxiety was measured were significant: there was a significant difference in the overall effect size when comparing parent-report and self-report measures of anxiety ($t(5, 19) = -2.86, p = .03$). There were no other significant differences in the strength of the association when comparing other ways in which anxiety was measured (i.e. clinical interview, combined child and parent reports).

Moderation by anxiety outcome

It was then examined whether there were effect size differences depending on the anxiety outcome. Given the small number of outcomes for the non-social anxiety subtypes (panic, separation anxiety, phobia, generalised anxiety, selective mutism), these outcomes were pooled together with the ‘any anxiety’ and ‘non-social anxiety’ outcomes. We collectively compared this ‘other anxiety’ composite to the social anxiety outcomes. There were significant associations between BI and social anxiety ($Z = 0.314, CI = 0.118 - 0.510$) and BI and other anxiety outcomes ($Z = 0.249, CI = 0.179 - .318$). However, the differences in these effect sizes were not significant: $t(20, 6) = 0.107, p = .91$, as illustrated in Figure 3.

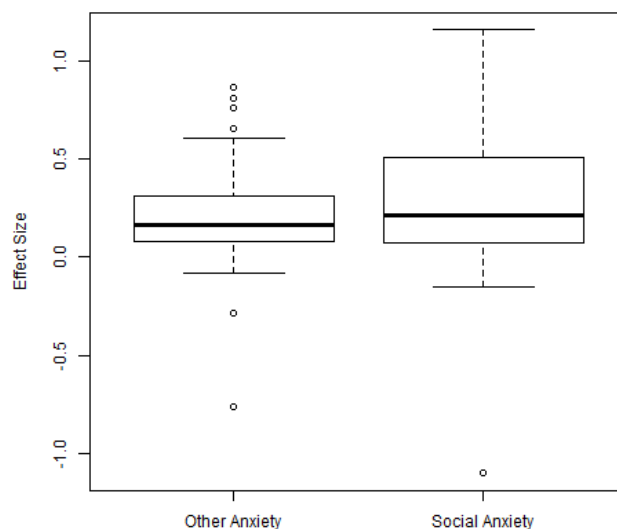


Figure 3. Box plot examining the differences in effect size between social anxiety outcomes and other (non-social) anxiety outcomes.

Publication bias

Visual inspection of the funnel plot (Figure 4), with symmetric distribution, and the outcome of the Egger test of asymmetry ($z = -0.896$, $p = .370$) did not suggest publication bias.

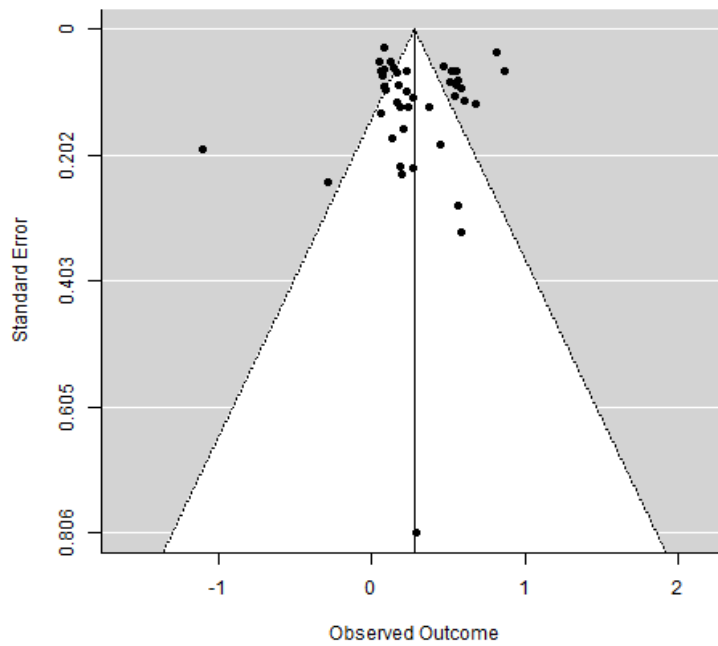


Figure 4. Funnel plot to visually assess for publication bias.

Note. Diagonal lines = 95% Confidence Interval, x-axis = magnitude of the weighted effect size for each sample, y-axis = standard error for each sample mean.

Discussion

This systematic review and meta-analysis aimed to quantify the strength and the significance of the association between BI and childhood anxiety. Previous meta-analyses examined the prospective association between BI and social anxiety disorder (Clauss & Blackford, 2012) and other anxiety subtypes (Sandstrom et al., 2019). The current meta-analysis also included studies using cross-sectional and retrospective designs in order to examine whether cross-sectional or retrospective associations of BI and child anxiety differ from their prospective association. Additionally, we set out to examine the effect of covariates on the strength of the relationship between BI and childhood anxiety, including how these constructs were defined (e.g. categorical or continuous) and measured (e.g. self-report, parent-report, clinical assessment), as well as investigating the differential associations between BI and different anxiety subtypes.

As hypothesised, we found an overall significant association between BI and childhood anxiety. This association was strongest for the social anxiety subtype, as previously demonstrated by Clauss & Blackford (2012) and Sandstrom et al. (2019), when compared to the other subtypes of anxiety, although this difference was not significant in our study. Genetics has been suggested as one possible explanation for the strong association between BI and social anxiety. Bourdon et al. (2019) conducted a twin-study to examine the genetic and environmental factors shared between childhood BI and anxiety disorders. They found that a significant proportion of genetic factors were shared between BI and social anxiety, whereas no other anxiety subtypes shared a significant proportion of genetic factors with BI. Alternatively, it has been argued that the strong association between BI and social anxiety could be accounted for by the conceptual overlap between behaviours/features present in both BI and social anxiety. Many assessments of BI use social contexts to measure this construct, e.g. response to a stranger. Dyson, Klein, Olino, Dougherty, and Durbin (2011) argue that BI is a heterogeneous construct that should be differentiated into social and non-social BI, which can explain

the differential outcomes and trajectories found in BI children. The cross-sectional study by Dyson et al. (2011) did not find a significant correlation between social and non-social BI. Additionally, the social and non-social BI outcomes were associated with distinct outcomes on the measures of anxiety symptoms and parent-reported temperament. Given this, it would be interesting for future research to employ a prospective design and to conduct both social and non-social BI assessments, in order to examine similarities and differences in these trajectories.

We found that there was a stronger association between BI and anxiety when anxiety was measured continuously (i.e. anxiety symptoms) than when anxiety was measured categorically (i.e. anxiety disorder diagnosis). Although a diagnosis of an anxiety disorder is considered a more robust measure within clinical practice than a continuous symptom measure, there are drawbacks of dichotomising variables within research. Altman and Royston (2006) explain that statistical power is lost through the use of dichotomous variables (i.e. meeting diagnostic criteria or not), resulting in a reduction in the statistical power to detect a relationship between the variables in question (i.e. BI and anxiety).

The inclusion in this meta-analysis of different study designs and the examination of covariates within the study design has also given rise to some novel findings. Here we were able to examine whether the strength of the association between BI and anxiety differed by study design. We did not find a significant difference when comparing the effect sizes between the prospective, cross-sectional and retrospective designs. There were substantially fewer retrospective study designs included in this meta-analysis so caution must be taken when making conclusions regarding this type of design. However, from these findings it is suggested that both prospective and cross-sectional designs can be considered valid means of examining the association between the constructs of BI and childhood anxiety.

We have also demonstrated (in the prospective studies) a significant effect of the time between BI and anxiety measurements. As the time between these two measurements increases, the strength of the association between BI and childhood anxiety decreases. This pattern was also shown by Clauss & Blackford (2012) in their meta-analysis examining the association between BI and social anxiety disorder. Hence, our findings demonstrate that this pattern, of a weakening association between BI and anxiety over time, extends beyond social anxiety to other types of anxiety presentations. These findings could suggest that the possible conceptual overlap of features associated with BI and anxiety symptoms diverges over time. Most often BI is measured in toddlerhood or early childhood and is reliant on behavioural observations, which would also be the case when anxiety is assessed in younger children. With increasing age children will develop skills in articulating their inner experiences and responding cognitively to anxiety-provoking situations, which may result in both greater accuracy in measuring anxiety symptoms as well as older children having more cognitive skills to rationalise and manage fears.

Our findings also showed a stronger association between BI and anxiety when BI was reported by parents, compared to BI being measured via observation or a compound measure of both parent-report and observation. This is consistent with the pattern of results of Chronis-Tuscano et al. (2009). In their longitudinal study of BI and childhood anxiety, children assessed as BI from parent-report (mothers only) were found to be twice as likely to meet the diagnostic criteria for social anxiety disorder in adolescence, than those who did not meet the criteria for BI from the parent-report measure. However, when BI was measured via observation or a composite measure from parent-report and observation a significant association was not found. Chronis-Tuscano et al. (2009) discuss the merits in non-research settings of parent-report measures of temperament as parents are able to provide information about their child in multiple contexts, in a cost- and time-effective way. However, they express caution of the sole use of parent-report measures of temperament within research settings, despite this only giving a limited sample of behaviour within an artificial context, due to the

risk of bias that comes from parental-report. This is particularly relevant in the case of assessing the constructs of BI and anxiety, which can present with similar behavioural features. Thus, our findings emphasise the importance of assessing BI in a way that is relevant for the context that the outcome is being applied to. BI was originally identified through observational methods and standardised observation methods have been developed from the work of Kagan and colleagues, e.g. the LAB-Tab (Planalp, Van Hulle, Gagne, & Goldsmith, 2017). Questionnaire parent-report measures for BI have since been developed, such as the BIQ which has been shown to be a reliable and valid measure of BI, which correlated well with behavioural observations of BI when completed by mothers and teachers (and slightly less well for fathers) (Bishop et al., 2003). A meta-analysis by Kostyrka-Allchorne et al. (2020) examined the predictive power of parent-rated questionnaire measures of infant temperament for psychopathology in childhood and adolescence, from 25 studies involving over 28 000 participants. The authors reported that there was an overall weak predictive power for later psychopathology from parent-report measures of temperament. However, BI measures were found to have a stronger predictive power for later psychopathology than measures of other temperamental constructs, and the authors emphasised the value of the use of widely accessible questionnaire-based measures for identifying at risk children who are likely to benefit from a preventative, early intervention, programme.

Another moderating factor of the effect size between BI and anxiety was how anxiety was measured. There was a significantly stronger association between BI and anxiety when anxiety was measured by parent-report than when anxiety was measured by self-report, the latter of which demonstrated a very weak association between BI and anxiety. Muris et al. (2003) compared the differences between adolescent- (11 – 15 years) and parent-reported BI and anxiety/depression symptoms (using the RCADS (Chorpita, Moffitt, & Gray, 2005)) and found only modest agreement between the parent and adolescent reports on all measures. Similarly, Baldwin and Dadds (2007) found low agreement between parent- and child- (8 – 13 years) reports when measuring anxiety

symptoms using the Multidimensional Anxiety Scale for Children (MASC) (Wei et al., 2014). Nauta et al. (2004), in a paper examining the psychometric properties of measures for child anxiety, discuss that *“in the assessment of childhood disorders, it is both common and recommended to include multiple informants, most commonly children, parents and teachers. Each informant may contribute information about different aspects of the disorder, thus complementing each other”* (p. 815). Hence, our findings and the findings from the empirical studies reported here call into question the accuracy of the use of only parent- or child-reports of childhood anxiety, and it is suggested that measures completed by multiple informants is a clinically more robust means of measuring anxiety in childhood.

A strength of this study is the large number of studies included in the quantitative synthesis. By including all study designs (prospective, cross-sectional and retrospective) we were able to capture the breadth of the literature quantitatively examining BI and childhood anxiety. This has allowed us to examine the difference between the prospective, cross-sectional, and retrospective associations between BI and anxiety in childhood. To our knowledge, this is the first time these differences have been examined quantitatively, although this has been debated theoretically in the literature. The findings we have presented contribute to the literature in understanding how the concepts of BI and childhood anxiety are related, as well as demonstrating factors that moderate this association.

Although a strength of this meta-analysis was the inclusion and synthesis of a large number of effect sizes, from studies covering a broad span of the research literature, this meant that the impact of heterogeneity was a potential limitation of this research. High heterogeneity was found in the meta-analysis, which suggests that the findings should be interpreted with caution and indicates the need to complete moderation analyses. However, it was considered appropriate to present our findings from the meta-analysis despite the high heterogeneity, given that moderation analyses were conducted in order to understand the heterogeneity. High heterogeneity was not unexpected given the range of anxiety outcomes being examined in the included studies, which were shown in

subsequent analyses to have significantly differing association strengths with BI. Additionally, the pooled effect size reported here is in line with the outcomes reported in comparable meta-analyses (e.g. Sandstrom et al., 2019; Clauss & Blackford, 2012) which included a much narrower range of studies.

An exclusion criterion for this meta-analysis was BI being measured by self-report. For the majority of studies this would not have been relevant as BI was most often measured in toddlerhood or early childhood. However, some cross-sectional studies of older children and adolescents were excluded on the basis of BI being self-reported. It would have been interesting to have examined whether self-reported BI differed from the other included types of BI measures in the strength of the association with the anxiety measures. There is debate in the literature about the accuracy of self-reported temperamental characteristics. However, there is a child-report version of the Behavioral Inhibition Questionnaire (BIQ) that has been shown to correlate with observational data and have good internal consistency and moderate agreement with parent-report versions (Broeren & Muris, 2010). The inclusion of self-report measures of anxiety allowed us to make some interesting observations regarding the differences in the association between BI and anxiety, depending on how anxiety had been measured. Thus, an interesting future direction would be to examine whether self-reported BI had a significant effect on the relationship between BI and anxiety.

We have investigated the association between a temperamental factor (BI) and presentations of childhood anxiety. However, longitudinal research has indicated that there is a more complex interactive relationship between BI and anxiety, whereby environmental and genetic factors moderate this relationship. Hudson and colleagues have been examining the predictors of childhood anxiety in a longitudinal study. They have consistently found children classified as BI at four years, when compared to behaviourally *uninhibited* children, are significantly more likely to be anxious, across early (Hudson, Dodd, Lyneham, & Bovopoulos, 2011), middle (Hudson & Dodd, 2012) and late

childhood into early adolescence (Hudson et al., 2019). However, parenting behaviours were found to moderate this relationship; with BI children (compared to behaviourally uninhibited children) having higher anxiety symptoms aged 11 (as measured by questionnaires and diagnostic interviews), only when the child had experienced maternal over-involvement at four years of age. We have seen in our meta-analysis that the strength of the association between BI and anxiety decreases as the time between these measures increases, and it would be interesting in the future to explore these interactive relationships. A comprehensive meta-analysis could incorporate a range of risk factors for childhood anxiety, such as temperamental factors (e.g. BI), parenting behaviours (e.g. overprotection, encouragement) and parental mental health (e.g. anxiety disorder diagnosis). This would allow us to understand the relative influence and interactive effects of these known risk factors in the development of childhood anxiety. It is important to note that caution must be taken when making conclusions regarding any research examining BI, given that this concept of BI was developed using limited sample characteristics and thus generalisations outside of these parameters cannot be made confidently.

It would also be interesting for future research to consider the resilience or protective factors relevant to childhood anxiety. Although BI has been clearly demonstrated, here and in previous research, as a risk factor for childhood anxiety, not all BI children go on to develop anxiety symptoms or disorders. Clauss & Blackford (2012) reported that almost half of the children with BI went onto develop social anxiety disorder, and the risks of developing other anxiety subtypes (e.g. generalised anxiety disorder) were even lower, as demonstrated here and by Sandstrom et al. (2019). Understanding the personality, parenting, and other environmental factors that mitigate the risks of BI would support the development of early-intervention programmes. A promising study in this area was presented by Rapee, Kennedy, Ingram, Edwards, & Sweeney (2005), in which a prevention programme for parents of pre-school BI children was found to result in a significant decrease in anxiety diagnoses one year later, when compared to a no intervention control group. This was a small-scale

study and it would be important for future research to consider how to deliver effective early-intervention programmes to target all children who could be at risk of developing anxiety problems.

In summary, this systematic review and meta-analysis is the first study, to our knowledge, to quantitatively examine the association between BI and childhood anxiety across all study designs. We found a significant relationship between BI and childhood anxiety, which was not significantly stronger in cross-sectional studies than prospective studies. The meta-analysis revealed some interesting new findings in regard to the study design factors which affected the relationship between BI and childhood anxiety, such as the way in which BI and anxiety were measured and whether anxiety was conceptualised as a categorical or continuous construct. Additionally, we have shown that the temporal relationship between BI and anxiety has a significant effect on the strength of the association between BI and anxiety. The findings from this meta-analysis also have some clinical implications. Assessing for BI at an early age appears to be a helpful indicator for later risk of developing anxiety in childhood and hence could be used as a means of screening infants to identify those likely to most require and benefit from a targeted early-intervention programme to mitigate this risk. It is suggested that future research examines the resilience factors associated with the development of childhood anxiety and that the interaction between temperament and other environmental factors is further explored.

Chapter 2: Effects of modification of maternal expressed anxiety and encouragement on infant affect and behaviour, using a social referencing paradigm.

Introduction

Childhood Anxiety

Anxiety disorders in childhood are a common problem, with increasing prevalence rates over time, and they can have long-term and detrimental consequences for the child's wellbeing, their future outcomes, and the wider family and community. One in eight (12.8%) five to 19-year-olds and one in 18 (5.5%) preschool children in England were found to have a mental disorder in 2017 (NHS Digital, 2018). These figures are in line with global prevalence rates, estimated from a global meta-analytic study, which reported a global prevalence rate of 13.8% for mental disorders in under 18s. Anxiety disorders were the most common disorders, with an estimated worldwide prevalence of 6.5% (Polanczyk et al., 2015).

Childhood anxiety disorders have been associated with negative educational outcomes, including more missed days at school (Creswell et al., 2015) as well as greater levels of school truanting and dropout, and lower levels of academic attainment (Public Health England, 2016). Longer term impacts of childhood anxiety disorders include lower earnings in adulthood (Public Health England, 2016) and greater costs for health and social care services (Creswell et al., 2015). Anxiety disorders in childhood have also been found to be associated with an increased risk of experiencing mental health problems in adulthood, including anxiety disorders (Essau et al., 2018), mood disorders, and substance misuse problems (Public Health England, 2016).

Parents of young children with anxiety disorders have been found to experience greater levels of worry, depression and fatigue, as well as difficulties in engaging with social activities and maintaining relationships, when compared to parents of children with other early childhood disorders (Towe-Goodman et al., 2014). Childhood anxiety disorders were also found to be associated with lost parent productivity (Creswell et al., 2015). For children, poorer levels of quality of life have been

shown to be associated with greater severity of anxiety symptoms (Ramsawh & Chavira, 2016) and a study of children with social phobia found that, compared to a control group without any mental health diagnoses, children with this anxiety disorder reported greater levels of loneliness, tearfulness, and feelings of dysphoria (Beidel, Turner, & Morris, 1999).

Development of Childhood Anxiety

Genetic, temperamental, and environmental factors have been implicated in the development of childhood anxiety. The literature suggests a complex picture in regard to the roles played by these different factors with genetic, temperamental, and environmental factors playing a greater or lesser role in the variability of childhood anxiety traits, depending on the age that anxiety is measured. There is research to suggest that environmental and temperamental factors have a stronger influence over the transmission of childhood anxiety than genetic factors. For example, in a children-of-twins study, Eley et al. (2015) found no evidence of significant genetic transmission of anxiety, but found evidence for significant environmental transmission of anxiety from parents to adolescent children. Additionally, Ahmadzadeh et al. (2019) conducted a study of adopted children (i.e. a context of no possible genetic transmission) and found that adoptive fathers' anxiety predicted their adopted child's anxiety symptoms at six to eight years of age.

The temperamental style of behavioural inhibition (BI) has been identified as a strong risk factor for the development of childhood anxiety disorders. The term and definition for BI comes from observational studies of young children conducted by Kagan et al. (1984); it describes a stable affective and behavioural pattern of fear, wariness, or reticence when facing unfamiliar situations (such as meeting a new person or seeing a novel object), and proximity seeking to familiar figures in such circumstances. A meta-analysis of prospective studies by Clauss and Blackford (2012) found that BI, compared to absence of BI, was associated with more than a sevenfold increase in the odds of

developing social anxiety disorder (SAD), even when controlling for the risk associated with parental psychopathology. More recently Sandstrom, Uher, & Pavlova (2019) published a meta-analysis examining the association between BI and all types of anxiety disorders. Early childhood BI was associated with an almost threefold increase in the odds of later developing an anxiety disorder in general, compared to the absence of BI.

The environmental factors that have been implicated in the development of childhood anxiety include parenting behaviours and parental anxiety disorders (e.g. Yap & Jorm, 2015; Moller, Nikolic, Majdandzic, & Bogles, 2016; Van der Bruggen et al., 2008; Lawrence, Creswell, Copper, & Murray, 2020). Murray, Creswell, & Cooper (2009), in a theoretical review of the literature, suggest that there is an interactive relationship between child temperament and parenting factors, whereby particular combinations of temperamental style and parenting behaviours have been found to be stronger predictors of childhood anxiety than temperament or parenting behaviours individually. A qualitative review by Ryan & Ollendick (2018) describes a conceptual model considering parenting as a moderating factor in the relationship between BI and childhood internalising problems, as well as temperament as a moderating factor between parenting behaviour and subsequent child anxiety.

Parenting Behaviour and Childhood Anxiety

Particular parenting behaviours have been implicated in the development of childhood anxiety disorders. A systematic review and meta-analysis conducted by Yap & Jorm (2015) examined the parenting factors associated with childhood anxiety, depression and internalising problems. They found evidence for an association between increased risk of childhood anxiety problems and each of inter-parental conflict, parental aversiveness (e.g. harsh criticism, parent-child conflict), parental over-involvement, and parental modelling of anxiety. Lower levels of childhood anxiety were associated with parental autonomy granting behaviours. Van der Bruggen et al. (2008) reported a meta-analysis

examining 23 studies, involving 1305 parent-child dyads, with a substantial association between child anxiety and parental control ($d = .58$).

Hudson and colleagues conducted a longitudinal study examining environmental risk factors for anxiety from pre-school age (3 – 4 years) (Hudson, Dodd, & Bovopoulos, 2011) to, most recently, early adolescence (Hudson, Murayama, Meteyard, Morris, & Dodd, 2019). At the first follow-up (two years after baseline measures) childhood anxiety at six years of age was found to be associated with the parent factors of maternal anxiety disorders and maternal over-involvement at baseline. In addition to these parent factors, the child having a temperament defined as behaviourally inhibited at age 3-4 years, compared to behaviourally uninhibited, was associated with increased risk for anxiety disorders at the age of six. The follow-up investigations of this longitudinal cohort suggested stability in the factors increasing the risk for childhood anxiety over time; higher levels of anxiety symptoms and disorders at the age of 12 were associated with the child being behaviourally inhibited, maternal anxiety disorder history, and (if present at the age of four) maternal over-involvement. An interesting interactive effect was reported by the researchers, whereby higher anxiety symptoms at age 12 in the behaviourally inhibited children were only found when maternal over-involvement was present when the child was four years old. Conversely, the behaviourally inhibited children who did not experience maternal over-involvement at the age of four years did not experience the same increased risk of anxiety at 12 years of age. These observational findings would be consistent with the hypothesis that modifications in parenting behaviours could decrease the risk of a child developing anxiety, even if the child's temperament would be expected to place them at greater risk of developing anxiety.

Experimental designs can provide strong evidence regarding the causal relationship between parenting behaviours and childhood anxiety disorders or symptoms. Rapee and colleagues reported on a long-term follow-up study investigating the efficacy of a prevention programme targeting children (3 – 5 years) who were identified as being at risk of developing anxiety or internalising

disorders on the basis of being assessed as temperamentally inhibited (analogous to BI) (Rapee, Kennedy, Ingram, Edwards, & Sweeney, 2005). The six-session group programme for parents (*Cool Little Kids*) was compared to a no intervention control. The sessions provided psychoeducation about anxiety and taught skills parents could use with their child to reduce the child's anxiety, including reducing unhelpful parenting strategies and helping the child to become more independent (i.e. increasing autonomy granting and reducing over-involvement). Data was collected one year after attendance at the programme and it was found that children whose parents attended the programme showed a significant decrease in anxiety diagnoses when compared to children whose parents were in the control condition. Thus, it is suggested from these findings that modifying parenting behaviours experienced by children at risk of developing anxiety disorders could play an important role in effectively reducing this risk. The participants of this programme were followed up eleven years later (mean age 15.4 years) and it was found that girls whose parents had attended the programme had significantly fewer internalising disorders, as well as significantly lower life interference, as compared to controls. However, this was only the case for girls; boys whose parents had attended the prevention programme did not show differences in internalising diagnoses or symptoms when compared to controls in mid-adolescence. The authors cautioned drawing strong conclusions about these gender differences due to the relatively small sample size and the mechanisms underlying these gender differences are unclear (Rapee, 2013).

The above literature presents a consistent finding of parental control and over-involvement being associated with increased risk of a child developing anxiety disorders. Rapee (2001) suggests that these parenting behaviours convey the message to the child of the world being dangerous and reduce opportunities for the child to develop skills for independent coping, instead reinforcing dependence.

Parenting Behaviours in Parents with Anxiety Disorders

Research shows that parents with anxiety disorders may behave differently towards their children than parents without anxiety disorders when faced with anxiety provoking situations (e.g. interacting with a stranger). It has been suggested that parental modelling of affect and behaviour plays a role in both the development and maintenance of childhood anxiety (and could help account for the increased risk of anxiety disorders in children of parents with anxiety disorders, compared to children whose parents do not have anxiety disorders), sometimes referred to as the intergenerational transmission of anxiety.

For example, Creswell, Apetroaia, Murray, & Cooper (2013) compared the behavioural parenting characteristics of mothers with and without anxiety disorders when interacting with their children (7 – 12 years), all of whom had anxiety disorders. The mothers who had anxiety disorder diagnoses differed to the non-anxious mothers in observations of intrusiveness, expressed anxiety, warmth, and the quality of the relationship, as well as self-reporting more negative emotions and more negative expectations of their child's performance in challenging tasks. From this, the authors suggested that maternal anxiety disorders may interfere with the maintenance of a positive and supportive mother-child interaction when facing stressful circumstances, and that these behaviours could be specifically targeted within treatment programmes for childhood anxiety.

Murray et al. (2012) examined the parenting behaviour of mothers with social anxiety disorder (SAD) or generalised anxiety disorder (GAD) when faced with disorder-specific stressors (child giving a speech or exploring a potentially scary object), which was compared to their behaviour in a non-threatening task. Mothers with SAD showed less encouragement and warmth, and more passivity and expressions of anxiety only when faced with a socially challenging situation. Whereas mothers with GAD showed less encouragement, warmth, and positive modelling, and greater passivity and

avoidance only when faced with the situation designed to elicit the core features of GAD (uncertainty and worry).

The findings from these studies suggest that the parenting behaviours that have been identified as increasing the risk of childhood anxiety are more often observed in parents with anxiety disorders, particularly when they are in a situation that is considered threatening within the context of their particular subtype of anxiety disorder.

Parenting Behaviours in Infancy

The impact of parenting behaviours implicated in the development of childhood anxiety have been evidenced from early in the post-natal year. Murray, Cooper, Creswell, Schofield, and Sack (2007) studied 10-week-old infants of mothers with SAD in a socially challenging situation of meeting a stranger, comparing the mothers' behaviour and infant responses to dyads where mothers did not have an anxiety disorder. Mothers with SAD engaged less with the stranger, were less encouraging of the infant's interaction with the stranger, and the infants subsequently showed less social responsiveness to the stranger. A similar finding was reported by Stein et al. (2012) in a study investigating mother-infant relationships at 10-months in groups of mothers with GAD, mothers with major depressive disorder, and mothers with no current or historical psychiatric disorder. When the mothers were primed to experience worry and rumination, those with GAD were less responsive to their infant's vocalisations than those without GAD, and these infants demonstrated more withdrawal and lower emotional tone than infants of mothers without GAD.

Infants' skills in using social referencing (SR) (i.e. making use of adults' emotional signals to determine how to respond when facing unfamiliar situations, such as meeting strangers or seeing new objects) develop around ten months of age (Feinman, 1982). Studies have been conducted with infants around this age to investigate the role of SR in learning anxious responses from parents. For

example, Murray et al. (2008) used a social referencing paradigm to compare mother and infant affect and behaviour between socially anxious mothers and a non-anxious control group, when the infants were 10-, and then 14-, months. Infants of mothers with social phobia showed increased avoidance of a stranger over time; this avoidance was predicted by maternal expressed anxiety and lower levels of encouragement to interact with the stranger. Infant avoidance was found to be even more pronounced if the infant's temperamental style was behaviourally inhibited.

Modification of Parenting Behaviour

Studies have shown that it is possible to modify parenting behaviours implicated in the development of childhood anxiety in non-clinical samples and that this, in turn, has an impact on children's anxiety. Using an experimental design with seven to 13 year olds and their non-anxious mothers, De Wilde & Rapee (2008) demonstrated that maternal control could be manipulated through explicit instruction. Mothers were allocated to one of two conditions; they were verbally instructed to be overly controlling or minimally controlling while their child prepared a speech. The child was then tasked with preparing a second speech alone – it was found that those who had experienced the maternal over-control condition demonstrated greater anxiety than those who had experienced the minimal control condition. The children in this study were a non-clinical sample and it is suggested from these findings that even in a single interaction a child's anxiety can be influenced by parental control.

Thirwall & Creswell (2010) extended the pilot study from De Wilde & Rapee (2008) using a sample of younger children (four and five years old). This study involved two experimental conditions – maternal controlling behaviours and maternal autonomy granting behaviours. Non-anxious mothers were shown training videos to demonstrate the types of behaviours they were to present in each condition. The child was then tasked with preparing a speech with their mother and then performing

the speech to camera - both experimental conditions were undertaken by each mother-child dyad, the order of which was counter-balanced. Maternal controlling behaviour was associated with the child having more negative predictions about the task and feeling less happy about the task. These findings provide more evidence for controlling parenting as a risk factor for the development of childhood anxiety as well as evidence that this behaviour can be modified through parent training using video tuition.

Gerull & Rapee (2002) demonstrated the modification of maternal modelling of anxiety in a sample of toddlers (15 to 20 months) and their non-anxious mothers. Mothers were instructed to show either a positive or negative (fear and disgust) facial expression while the toddler was shown a potentially anxiety provoking object. The children were shown these objects again after ten minutes and on this occasion the mother was instructed to show a neutral expression. Infants showed increased expressions of fear and avoidance when presented with the object to which their mother had previously reacted negatively.

Modification of maternal behaviour using video tuition has also been demonstrated in a study involving mother-infant (12 to 14 months) dyads (de Rosnay, Cooper, Tsigaras, & Murray, 2006). In this study mothers (without social anxiety disorder) were shown a training video of socially anxious and non-socially anxious interactions with a stranger before undertaking social referencing tasks with their infant. The training videos demonstrated the desired behaviour for the mother to present in the two experimental conditions (socially anxious and non-anxious) when talking to a stranger in front of their infant. The infants' responses to the strangers were compared between these two conditions. In the socially anxious condition infants were significantly more fearful and avoidant with the stranger when compared to the non-anxious condition. This effect was even more marked in the infants whose temperament was categorised as 'high fear' (analogous to BI); whereby high fear infants demonstrated even greater avoidance of the stranger in the socially anxious condition. The findings

from this study provide evidence for the efficacy of training videos for the modification of maternal anxiogenic behaviour, as well as demonstrating the impact of maternal anxious behaviour on infant affect and behaviour, particularly when infants are temperamentally fearful.

More recently, research has shown that parenting behaviours implicated in childhood anxiety can be modified within clinical populations. Ewing et al. (2019) successfully demonstrated the efficacy of a short video tutorial for parents to support their child (aged between five and nine years) to overcome fears (e.g. snakes, beetles). Parenting behaviours implicated in overcoming fears (e.g. encouraging and reinforcing children's approach behaviour) were shown to parents in a ten-minute video tutorial before supporting their child to face the feared stimuli, which was compared to a control video. This randomised-controlled trial included both clinical (confirmed anxiety diagnoses) and non-clinical parent populations. The video tutorial was found to be equally effective in modifying parenting behaviour across both groups of parents, whereby parents who had watched the video tutorial showed increased behaviours associated with children overcoming anxiety (i.e. encouraging approach) and decreased behaviours associated with maintenance of childhood anxiety (i.e. encouraging avoidance), as compared to parents who watched the control video. The increase in parenting behaviours encouraging approach and the decrease in behaviours encouraging avoidance were associated with increased child approach towards the feared object. Child anxiety did not moderate the impact of the video tutorial on parenting behaviours and thus the use of video tutorials targeting parenting behaviour implicated in childhood anxiety appears to be a useful tool for overcoming childhood phobias (Ewing et al., 2019).

The Current Study

The research outlined here illustrates the importance of maternal behaviours in the development of childhood anxiety and, in particular, maternal expressed anxiety and encouragement

in social referencing tasks in infancy. Additionally, the current literature suggests that infant temperament (in particular BI) moderates the strength of the association between parenting behaviours and childhood anxiety. Video tuition has also been shown to be an effective tool in modifying maternal behaviours implicated in infant anxiety development.

What has not yet been examined is whether maternal behaviours associated with lower levels of infant anxiety (i.e. encouragement) can be modified during infancy, whether video guidance can be effective in modifying these maternal behaviours during infancy, and whether successful modification would be associated with changes in infant affect and behaviours.

This paper reports on an experimental study with a non-clinical sample of mothers with 10 to 14-month infants, examining; the effect of training videos on maternal behaviours in a social interaction implicated in infant anxiety, whether maternal behaviours had an impact on infant affect and behaviour, and whether infant temperament moderated the relationship between maternal behaviours and infant affect and behaviour.

Aims. The aims of this study are; i) to test whether maternal expressed anxiety and encouragement can be modified in mothers of infants between 10 and 14 months using a training video. If the mothers' behaviour is significantly different in the anxious and encouraging conditions after watching the training videos, the second aim is: ii) to examine whether modifications in parenting behaviour (i.e. expressed anxiety and encouragement in interactions with a stranger) are associated with infant affect and infant behaviour.

Hypotheses.

i) we will successfully modify maternal behaviours implicated in the development of childhood anxiety (i.e. expressed anxiety and encouragement) using specific video training with non-

anxious mothers, as demonstrated in the De Wilde & Rapee (2008), Ewing et al. (2019) and Thirwall & Creswell (2010) studies,

ii) maternal expressed anxiety during an interaction with a stranger will have an impact on the affect and behaviour of 10 to 14 month infants. It is predicted that infants will display more signs of anxiety and be emotionally less positive towards the stranger when mothers express anxiety, than when mothers use encouragement behaviours.

iii) the impact of maternal expressed anxiety (during an interaction with a stranger) on infant anxiety will be moderated by infant temperament, whereby infants with a negative temperamental style analogous to BI (e.g. high fear, negative affect), compared to infants with a less negative temperamental style, are predicted to demonstrate more anxious behaviours in the condition of maternal expressed anxiety.

Method

Ethics

Ethical approval to carry out this study was granted by the University of Southampton's Research Integrity and Governance Committee (ERGO ID: 47178).

Participants

Mother-infant dyads were recruited through poster advertisement in local nurseries and baby/toddler groups in the south of England and through word of mouth. Twenty-two infants between 10 and 14 months of age took part in the study with their mothers. Data from 19 mother-infant dyads was included in the analysis (12 female, 7 male); there were two incomplete data sets due to the infant becoming too distressed to complete the study tasks in the second condition and one infant was highly distressed throughout both study tasks and this data set was excluded from the analysis. Of note, all three of the excluded data sets were from monozygotic twin pairs. One twin from a monozygotic twin pair and both twins from a dizygotic twin pair were able to complete the study successfully.

All mothers were over 18 years ($M = 33.95$, $SD = 4.54$, range = 28 - 43) and English speaking. All infants met the recruitment criteria of being born full term (37 weeks or later) with no additional needs (e.g. developmental delays, congenital health conditions).

Design and Procedure

The mother-infant dyads participated in the social referencing tasks in a research laboratory in a university psychology department. Prior to attending the university to participate in the study tasks the mothers completed an online survey (described below) comprising questions about their anxiety (Penn State Worry Questionnaire; Meyer, Miller, Metzger, & Borkovec, 1990, and Social

Phobia Inventory; Conner et al., 2000) and their infant's temperament (Infant Behavior Questionnaire – Revised version, very short form (IBQ-R VSF) (Putnam, Helbig, Gartstein, Rothbart, & Leerkes, 2014)). At the university laboratory, mothers completed a paper questionnaire about their mood (described below) and the consent form.

The experiment comprised two conditions: 'anxious' and 'encouraging'. This study used a within-subjects design, with all mother-infant dyads experiencing both conditions. The order of the conditions was counter-balanced across the sample to minimise order effects.

At the university laboratory, mothers were told which condition of the social referencing tasks they would be participating in first (anxious or encouraging) and then shown a two-minute instructional video demonstrating the behaviour for her to display during the task – the infant sat with their mother while the videos were being shown. The video was watched jointly with the experimenter; once on mute to encourage mothers to focus on non-verbal features such as facial expressions, eye contact, and gestures and then repeated with sound to hear how the mother was responding verbally to the stranger. The experimenter drew the mothers' attention to salient aspects of the instructional videos (e.g. little eye contact in the anxious condition; smiling and open gestures in the encouraging condition).

After watching the instructional video for the first condition the mother-infant dyad were participating in, the experimental tasks began. The infant was secured into a highchair that was positioned between two arm-chairs facing each other (3 metres apart). In part 1, the mother and infant were alone in the room together for approximately 30 seconds before the stranger entered the room and initiated conversation with the mother for 90 seconds – during this time the stranger and mother did not look to the infant. The questions asked by the stranger were about the infant and the family e.g., 'what are their favourite toys?', 'do they have any brothers or sisters at home?'. The experimenter, standing out of sight of the infant and mother (behind a screen), prompted the stranger

and the mother after 90 seconds to swap chairs. For part 2, (the next 60 seconds) the mother looked at a laptop, so as to not engage with their infant, and the stranger attempted to engage the infant by speaking to them, saying their name, and showing and offering them a soft toy animal. The experimenter (remaining out of sight) prompted at the end of the 60 seconds and the stranger ended the interaction by reaching out their arm to touch the infant. The procedure was repeated for the second experimental condition but with a different stranger.

After completing the experimental tasks the two strangers returned to meet the mother-infant dyad. They behaved in a calm and friendly way with the infants and their mother, to foster a more natural and positive interaction to end the experience of participating in the study.

Materials and Measures

Maternal Anxiety. Maternal worry was measured using the Penn State Worry Questionnaire (PSWQ) (Meyer, Miller, Metzger, & Borkovec, 1990); a 16-item validated and reliable self-report measure of worry for use in adult populations. Scores on the PSWQ range from 16 - 80, with scores over 50 being considered indicative of generalised anxiety disorder (Brown, Antony, & Barlow, 1992).

Maternal social anxiety was measured using the Social Phobia Inventory (SPIN); a 17-item validated and reliable self-report measure of social anxiety for use in adult populations. Scores range from 0 – 68, with scores of 19 and above being considered indicative of social anxiety disorder (Connor et al., 2000).

Maternal Mood. Maternal mood was measured using the Edinburgh Postnatal Depression Scale (EPDS); a ten-item self-report measure of symptoms of postnatal depression that is widely used in studies of postnatal infant development. The score range on the EPDS is 0 – 30, with likely depression considered for scores above 12 or 13 (Cox, Holden, & Sagovsky, 1987).

Infant Temperament. Infant temperament was measured using the very short form of the Infant Behavior Questionnaire – Revised version (IBQ-R VSF) (Putnam et al., 2014). This 37-item parent-report questionnaire produces three scale scores: i) Surgency (e.g. ‘when tossed around playfully how often did the baby laugh?’, ‘how often during the week did your baby move quickly toward new objects?’), ii) Negative Affect (e.g. ‘when introduced to an unfamiliar adult, how often did the baby refuse to go to the unfamiliar person?’, ‘when introduced to an unfamiliar adult, how often did the baby cling to a parent?’), and iii) Effortful Control (e.g. ‘when showing the baby something to look at, how often did s/he soothe immediately?’, ‘how often during the last week did the baby enjoy gentle rhythmic activities, such as rocking or swaying?’). We used the Negative Affect scale scores, because its items consider infant behaviour associated with Behavioural Inhibition (BI), such as wariness with strangers, and clinginess with parents. This ‘negative affect’ scale will henceforth be referred to as ‘negative temperament’ for clarity. This very short-form version of the measure was chosen in order to reduce the burden on the participants, whilst allowing us to gain an indication of the infants’ temperament and their propensity to temperamental features analogous to BI.

Strangers. The participants met two strangers during the study – one in each experimental condition. The strangers used were always two white males of average build and height.

Instructional Videos. Two instructional videos were produced to demonstrate the desired behaviour for the mother to display during each condition. The video showed a mother (white female) with her infant (both on screen) speaking to a male stranger (off screen). The mother shown in the video was not involved in the experimental tasks. The stranger spoke to the mother in the same manner in each condition and asked the same questions. The difference between the conditions was how the mother interacted with the stranger.

In the encouraging condition the mother presented as relaxed and enjoying the conversation. Non-verbally, she was observed to smile at the stranger, engage in typical levels of eye-contact, use

emphatic gestures as she spoke, and have an open body posture. Verbally, she answered the stranger's questions with ease, expanded on her answers, and her tone of voice and intonation sounded relaxed.

In the anxious condition the mother presented as uncomfortable throughout the interaction with the stranger. Non-verbally, she was observed to engage in minimal eye-contact (mostly looking down to the floor), she did not smile often, and her body language conveyed an anxious presentation, e.g. wringing of hands, fidgety with hands and arms, and stiff posture. Verbally, the mother provided minimal responses to the stranger's questions (e.g. a one-word answer or short utterance), she did not attempt to continue the conversation, resulting in lengthy pauses and silences. Her tone of voice and intonation sounded tense and unrelaxed. The behaviour demonstrated in the video for the anxious condition was based on clinical descriptions of social anxiety (Bates, 2006) and from the author's experience of working in therapeutic settings with clients with a diagnosis of Social Anxiety Disorder.

Video Coding. The video recordings were coded using the coding schemes used in the social referencing paradigm studies by de Rosnay et al. (2006), Murray et al. (2008) and Aktar et al. (2013; 2014). The coding schemes provided detailed descriptions for the observed behaviour of the infants and the mothers, in order to rate the observed behaviour on a 1 – 5 point scale. The behavioural observations of the mother were: maternal expressed anxiety and maternal expressed encouragement, which were observed in part 1 when the mother was speaking to the stranger. The behavioural observations of the infant were: fear, avoidance, positive emotional tone, and negative emotional tone, which were observed in part 2 when the stranger was interacting with the infant. A count of the number of times the infant looked to their mother in part 1 and in part 2 was also taken from the video recordings.

The videos were coded by two raters who were blind to the experimental condition, to minimise expectation bias. The experimenter double-coded 30% of the video recordings as a means of monitoring inter-rater reliability. Inter-rater reliability was substantial: weighted- $\kappa = 0.689$ (Landis & Koch, 1977).

Maternal Expressed Anxiety. To examine the efficacy of the training videos a five-point scale was used to rate maternal expressed anxiety, including facial expressions of anxiety (e.g. anxious expression, frozen face, grimaces) and body expressions of anxiety (e.g. rigid posture, hand wringing) and verbal expressions of anxiety (e.g. agitated speech, highly constrained speech, nervous laughter, rushed manner of speech).

Maternal Expressed Encouragement. To examine the efficacy of the training videos a five-point scale was used to rate maternal encouragement, including positive facial expressions (e.g. smiling), positive gestures (e.g. nodding, open gestures), positive body language (e.g. open posture, leaning towards the stranger) and positive speech utterances.

Infant Behaviour. The infants' looks to their mothers were counted in part 1 and in part 2. For part 1 the tally of looks from infant to mother was firstly to ensure that they had observed their mothers' behaviour in each condition. The number of looks from infant to mother in parts 1 and 2 was also considered a rudimentary indication of infant anxiety, with a greater number of looks being expected when the infant felt more anxious about the situation and hence looking for more reassurance from their mother.

As a measure of infant anxiety, the infants' behaviour when interacting with the strangers was coded on five-point scales for: i) infant fear (e.g. facial expressions of wariness and fear, postural behaviours such as freezing), ii) avoidance of stranger (e.g. attempts to avoid contact with the stranger through gaze avoidance, physically withdrawing or turning away); iii) positive emotional tone (e.g.

expressions of happiness or contentment) and iv) negative emotional tone (e.g. expressions of distress or sadness).

Results

All of the data analysis was conducted in R (version 3.5.2).

Maternal Mental Health

Maternal Mood

The mean score on the EPDS was 5.84 (SD = 4.47, range = 1 – 20 (maximum possible range = 0 – 30)). The distribution of the EPDS scores was highly positively skewed (see Appendix F); one participant scored in the clinical range indicative of likely depression and all other participants scored below the range considered indicative of likely depression (12 and below).

Maternal Social Anxiety

The mean score on the SPIN was 14.21 (SD = 10.43, range = 3 – 36 (maximum possible range = 0 – 68)). There was a positive skew in the distribution of scores from this measure (see Appendix F), with most participants scoring at or below the clinical cut-off for social anxiety disorder.

Maternal Worry

The mean score on the PSWQ was 43.00 (SD = 13.23, range = 18 – 72 (maximum possible range = 16 – 80)), which is below the suggested clinical cut-off score for GAD of 50 (Brown et al., 1992). The scores on the PSWQ showed a normal distribution (see Appendix F) with most participants scoring below the clinical cut-off for GAD.

Maternal Affect in Experimental Conditions

To determine the efficacy of the training videos for the two experimental conditions (anxious and encouraging) expressed maternal anxiety and encouragement were coded from part 1 of the

video data (stranger speaking to the mother, in view of their infant). Paired *t*-tests were conducted to examine the differences in maternal expressed anxiety and encouragement in the two conditions.

The mean rating (from 1 – 5 scale) of maternal expressed anxiety in the anxious condition was 3.895 and the mean rating of maternal expressed anxiety in the encouraging condition was 1.737. This difference was highly significant: $t(18) = 8.808$, $p < .0001$, with a very large effect size: $d = 2.564$.

The mean rating (from 1 – 5 scale) of maternal expressed encouragement in the encouragement condition was 4.053 and the mean rating of maternal expressed encouragement in the anxious condition was 1.737. This difference was also highly significant: $t(18) = 10.065$, $p < .0001$, with a very large effect size: $d = 3.213$.

Infant Looks to Mother

In part 1 (stranger speaking to mother with infant watching for 90 seconds) all infants looked in the direction of their mother's face at least twice. In the anxious condition the mean number of looks (from infant to mother) was 7.789 (SD = 3.852, range = 2 – 17). In the encouraging condition the mean number of looks was 7.578 (SD = 3.849, range = 4 – 15).

The order of the experimental condition was counter-balanced across the sample. To examine whether order influenced the number of times the infant looked to their mother in part 1, a repeated measures ANOVA was conducted (order as a between-subjects factor and condition as within-subjects factor). There was no main effect of order on the number of times the infant looked to their mother in part 1: $F(1, 17) = 0.536$, $p = .474$, nor was there a main effect of condition: $F(1, 17) = 0.078$, $p = .783$. There was no interaction between order and condition: $F(1, 17) = 0.024$, $p = .879$.

A second ANOVA was conducted to examine whether infant gender or negative temperament (as measured in the IBQ-R VSF, parent-report measure) influenced the number of looks from infant to

mother in part 1 (with gender and infant negative affect as between-subjects factors and condition as within-subjects factor). There was no main effect of infant gender ($F(1, 15) = 2.676, p = .123$) or infant negative affect ($F(1, 15) = 1.130, p = .305$) on infant looks to mother in part 1. All two-way interactions were considered and there were no significant interactions.

In part 2 (stranger interacting with infant for 60 seconds) the number of looks from the infant towards their mother's face was also counted. In part 2 not all infants looked to their mothers. In the anxious condition the mean number of looks was 5.105 ($SD = 5.216, range = 0 - 22$). In the encouraging condition the mean number of looks was 4.211 ($SD = 3.706, range = 0 - 12$). Again, it was first examined whether the order of the experimental conditions influenced the number of times the infant looked to their mother. A repeated measures ANOVA was conducted (order as a between-subjects factor and condition as within-subjects factor). There was no main effect of order on the number of times the infant looked to their mother in part 2: $F(1, 17) = 0.003, p = .957$, nor was there a main effect of condition: $F(1, 17) = 0.489, p = .503$. There was no interaction between order and condition: $F(1, 17) = 1.870, p = .189$.

An additional ANOVA was conducted to examine whether infant gender or negative temperament influenced the number of looks from the infant to their mother in part 2. There was a main effect of gender ($F(1, 15) = 4.768, p = .045$), whereby in part 2 female infants looked to their mothers significantly more times (mean number of looks = 5.792) than male infants looked to their mothers (mean number of looks = 2.714), irrespective of the experimental condition. The effect size for this difference was medium ($d = 0.718$). There were no significant two-way interactions.

The main effect of infant negative temperament was nearing significance ($F(1, 15) = 4.488, p = .051$). Post-hoc analysis indicated a positive trend between higher scores for infant negative affect and greater number of looks in part 2 (see Figure 5). Examination of Figure 5 identified a potential outlier (number of looks > 20); regression analysis was re-run without this data point and a significant

association was found between infant negative temperament and the number of looks to mother ($F(1, 35) = 11.00, p = .002, R^2_{adj} = 0.217$).

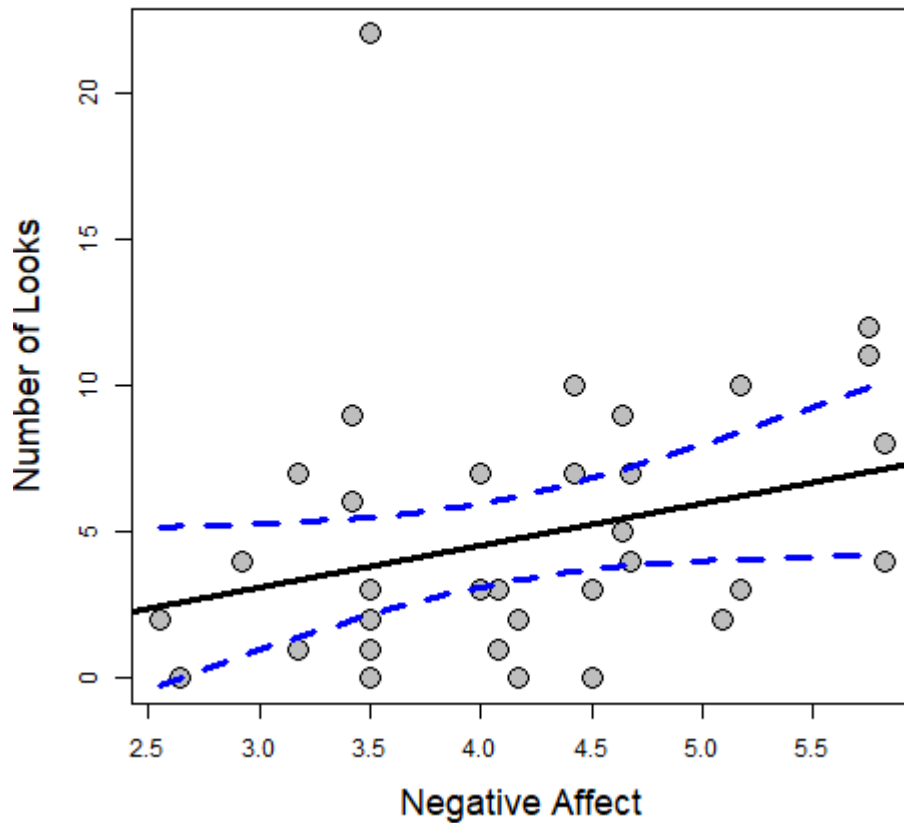


Figure 5. Linear regression (black line) between infant negative temperament (measured by the IBQ-R VSF) and number of looks from infant to mother in part 2.

Note. $F(1, 36) = 3.756, p = .060, R^2_{adj} = 0.069$. Blue dashed lines indicate 95% confidence interval.

Infant Affect

Order of Experimental Conditions

Repeated measures ANOVAs were conducted for each dimension of infant affect measured (fearfulness, avoidance, positive emotional tone, and negative emotional tone). For each infant affect dimension, first an ANOVA was conducted to test if the order of the experimental conditions

influenced infant affect (order between-subjects factor and condition within-subjects factor). There was no indication that infant affect was influenced by the order of the experimental conditions, see Table 1.

Table 1

ANOVA to Explore if Order of the Experimental Conditions Influenced Infant Affect

Infant Affect Dimension	<i>df</i>	<i>F</i>	<i>p</i>
Fearfulness	1, 32	0.000	.983
Avoidance	1, 32	0.022	.884
Positive Emotional Tone	1, 32	0.008	.931
Negative Emotional Tone	1, 32	0.012	.909

Infant Fearfulness

The first repeated measures ANOVA for this infant affect dimension explored whether experimental condition influenced infant fearfulness. No main effect of condition was found ($F(1, 32) = 0.000, p = .983$). There was no significant interaction between condition and order ($F(1, 32) = 2.375, p = .722$).

A second ANOVA explored the influence of gender, infant negative temperament (measured by the IBQ-R VSF), and underlying maternal social anxiety (measured by the SPIN) on infant fearfulness (condition as within-subjects factor and gender, infant negative affect, and maternal social anxiety as between-subjects factors). There was no main effect of gender ($F(1, 20) = 2.831, p = .108$), nor was there a main effect of maternal social anxiety ($F(1, 20) = 0.033, p = .858$). There was a main effect of infant negative temperament: $F(1, 20) = 9.820, p = .005$. Post-hoc regression analysis was conducted to examine this main effect (see Figure 6); higher scores for infant negative temperament were significantly associated with higher scores of fearfulness. All two-way interactions were considered and no significant interactions were found.

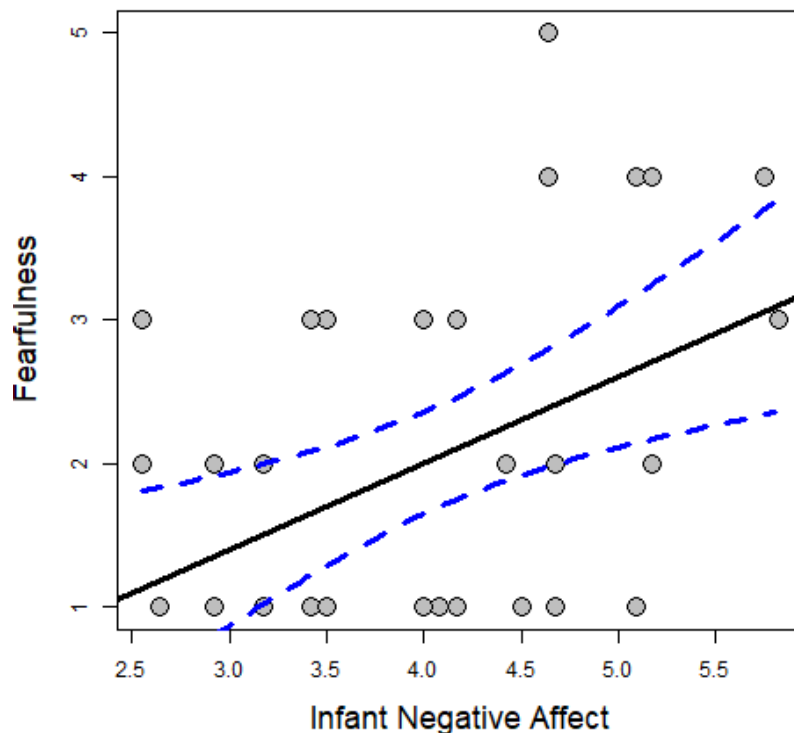


Figure 6. Linear regression (black line) between infant negative affect (measured by the IBQ-R VSF) and infant fearfulness.

Note. $F(1, 36) = 10.64, p = .002, R^2_{adj} = 0.207$. Blue dashed lines represent 95% confidence intervals.

Infant Avoidance

An ANOVA was conducted to explore whether the experimental condition influenced infant avoidance. No main effect of condition was found ($F(1, 32) = 0.067, p = .789$). No significant interaction between condition and order was found ($F(1, 32) = 0.053, p = .819$).

The second ANOVA explored the influence of gender, infant negative temperament (measured by the IBQ-R VSF), and underlying maternal social anxiety (measured by the SPIN) on infant avoidance (condition as within-subjects factor and gender, infant negative affect, and maternal social anxiety as between-subjects factors). There was no main effect of gender ($F(1, 20) = 0.790, p = .385$), maternal social anxiety ($F(1, 20) = 0.352, p = .560$) or infant negative temperament ($F(1, 20) = .388, p = .540$). All two-way interactions were considered and no significant interactions were found.

Infant Positive Emotional Tone

An ANOVA was conducted to explore whether the experimental condition influenced infant positive emotional tone. No main effect of condition was found ($F(1, 32) = 0.017, p = .898$). No significant interaction between condition and order was found ($F(1, 32) = 0.543, p = .467$).

The second ANOVA explored the influence of gender, infant negative temperament (measured by the IBQ-R VSF), and underlying maternal social anxiety (measured by the SPIN) on infant positive emotional tone (condition as within-subjects factor and gender, infant negative affect, and maternal social anxiety as between-subjects factors). There was no main effect of infant negative temperament ($F(1, 20) = 3.465, p = .077$), nor was there a main effect of maternal social anxiety ($F(1, 20) = 1.205, p = .285$). There was a main effect of gender ($F(1, 20) = 6.633, p = .018$). For male infants the mean score for positive emotional tone was 2.857 and for female infants the mean score was 1.792.

Infant Negative Emotional Tone

An ANOVA was conducted to explore whether the experimental condition influenced infant negative emotional tone. No main effect of condition was found ($F(1, 32) = 0.217, p = .644$). No significant interaction between condition and order was found ($F(1, 32) = 0.556, p = .461$).

The second ANOVA explored the influence of gender, infant negative temperament (measured by the IBQ-R VSF), and maternal social anxiety (measured by the SPIN) on infant negative emotional tone (condition as within-subjects factor and gender, infant negative affect, and maternal social anxiety as between-subjects factors). There was no main effect of gender ($F(1, 20) = 1.800, p = .195$), maternal social anxiety ($F(1, 20) = 0.461, p = .505$), or infant negative temperament ($F(1, 20) = 3.694, p = .069$). All two-way interactions were considered and no significant interactions were found.

Discussion

This study set out to examine, first; whether maternal expressed anxiety and encouragement can be modified in mothers of 10 to 14-month-old infants via a training video and, second; whether such modifications in parenting behaviour had an impact on infant affect and behaviour.

Previous research by de Rosnay et al. (2006) demonstrated that video training was effective in modifying maternal expressed anxiety in mothers, which was compared to mothers expressing 'neutral' behaviour in a social referencing paradigm. As well as higher levels of expressed anxiety; lower levels of parental encouragement have also been identified as a parenting behaviour that is associated with parental anxiety and, subsequently, childhood anxiety (e.g. Murray et al., 2012; Lawrence, Creswell, Cooper, & Murray, 2020). Therefore, this study aimed to complement and expand on the findings from de Rosnay et al. (2006) by examining whether maternal encouragement behaviours could also be modified by video training, and then to compare infant affect and behaviour in response to maternal expressed anxiety and encouragement.

Here we show, as expected, the training videos for maternal expressed anxiety and encouragement were effective in modifying maternal behaviour on both the dimensions of expressed anxiety and encouragement. Mothers acted to demonstrate significantly greater levels of anxiety in the anxious condition and, conversely, mothers acted to demonstrate significantly greater levels of encouragement in the encouragement condition. The order of the experimental conditions the infants experienced did not have an impact on any of the infant affect and behaviour outcomes measured. Thus, the two experimental conditions the infants were exposed to were significantly different in regards to their mothers' behaviour and therefore any main effects of the experimental condition found can be considered as attributable to the experimental condition.

This study also aimed to examine the role of infant temperament on infant affect and behaviour, in response to maternal expressed anxiety or encouragement. There is strong evidence from meta-analytic studies that the temperamental style of behavioural inhibition (BI) is associated with a greater risk for developing an anxiety disorder in childhood, particularly social anxiety disorder (e.g. Clauss & Blackford, 2012; Sandstrom et al., 2019). The literature examining factors that influence childhood anxiety have identified that temperamental factors of the infant or child can moderate the relationship between parenting behaviours and anxiety in the infant or child. For example, the longitudinal research presented by Hudson and colleagues (e.g. Hudson et al., 2011; 2012; 2019) has found that infants identified as behaviourally inhibited were at an increased risk of anxiety at the age of 12, only if they experienced maternal over-involvement in early childhood. Similarly, de Rosnay et al. (2006) reported an interactive relationship between temperamental and parenting factors, whereby infant avoidance of a stranger was more pronounced for the infants with a temperamental style of high fear, when their mother was expressing anxiety. Hence, it was predicted in this study that infants with a temperamental style indicative of greater negative affect were more likely to demonstrate fearful, avoidant, and negative responses to a stranger when their mothers were acting anxiously.

Unexpectedly, there was no main effect of the experimental condition on any of the infant affect and behaviour outcomes, nor were there any significant interactions between condition and infant temperamental style. It was expected that infants would demonstrate greater fearfulness, avoidance, and negative emotional tone (and lower positive emotional tone) in the expressed anxiety condition, and that this difference would be more pronounced in infants with a temperamental style indicative of greater negative temperament (a temperamental dimension associated with BI, including fearfulness and clinginess in novel situations). These findings are in contrast to the finding from de Rosnay and colleagues (2006) (who used a similar social-referencing paradigm and video coding protocol to examine infant affect and behaviour in response to maternal expressed anxiety or

maternal neutral responses to a stranger). They reported a main effect of condition, whereby infants were significantly more fearful in the anxious condition as compared to the non-anxious condition, as well as reporting an interaction between condition and temperamental fear, in which high fear infants were more avoidant in the anxious condition. This study also used a non-clinical sample of mothers, although the infant age-range was narrower than in the present study (12 – 14 months, as opposed to our inclusion range of 10 – 14 months). Infants' skills in social referencing have been shown to develop from around ten months of age (Feinman, 1982). It is therefore possible that the social referencing skills were better developed in the 12 – 14 month infants and in our study the lower age threshold meant that overall the infant sample were able to make less use of their mothers' cues via social referencing.

The findings from our study suggested that infant factors of gender and temperament played a greater role in infant affect and behaviour outcomes than did maternal behaviour. In part 1 there were no significant gender differences in regards to the number of looks from the infant to the mother. In part 1 the infant is only observing the strangers speaking with their mother, whereas in part 2 the strangers engage directly with the infant and is therefore considered a more socially threatening situation for the infant. In relation to gender; during the interactions with the strangers (part 2), female infants looked to their mothers significantly more times than male infants – this finding may indicate higher levels of anxiety, irrespective of experimental condition. Alternatively, it could suggest that female infants had more developed social referencing skills and thus were more likely to turn to their mothers for cues as to how to respond in the novel situation. Additionally, male infants displayed higher levels of positive emotional tone than female infants when interacting with the strangers– indicating lower levels of anxiety and higher levels of enjoyment and contentment, irrespective of the experimental condition. The strangers in the study were male, so these findings may relate to the interaction between infant and stranger gender, with female infants experiencing a social interaction with a male adult as more anxiety-provoking than male infants. Of note, a similar social referencing

study with mother-infant dyads, conducted by Murray et al. (2008), used female strangers and it was found that male infants (of socially anxious mothers) showed increasing fearful responses to the female strangers, which was not a pattern seen in the female infants (of socially anxious mothers). It would be interesting to repeat our study using female strangers to examine whether these gender differences remain. Repetition of the current study with a larger sample size would also be useful to determine how reliable these gender differences are. Additionally, repetition of the current study could consider the role of infant attachment security, which is a potential confounding variable that may play a mediating role between maternal behaviour and infant affect and behaviour.

In relation to infant temperament; we found that infants with a more negative temperament were significantly more likely to demonstrate anxiety than infants with lower temperamental negativity, irrespective of whether their mother had acted anxiously or encouragingly towards the strangers. Temperamental negative affect was associated with greater levels of fearfulness and more looks to their mother during the interactions with the strangers. This finding suggests that infants with a temperamental style associated with greater negativity experience higher levels of anxiety and seek more reassurance from their mothers in novel situations. However, these were the only outcome domains in which this pattern was found; there were no significant associations between negative temperament and infant avoidance, negative emotional tone, or positive emotional tone. Comparable findings to these have been reported by Aktar et al. (2013) who reported that infants who were more behaviourally inhibited showed greater fear during a social referencing task, but, similarly, did not show more avoidance with a stranger than did children who were not behaviourally inhibited. It is difficult to draw strong conclusions from these findings, as both fearfulness and avoidance are considered indicators of anxiety, but it is suggested that in the presence of higher temperamental negative affect, temperamental style has a stronger influence over infant anxiety than maternal behaviours.

We know that the two experimental conditions were significantly different in regards to maternal behaviour observed by the infants, with mothers consistently acting in an anxious manner in the anxious condition and acting in an encouraging manner in the encouraging condition. However, we considered whether underlying maternal social anxiety symptoms (as measured by the SPIN) may have an effect on how the infants responded to the strangers. It was wondered whether infants of mothers with greater levels of social anxiety may be more sensitive in general to novel social situations based on previous learning experiences from their mothers. Social anxiety scores from the SPIN did not have a main effect on any of the infant affect outcomes, nor did SPIN scores have any interactive effects with other factors, such as experimental condition. However, it is important to note that this was a non-clinical sample, with the majority of the sample scoring below the suggested cut-off score for social anxiety, and the highest social anxiety score in the sample (36) being substantially lower than the maximum possible score on this scale (68). Hence, a clinical sample of socially anxious mothers may yield different outcomes with respect to the impact of underlying social anxiety.

Our study design used a non-clinical sample of mothers, which allowed us to examine infant learning from the social referencing paradigm with infants who did not have a learning history of observing anxious behaviours. However, this means that there are some limitations in regards to the generalisability of the findings. Use of a non-clinical sample of mothers means that we are unable to conclude that the training videos to modify maternal behaviour would be equally as effective in a sample of mothers who are experiencing an anxiety disorder. It is important to recognise that acting in an encouraging way is likely to be a much more difficult task for anxious mothers than the task of acting anxiously for non-anxious mothers (as required for our non-clinical sample). However, Ewing et al. (2019) demonstrated equally effective outcomes from video training of parenting behaviours implicated in the development of childhood anxiety, between groups of mothers with and without anxiety disorders with primary school aged children. It is therefore expected that our findings would

translate to clinical populations but this has not yet been demonstrated in a clinical sample with infants.

The self-selecting sample for this study is likely to have encountered bias in relation to participant characteristics. The maternal mood and anxiety questionnaires suggest this may have been the case, particularly for maternal depression, with all but one participant scoring below the clinical cut-off score for likely postnatal depression. Additionally, the majority of participants did not score within the range indicative of social anxiety disorder, and of those who did score above this cut-off they were within the mild or moderate ranges for symptom severity. Anecdotally, a number of mothers commented on how unusual it would be for their infant to see them acting anxiously when speaking to a stranger. Hence, infants observing only a 90-second anxious interaction between their mother and a stranger may have had limited impact on the infant's response to the strangers, within the wider context of their life experiences of witnessing their mother speaking to strangers in novel situations. Given that previous research has demonstrated that the parenting behaviours associated with infant or child anxiety are more frequently seen in mothers with anxiety disorders, it would be helpful to investigate the same research questions using a sample of mothers with anxiety disorders. Murray et al. (2008) used a social referencing paradigm to compare infant responses to a stranger between mothers with and without social phobia, without attempts to modify maternal behaviour. An expansion of this study using a clinical sample, with the addition of modification of maternal behaviour could be a helpful next step in understanding the applicability of our findings within a clinical population.

A limitation in terms of the infant sample characteristics was with respect to the inclusion of twins, which had not been foreseen when planning the study or identified within the research literature. Three pairs of twins completed the study - each twin completed both experimental conditions individually with their mother, while the other twin was looked after outside of the

research laboratory. One set of twins (dizygotic) successfully completed the study tasks, however, in one set of (monozygotic) twins both infants were too distressed to complete the experimental tasks and in the third set of (monozygotic) twins only one twin was able to complete the experimental tasks, with the other being too distressed. Although these are very small numbers to comment on, it is hypothesised that the experimental tasks were more distressing for twin infants, who are usually used to the constant company of their twin, than for single infants. A future direction to explore this could involve the inclusion of a larger number of twin pairs to examine the reliability of these differences in a similar study. Alternatively, to mimic more natural contexts, twin pairs could participate in the experimental conditions together, which would allow us to investigate both the impact of social referencing from the parent and from the other twin.

In regards to the strengths of this research; to our knowledge, this is the first time that it has been demonstrated that it is possible to modify both maternal expressed anxiety and encouragement via video tuition. The use of video tuition in research can therefore be considered an outcome-effective as well as a cost- and time-effective means to support parents in modifying parenting behaviours that are implicated in the development of childhood anxiety. Research consistently points to the importance of early-intervention for preventing the development of anxiety in childhood; hence, the young age of our infant participants is a strength in demonstrating that video-tuition for parents can be an effective intervention tool from a young age.

As noted above, a useful area for future research would be to examine the efficacy of the training videos in a clinical sample of mothers with anxiety disorders and to explore the impact of the mothers' different style of responding (anxious or encouraging) on infant affect and behaviour. This would tell us whether it would be beneficial to pursue the development of intervention programmes that use video training tools for mothers with anxiety disorders, whose children will be at a greater risk of developing an anxiety disorder from intergenerational transmission, via parental modelling of

anxious behaviour. The use of video technology has been used as an intervention tool with other clinical populations. For example, Video Interaction Guidance™ (VIG) is an intervention used to develop attunement and bonding in parent-child relationships, with the aim of fostering secure attachments. Typically a practitioner in VIG would support a parent to reflect on video clips of their interactions between the parent and child, to support the enhancement of attuned communication (Association for Video Interaction Guidance UK, 2020). A randomised control trial by Stein et al. (2018) examined the efficacy of video-feedback therapy (VFT) for mothers with postnatal depression, which was compared to a control intervention (whilst all mothers were also undergoing additional cognitive behavioural therapy for depression). It would be interesting therefore for future research to employ a similar study design in order to consider how VIG or VFT interventions could be used to support parents with anxiety disorders in modifying their interactions with their children, as a tool for preventing the environmental transmission of anxiety from parenting behaviours.

In our study the infants observed their mother interacting with a stranger and then interacted with the stranger themselves, without any further input from their mother. This allowed us to specifically examine the infants' learning through social referencing from watching their mother interact with the strangers. However, in a more natural setting it is more likely that the mother would continue to be involved in an interaction between their infant and a stranger, and hence their behaviour would continue to influence how the infant responds. Murray et al. (2008) examined socially anxious and non-socially anxious mothers' responses to their infant interacting with a stranger and reported lower levels of encouragement, warmth, and positive modelling in mothers with social anxiety. It would be useful in future work to extend the current study paradigm to explore whether maternal behaviour during the infant-stranger interaction resulted in the infant responding differently in the two experimental conditions.

Another important research avenue to explore would be regarding father-infant dyads, using a similar research paradigm. Aktar et al. (2013) presented findings from a study involving both mothers and fathers of 12-month old infants. They demonstrated parental differences during a social referencing paradigm, whereby for fathers there was an association between, on the one hand, infant anxiety and, on the other hand, more paternal expressed encouragement and less expressed anxiety, which was not found to be the case for mothers. The mechanisms for these differences were unclear and it would be interesting to explore and understand any parenting differences implicated in childhood anxiety. Meta-analyses by Moller, Nikolic, Majdandzic, and Bogels (2016) examined the association between childhood anxiety (and known precursors to childhood anxiety, such as inhibited temperament) and parenting behaviours in mothers and fathers. Although the associations found between parenting behaviours and childhood anxiety were small, parenting behaviours were more strongly associated with childhood anxiety than child anxiety precursors. The authors reported that there was not a significant difference between mothers and fathers in terms of the behaviours of overprotection and over-involvement. However, a difference was found between mothers and fathers on the dimension of challenging parenting, whereby fathers' challenging parenting was related to less anxiety in the child, which was not the case for mothers. These findings led the authors to conclude that fathers' parenting is equally as important as mothers' parenting in relation to child anxiety outcomes. To date, a lot of research in child development has overlooked the role of fathers and it is important for future research to examine the influence of both mothers and fathers parenting behaviours on the development of childhood anxiety. This area of research is particularly relevant in light of changing trends in childcare practices in recent years, with more fathers taking on childcare responsibilities. These trends have been acknowledged by government legislation; in the UK in 2015 shared parental leave was introduced by the UK Government, allowing mothers and fathers to share parental leave between them.

In conclusion, this study has demonstrated that video tuition is effective in modifying parenting behaviours that have been implicated in the development of childhood anxiety, namely expressed anxiety and encouragement. This finding has contributed to the evidence-base for the utility of video tuition for modifying parenting behaviours in infancy, and shown that both factors that increase the risk of childhood anxiety (i.e. expressed anxiety) and decrease the risk of childhood anxiety (i.e. encouragement) are modifiable. We have suggested that it would be worthwhile further investigating the efficacy of video tuition in clinical samples, to understand if mothers with anxiety disorders are equally able to modify their parenting behaviours using this method of intervention.

Similarly to other research findings in this field, our findings suggest a complex relationship between anxiety and the influence of parenting factors and temperamental factors. In our non-clinical sample it appeared that the temperamental style indicative of negative temperament had a greater influence over the infant anxiety than parenting factors; whereby negative temperament was found to be significantly associated with infant anxiety and parent factors were not found to be significantly associated with infant anxiety. There also appeared to be an interactive effect between infant and stranger gender, with female infants' behaviour indicating greater levels of anxiety than male infants' behaviour, when interacting with the male strangers. Expanding the study to modify some of the potential contributing and confounding factors identified in this study (such as inclusion of fathers, further exploration of twin relationships, and examination of the impact of different gender strangers) would be helpful avenues to explore in future research.

Appendix A – Summary of the 55 Studies Included in the Meta-Analysis

Table 2

Summary of the 55 Studies Included in the Meta-Analysis

Author	Year	Design	BI measure	Mean age of BI measure	BI Type	Anxiety measure	Mean age of anxiety measure	Anxiety type	Anxiety outcome
Abend	2019	Prospective	Observational and parent-report	2.62	Categorical	Child and parent combined report	10.51, 13.04	Categorical	Any Anxiety
Ale	2010	Cross-sectional	Parent-report	4.42	Continuous	Parent-report	4.42	Continuous	Social Anxiety
Auday	2019	Cross-sectional	Parent-report	10.81	Continuous	Parent-report	10.81	Continuous	Any Anxiety
Biederman	1993	Prospective	Observational	1.75, 5.12	Categorical	Clinical Interview	7.45, 11.25, 5.12, 6.38	Categorical	Any Anxiety, Separation Anxiety, Social Anxiety, Phobia, Panic
Bourdon	2019	Prospective	Parent-report	N/A	Continuous	Child and parent combined report	11.22	Continuous	Social Anxiety, Generalised Anxiety, Separation Anxiety, Panic
Broeren	2009	Cross-sectional	Parent-report	6.09	Continuous	Parent-report	6.09	Continuous	Any Anxiety
Buss	2011	Prospective and Cross-sectional	Observational	2	Continuous	Parent-report	2, 3, 4, 5	Continuous	Any Anxiety

ROLE OF TEMPERAMENT AND PARENTING BEHAVIOURS ON ANXIETY

Author	Year	Design	BI measure	Mean age of BI measure	BI Type	Anxiety measure	Mean age of anxiety measure	Anxiety type	Anxiety outcome
Buzzell	2017	Prospective	Observational and parent-report	2.62	Continuous	Parent-report	13.58	Continuous	Social Anxiety, Generalised Anxiety
Chronis-Tuscano	2009	Prospective	Observational and parent-report	1.2 – 7*	Categorical	Clinical interview	15.05	Categorical	Any Anxiety Social Anxiety
Dougherty	2013	Cross-sectional	Observational	3.6	Continuous	Clinical interview	3.6	Categorical	Any Anxiety
Dyson	2011	Cross-sectional	Observational and parent-report	3.5	Categorical	Clinical interview	3.5	Categorical	Social Anxiety, Phobia, Separation Anxiety, Generalised Anxiety
Edwards	2010	Prospective, Cross-sectional, Retrospective	Parent-report	3.95, 5	Continuous	Parent-report	3.95, 5	Continuous	Any Anxiety
Elliott	2011	Cross-sectional	Parent-report	5.61	Categorical	Parent-report	5.61	Continuous	Any Anxiety
Fu	2017	Cross-sectional	Parent-report	6.06	Categorical	Observational	6.06	Continuous	Any Anxiety

ROLE OF TEMPERAMENT AND PARENTING BEHAVIOURS ON ANXIETY

Author	Year	Design	BI measure	Mean age of BI measure	BI Type	Anxiety measure	Mean age of anxiety measure	Anxiety type	Anxiety outcome
Hirshfeld-Becker	2007	Prospective	Observational	4.19	Categorical	Clinical interview	9.28	Categorical	Social Anxiety
Howard	2017	Cross-sectional, Prospective	Parent-report	4	Continuous	Parent-report	4, 5	Continuous	Any Anxiety
Hudson	2011	Cross-sectional, Prospective	Observational and parent-report	4	Categorical	Clinical interview	4, 6.03	Categorical	Any Anxiety
Hudson	2012	Prospective	Observational and parent-report	4	Categorical	Clinical interview	8.9	Categorical	Any Anxiety
Hudson	2018	Prospective	Observational and parent-report	4	Categorical	Clinical interview	11.8	Categorical	Any Anxiety
Kiel	2016	Cross-sectional, Prospective	Observational	2.1	Continuous	Parent-report	2.1	Continuous	Separation Anxiety
Kim	2016	Cross-sectional	Parent-report	13.4	Continuous	Parent-report	13.4	Continuous	Any Anxiety

ROLE OF TEMPERAMENT AND PARENTING BEHAVIOURS ON ANXIETY

Author	Year	Design	BI measure	Mean age of BI measure	BI Type	Anxiety measure	Mean age of anxiety measure	Anxiety type	Anxiety outcome
Kotelnikova	2013	Cross-sectional	Observational	7.41	Continuous	Parent-report	7.41	Continuous	Any Anxiety
Lahat	2018	Prospective	Observational and parent-report	2.62	Continuous	Parent-report, Self-report, Child and parent combined reports	10-13*	Continuous	Social Anxiety
Lamm	2014	Prospective	Observational and parent-report	2.62	Continuous	Parent-report	7.64	Continuous	Any Anxiety
Majdandzic	2018	Prospective	Observational	1.05	Continuous	Parent-report	2.55, 4.56	Continuous	Any Anxiety
Manassis	1995	Cross-sectional	Observational	3	Categorical	Clinical interview	36.3m	Categorical	Any Anxiety
McClellan	2019	Prospective	Parent-report	1.4	Continuous	Parent-report	48.8m	Continuous	Any Anxiety

ROLE OF TEMPERAMENT AND PARENTING BEHAVIOURS ON ANXIETY

Author	Year	Design	BI measure	Mean age of BI measure	BI Type	Anxiety measure	Mean age of anxiety measure	Anxiety type	Anxiety outcome
Mian	2011	Prospective	Parent-report	3	Continuous	Parent-report	6.03, 8.01	Continuous	Any Anxiety
Morales	2017	Cross-sectional	Parent-report	10.19	Categorical	Clinical interview	10.19	Continuous	Social Anxiety
Mumper	2019	Prospective	Observational	3.55, 3.56	Continuous	Self-report	9.17, 12.67	Continuous	Any Anxiety
Muris	2002	Cross-sectional	Parent-report	12.7	Continuous	Parent-report	12.7	Continuous	Any Anxiety
Muris	2009	Cross-sectional	Parent-report	10.54	Continuous	Parent-report	10.54	Continuous	Any Anxiety
Muris	2011	Prospective	Parent-report	6.6	Continuous	Clinical interview, Parent-report	5-11*	Continuous	Social Anxiety, Any (Non-Social) Anxiety
Muris	2016	Cross-sectional	Parent-report	4.10, 4.98, 4.99	Continuous	Parent-report	4.98	Continuous	Social Anxiety, Any (Non-Social) Anxiety, Selective Mutism

ROLE OF TEMPERAMENT AND PARENTING BEHAVIOURS ON ANXIETY

Author	Year	Design	BI measure	Mean age of BI measure	BI Type	Anxiety measure	Mean age of anxiety measure	Anxiety type	Anxiety outcome
Natsuaki	2013	Prospective, Cross-sectional, Retrospective	Observational	1.17, 2.25	Continuous	Parent-report	1.17, 2.25	Continuous	Social Anxiety
Niditch	2018	Prospective	Parent-report	0.5	Continuous	Parent-report	6	Continuous	Any Anxiety
Olino	2014	Cross-sectional	Observational	3.56	Continuous	Clinical interview	3.56	Categorical	Generalised Anxiety, Separation Anxiety, Panic, Social Anxiety, Phobia
Pahl	2012	Cross-sectional	Parent-report	4.45	Continuous	Parent-report	4.45	Continuous	Any Anxiety
Paulus	2015	Retrospective	Parent-report	59m**	Categorical	Parent-report	6.1	Categorical	Any Anxiety, Social Anxiety, Separation Anxiety, Phobia
Perez-Edgar	2014	Prospective	Observational and parent-report	2.62	Continuous	Parent-report, self-report	16.33	Continuous	Any Anxiety
Rapee	2014	Prospective	Observational and parent-report	3.8	Categorical	Clinical interview	15.4	Categorical	Any Anxiety, Separation Anxiety, Generalised Anxiety, Social Anxiety, Phobia, OCD

ROLE OF TEMPERAMENT AND PARENTING BEHAVIOURS ON ANXIETY

Author	Year	Design	BI measure	Mean age of BI measure	BI Type	Anxiety measure	Mean age of anxiety measure	Anxiety type	Anxiety outcome
Reeb-Sutherland	2009	Prospective	Observational and parent-report	2.62	Continuous	Clinical interview	15.1	Categorical	Any Anxiety
Reeb-Sutherland	2015	Prospective	Observational and parent-report	2.62	Continuous	Clinical interview	15.06	Categorical	Any Anxiety
Schwartz	1999	Prospective	Observational	1-2*	Categorical	Clinical interview	13	Categorical	Social Anxiety
Stumper	2017	Prospective, Cross-sectional	Observational, parent-report	3	Continuous	Clinical interview	3, 9	Continuous	Any Anxiety
Suzrez	2019	Cross-sectional	Parent-report	9.97	Continuous	Clinical interview	9.97	Continuous	Social Anxiety
Troller-Renfree	2019a	Prospective	Observational and parent-report	2.62	Continuous	Parent-report, Self-report	12	Continuous	Any Anxiety
Troller-Renfree	2019b	Prospective	Observational and parent-report	2.62	Continuous	Parent-report, Self-report	13	Continuous	Any Anxiety
van der Linden	2013	Cross-sectional	Parent-report	10.54	Continuous	Parent-report	10.54	Continuous	Any Anxiety

ROLE OF TEMPERAMENT AND PARENTING BEHAVIOURS ON ANXIETY

Author	Year	Design	BI measure	Mean age of BI measure	BI Type	Anxiety measure	Mean age of anxiety measure	Anxiety type	Anxiety outcome
Volbrecht	2010	Prospective	Observational and parent-report	3.04	Continuous	Parent-report	7.51	Continuous	Separation and Generalised Anxiety
Vreeke & Muris	2012	Cross-sectional	Parent-report	9.07	Continuous	Parent-report	9.07	Continuous	Any Anxiety
Vreeke	2012	Prospective	Parent-report	3.59	Continuous	Parent-report	4.47	Continuous	Any Anxiety
Vreeke	2013	Prospective, Cross-sectional, Retrospective	Parent-report	4.54	Continuous	Parent-report	4.54	Continuous	Social Anxiety, Any (Non-Social) Anxiety
West	2007	Cross-sectional	Parent-report	13.5	Continuous	Self-report	13.5	Continuous	Social Anxiety
White	2017	Prospective	Observational and parent-report	2.61	Continuous	Parent-report	5.26, 7.64	Continuous	Any Anxiety

Note. * = mean age range not reported, age range reported instead. ** = mean age or age range not reported, age of recruitment reported instead.

Appendix B – Summary of all excluded articles reviewed at full text stage

Full Text Review, n = 234

Total included meeting eligibility criteria, n =70

Excluded studies not meeting eligibility criteria, n = 164

Excluded studies due to data unavailable or inappropriate for meta-analysis, n = 15

Total Excluded studies (not meeting eligibility criteria and data unavailable or inappropriate for meta-analysis) n = 179

Total included in meta-analysis, n = 55

Excluded due to no original data (e.g. review paper, data replicated from another included study) n = 23

Auday (2018)	Mihalopoulos (2015)
Axelson (2001)	Muris (2003)
Bayer (2011)	Nozadi (2016)
Biederman (1990)	Pahl (2010)
Broeren (2013)	Pass (2012)
Buffred (2018)	Rosenbaum (1993)
Degnan (2014)	Slyvester (2016)
Dodd (2012 [2011 online])	Taber-Thomas (2016)
Henderson (2018)	Thai (2016)
Hirshfeld (1992)	White (2018)
Hirshfeld (1997 [Associations Between])	Dougherty (2011)
Hirshfeld (1997 [Expressed Emotion])	

Excluded due to BI measured using self-report, n = 14

Askew (2015)	Fu (2019)
Barry (2018)	Hill (2010)
Bessdo-Baum (2012)	Messer (2004)
Brozina (2006)	Rohrbacher (2008)
Coles (2006)	Bas-Hoogendam (2018)
Field (2006)	Beesdo (2010)
Frost Bellgowan (2015)	Lim (2013)

ROLE OF TEMPERAMENT AND PARENTING BEHAVIOURS ON ANXIETY

Excluded due to no valid quantitative BI measure, n = 35

Aktar (2019 [2018 online])	Kwon (2019)
Battaliga (2004)	Lee (2017)
Bloemsma (2013)	Leen-Feldner (2004)
Bortoluzzi (2014)	Lupattelli (2018)
Canals (1992)	Martin (2007)
Clarbour (2004)	Mian (2015)
Dadds (2008)	Orri (2018)
DePrince (2009)	Pappa (2014)
Dipietro (2018)	Prior (2000)
FernandezdelaCruz (2018)	Rogers (2017)
Goldstein (2010)	Roughan (2011)
Gouze (2017)	Rubin (2013)
Grant (2009)	Schrock (2010)
Hardway (2013)	Stochl (2015)
Hermansen (2016)	Trevo-Clemmens (2017)
Huang-Pollock (2017)	Tsui (2017)
Husby (2017)	Weeks (2016)
Kerr (1997)	

Excluded due to no valid quantitative anxiety measure, n = 80

Aktar (2013)	Buss (2018)
Aktar (2014)	Davies (2011)
Aktar (2018 [2017 online])	Degnan (2008)
Ballespi (2012)	Eisenberg (1998)
Battaliga (1997)	Fang (2018)
Biederman (2001 [Lack of Association])	Gensthaler (2013)
Biederman (2001)	Gensthaler (2016)
Bohlin (2009)	Georgiou (2014)
Broeren (2013)	Gest (1997)
Brooker (2013)	Gibler (2018)
Brooker (2016)	Gunnar (2011)

ROLE OF TEMPERAMENT AND PARENTING BEHAVIOURS ON ANXIETY

Guyer (2015)
Hastings (2008)
Hastings (2011)
Hastings (2015 [2014 online])
He (2017)
Hirshfeld-Becker (2002)
Hirshfeld-Becker (2004)
Jockman (2014)
Johnson (2016)
Johnson (2016 [A Longitudinal Investigation])
Kalomiris (2016)
Kiel (2014)
Koomen (2003)
Letcher (2009)
Liu (2019)
Lozoff (2014)
Marysko (2010)
Meysamie (2013)
Mills (2012)
Moehler (2006)
Moehler (2007)
Moehler (2007 [International J..])
Moehler (2008)
Morales (2015)
Morris (2014)
Mount (2010)
Muris (2006)
Murray (2012)
Olino (2010)
Otto (2007)
Pauli-Pott (2007)
Perez-Edgar (2010)
Perez-Edgar (2011)
Putman (2005)
Raishevich (2010)
Rickman (1994)
Robinson (1992)
Rosenbaum (1991)
Rosenbaum (1992)
Rosenbaum (2000)
Schwartz (2015)
Schwartz and Kunwar (2015)
Schweizer (2018)
Smith (2019)
Smoller (2001)
Smoller (2003)
Smoller (2005)
Smoller (2008)
Soo (2007)
Stellern (2014)
Stevenson-Hinde (2011)
Stevenson-Hinde (2013)
Trucco (2017)
van Brakel (2006)
van der Voort (2014)
Vandermeer (2018)
Vrticka (2013)
Williams (2014)

ROLE OF TEMPERAMENT AND PARENTING BEHAVIOURS ON ANXIETY

Excluded due to anxiety measured post 18 years, n = 6

Fan (2015)	Moffitt (2007)
Frenkel (2015)	Roy (2014)
Hardee (2013)	Caspi (1996)

Excluded due to other reasons, n = 6

Bischof (2018) – Only children with autism measured	Rapee (2005) - This is an intervention study that does not present base-line data.
Hirshfeld-Becker (2010) – No baseline, participants already had anxiety disorders	Rapee (2013) - This is a follow-up for an intervention study that does not present base-line data.
Rapee (2002) – This is an intervention study that does not report base-line data (the data required is in Shamir-Essakow, 2005).	Sheynin (2014) – Adult participants

Met Inclusion but Excluded due to no response from author for data, n = 5

Anticich (2013)
Barker (2015)
Essex (2010)
Murray (2014)
Shamir-Essakow (2005)

Met Inclusion but Excluded due to data in inappropriate format for meta-analysis (e.g. categorical data for BI and anxiety with no comparison group), n = 10

Bayer (2019) – only BI children, no comparison
Buffred (2018) – only children meeting anxiety diagnosis
Chronis-Tuscano (2015) – only BI children, no comparison
Kennedy (2009) – only BI children, no comparison
Lau (2017) – only BI children, no comparison
Harrewijn (2019)
Lewis-Morrarty (2012)
Lewis-Morrarty (2015)
Marakovitz (2011)
Winchstrom (2013)

Appendix C - Graph to illustrate relationship between Fisher's Z and Pearson's r

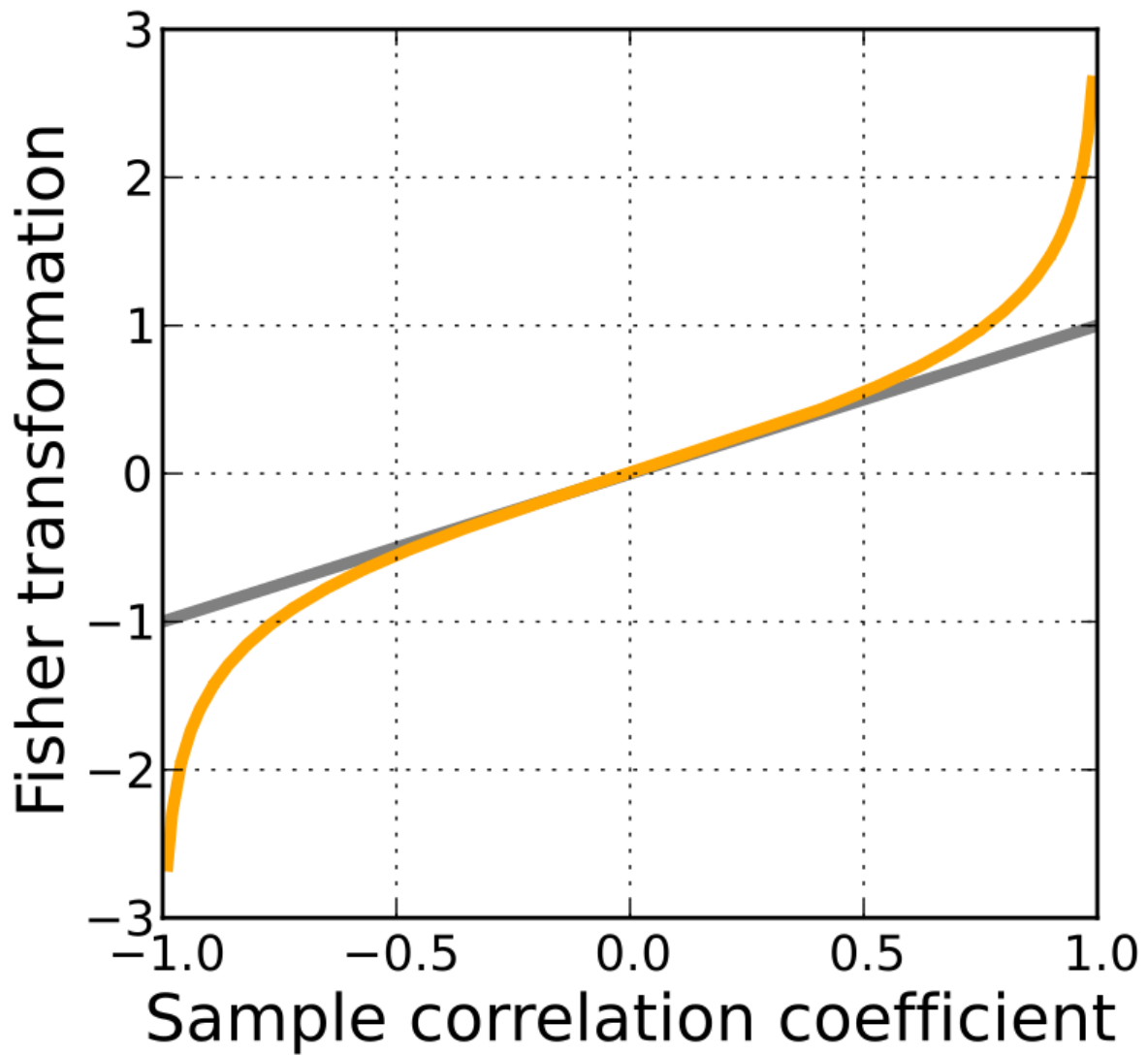


Figure 7. Graph to illustrate relationship between Fisher's Z and Pearson's r.
Note. Orange line = transformation (Z), grey line = original function (r). Source: By Skbkekas, CC BY-SA 3.0, <https://commons.wikimedia.org/w/index.php?curid=8776266>

Appendix D – Standard Quality Assessment Criteria

Table 3

Assessment of Study Quality for the 55 Included Studies using the 11 Relevant Criteria from Kmet et al. (2004) Standard Quality Assessment Criteria for Quantitative Studies.

Study Number	Study	1. Question / objective sufficiently described?	2. Study design evident and appropriate?	3. Method of subject/comparison group selection or source of information/input variables described and appropriate?	4. Subject (and comparison group, if applicable) characteristics sufficiently described?	8. Outcome and (if applicable) exposure measure(s) well defined and robust to measurement / misclassification bias?	9. Sample size appropriate?	10. Analytic methods described/justified and appropriate?	11. Some estimate of variance is reported for the main results?	12. Controlled for confounding?	13. Results reported in sufficient detail?	14. Conclusions supported by the results?	Total score	Summary Score
1	Abend (2019)	2	2	2	2	2	2	2	2	2	2	2	22	1.00
2	Ale (2010)	2	2	2	2	2	0	2	0	2	2	2	18	0.82
3	Auday (2019)	2	2	2	2	2	2	2	0	2	2	2	20	0.91
4	Biederman (1993)	2	2	2	2	2	2	2	0	2	2	2	20	0.91

ROLE OF TEMPERAMENT AND PARENTING BEHAVIOURS ON ANXIETY

Number	Study	1. Question / objective sufficiently described?	2. Study design evident and appropriate?	3. Method of subject/comparison group selection or source of information/input variables described and appropriate?	4. Subject (and comparison group, if applicable) characteristics sufficiently described?	8. Outcome and (if applicable) exposure measure(s) well defined and robust to measurement / misclassification bias?	9. Sample size appropriate?	10. Analytic methods described/justified and appropriate?	11. Some estimate of variance is reported for the main results?	12. Controlled for confounding?	13. Results reported in sufficient detail?	14. Conclusions supported by the results?	Total score	Summary Score
5	Bourdon (2019)	2	2	2	2	2	2	2	2	2	2	2	22	1.00
6	Broeren (2009)	2	2	2	2	2	2	2	2	2	2	2	22	1.00
7	Buss (2011)	2	2	2	2	2	2	2	2	2	2	2	22	1.00
8	Buzzell (2017)	2	2	2	2	2	2	2	2	2	2	2	22	1.00
9	Chronis-Tuscano (2009)	2	2	2	2	2	2	2	2	2	2	2	22	1.00
10	Dougherty (2013)	2	2	2	2	2	2	2	2	2	2	2	22	1.00
11	Dyson (2011)	2	2	2	2	2	2	2	2	2	1	2	21	0.95
12	Edwards (2010)	2	2	2	2	2	2	2	2	2	2	2	22	1.00

ROLE OF TEMPERAMENT AND PARENTING BEHAVIOURS ON ANXIETY

Number	Study	1. Question / objective sufficiently described?	2. Study design evident and appropriate?	3. Method of subject/comparison group selection or source of information/input variables described and appropriate?	4. Subject (and comparison group, if applicable) characteristics sufficiently described?	8. Outcome and (if applicable) exposure measure(s) well defined and robust to measurement / misclassification bias?	9. Sample size appropriate?	10. Analytic methods described/justified and appropriate?	11. Some estimate of variance is reported for the main results?	12. Controlled for confounding?	13. Results reported in sufficient detail?	14. Conclusions supported by the results?	Total score	Summary Score
13	Elliott (2011)	2	2	2	2	2	1	2	2	2	2	2	21	0.95
14	Fu (2017)	2	2	2	2	2	2	2	2	2	2	2	22	1.00
15	Hirshfeld-Becker (2007)	2	2	2	2	2	2	2	2	2	2	2	22	1.00
16	Howard (2017)	2	2	2	2	2	1	2	2	2	2	2	21	0.95
17	Hudson (2011)	2	2	2	2	2	2	2	2	2	2	2	22	1.00
18	Hudson (2012)	2	2	2	2	2	2	2	2	2	2	2	22	1.00
19	Hudson (2018)	2	2	2	2	2	1	2	0	2	2	2	19	0.86
20	Kiel (2016)	2	2	2	2	2	2	2	2	2	2	2	22	1.00
21	Kim (2016)	2	2	2	2	2	1	2	0	2	2	2	19	0.86

ROLE OF TEMPERAMENT AND PARENTING BEHAVIOURS ON ANXIETY

Number	Study	1. Question / objective sufficiently described?	2. Study design evident and appropriate?	3. Method of subject/comparison group selection or source of information/input variables described and appropriate?	4. Subject (and comparison group, if applicable) characteristics sufficiently described?	8. Outcome and (if applicable) exposure measure(s) well defined and robust to measurement / misclassification bias?	9. Sample size appropriate?	10. Analytic methods described/justified and appropriate?	11. Some estimate of variance is reported for the main results?	12. Controlled for confounding?	13. Results reported in sufficient detail?	14. Conclusions supported by the results?	Total score	Summary Score
22	Kotelnikova (2013)	2	2	2	2	2	1	2	0	2	1	2	18	0.82
23	Lahat (2018)	2	2	2	2	2	1	2	0	2	2	2	19	0.86
24	Lamm (2014)	2	2	2	2	2	2	2	0	1	2	2	19	0.86
25	Majdandzic (2018)	2	2	2	2	2	2	2	1	2	2	2	21	0.95
26	Manassis (1995)	2	2	2	2	2	0	2	0	2	2	2	18	0.82
27	McLean (2019)	2	2	2	2	2	1	2	2	2	2	2	21	0.95
28	Mian (2011)	2	2	2	2	2	2	2	2	2	2	2	22	1.00
29	Morales (2017)	2	2	2	2	2	2	2	2	2	2	2	22	1.00
30	Mumper (2019)	2	2	2	2	2	1	2	2	1	2	2	20	0.91
31	Muris (2002)	2	2	2	2	2	2	2	1	0	2	2	19	0.86

ROLE OF TEMPERAMENT AND PARENTING BEHAVIOURS ON ANXIETY

Study Number	Study	1. Question / objective sufficiently described?	2. Study design evident and appropriate?	3. Method of subject/comparison group selection or source of information/input variables described and appropriate?	4. Subject (and comparison group, if applicable) characteristics sufficiently described?	8. Outcome and (if applicable) exposure measure(s) well defined and robust to measurement / misclassification bias?	9. Sample size appropriate?	10. Analytic methods described/justified and appropriate?	11. Some estimate of variance is reported for the main results?	12. Controlled for confounding?	13. Results reported in sufficient detail?	14. Conclusions supported by the results?	Total score	Summary Score
32	Muris (2009)	2	2	2	2	2	2	2	1	2	2	2	21	0.95
33	Muris (2011)	2	2	2	2	2	2	2	2	0	2	2	20	0.91
34	Muris (2016)	2	2	1	2	2	1	2	2	2	2	2	20	0.91
35	Natsuaki (2013)	2	2	2	2	2	0	2	0	2	2	2	18	0.82
36	Niditch (2018)	2	2	2	2	2	2	2	2	2	2	2	22	1.00
37	Olino (2014)	2	2	2	2	2	2	2	0	0	2	2	18	0.82
38	Pahl (2012)	2	2	2	2	2	2	2	2	0	2	2	20	0.91
39	Paulus (2015)	2	2	2	2	2	2	2	2	0	2	2	20	0.91
40	Perez-Edgar (2014)	2	2	2	2	2	0	2	2	2	2	2	20	0.91
41	Rapee (2014)	2	2	2	2	2	1	2	0	2	2	2	19	0.86

ROLE OF TEMPERAMENT AND PARENTING BEHAVIOURS ON ANXIETY

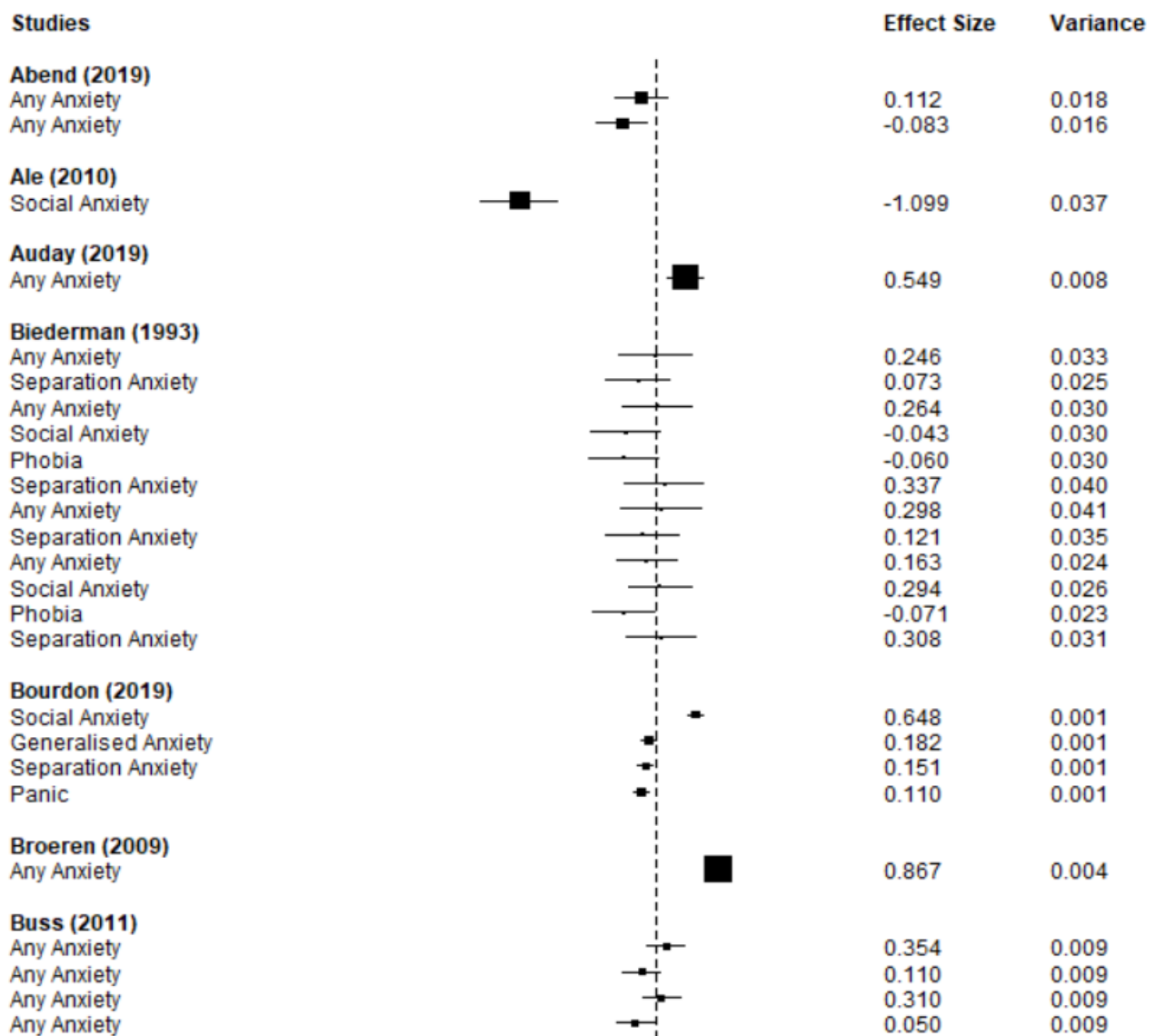
Number	Study	1. Question / objective sufficiently described?	2. Study design evident and appropriate?	3. Method of subject/comparison group selection or source of information/input variables described and appropriate?	4. Subject (and comparison group, if applicable) characteristics sufficiently described?	8. Outcome and (if applicable) exposure measure(s) well defined and robust to measurement / misclassification bias?	9. Sample size appropriate?	10. Analytic methods described/justified and appropriate?	11. Some estimate of variance is reported for the main results?	12. Controlled for confounding?	13. Results reported in sufficient detail?	14. Conclusions supported by the results?	Total score	Summary Score
42	Reeb-Sutherland (2009)	2	2	2	2	2	1	2	2	0	2	2	19	0.86
43	Reeb-Sutherland (2015)	2	2	2	2	2	1	2	0	2	2	2	19	0.86
44	Schwartz (1999)	2	2	2	2	2	1	2	0	0	2	2	17	0.77
45	Stumper (2017)	2	2	2	2	2	2	2	2	2	2	2	22	1.00
46	Suarez (2019)	2	2	2	2	2	2	2	2	0	2	2	20	0.91
47	Troller-Renfree (2019 [a])	2	2	2	2	2	2	2	2	2	2	2	22	1.00

ROLE OF TEMPERAMENT AND PARENTING BEHAVIOURS ON ANXIETY

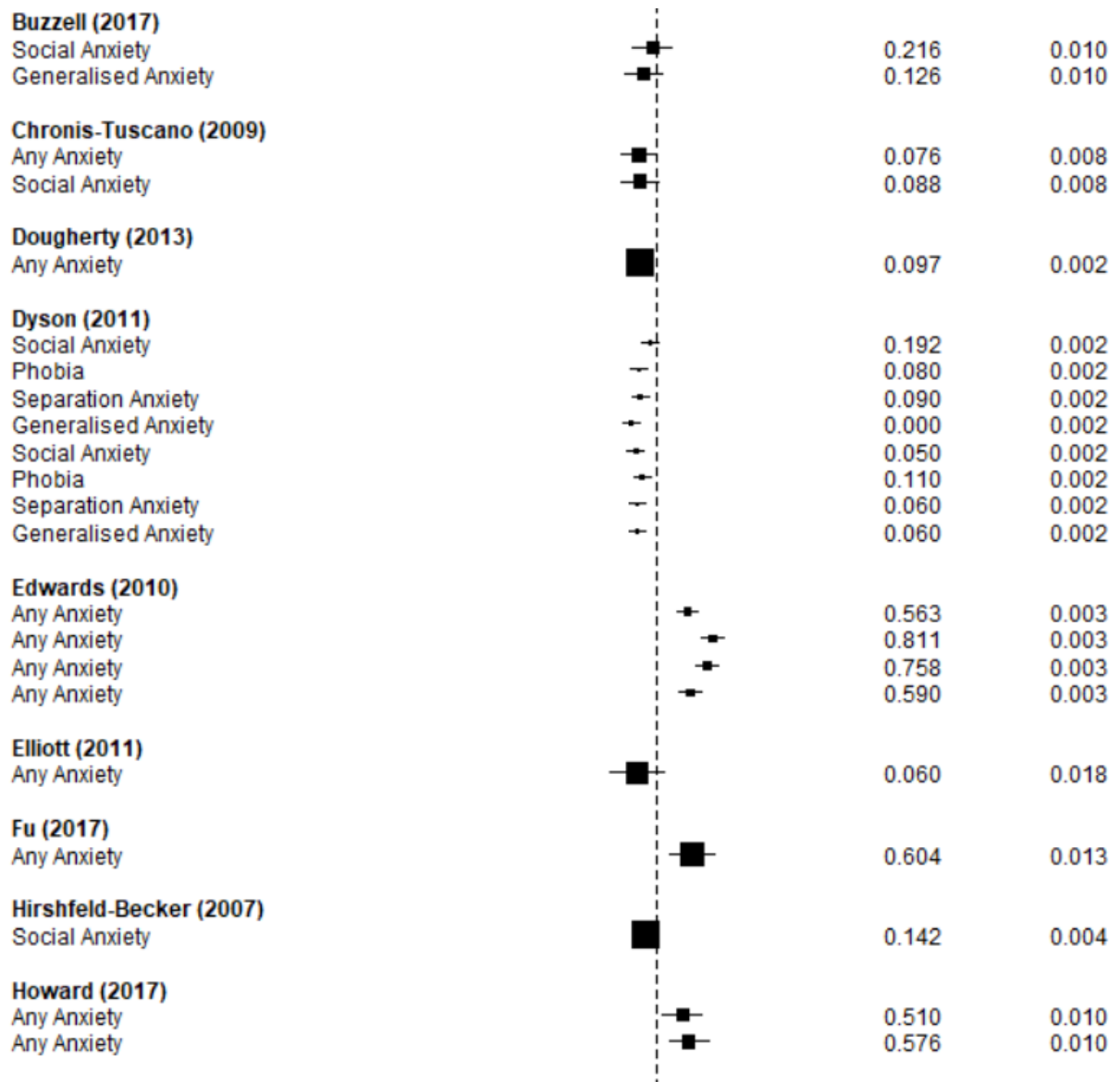
Number	Study	1. Question / objective sufficiently described?	2. Study design evident and appropriate?	3. Method of subject/comparison group selection or source of information/input variables described and appropriate?	4. Subject (and comparison group, if applicable) characteristics sufficiently described?	8. Outcome and (if applicable) exposure measure(s) well defined and robust to measurement / misclassification bias?	9. Sample size appropriate?	10. Analytic methods described/justified and appropriate?	11. Some estimate of variance is reported for the main results?	12. Controlled for confounding?	13. Results reported in sufficient detail?	14. Conclusions supported by the results?	Total score	Summary Score
48	Troller-Renfree (2019)	2	2	2	2	2	2	2	2	2	2	2	22	1.00
49	van der Linden (2013)	2	2	2	2	2	2	2	0	0	2	2	18	0.82
50	Volbrecht (2010)	2	2	2	2	2	2	2	2	2	2	2	22	1.00
51	Vreek and Muris (2012)	2	2	2	2	2	2	2	2	2	2	2	22	1.00
52	Vreeke (2012)	2	2	2	2	2	2	2	0	1	2	2	19	0.86
53	Vreeke (2013)	2	2	2	2	2	1	2	1	2	2	2	20	0.91
54	West (2007)	2	2	2	2	2	1	2	2	2	2	2	21	0.95
55	White (2017)	2	2	2	2	2	2	2	2	2	2	2	22	1.00

Appendix E – Forest Plot

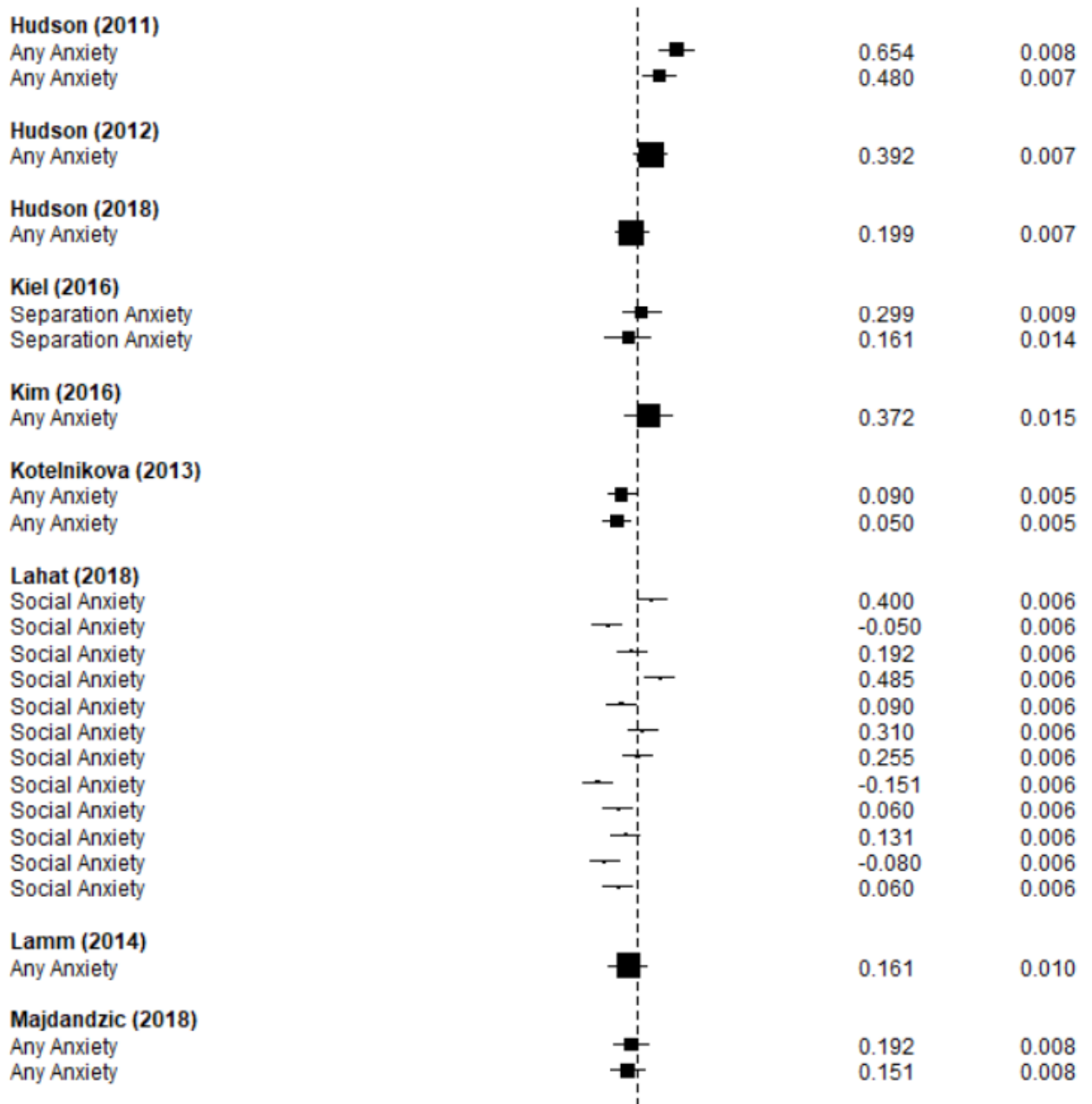
(extended over 6 pages - page 1 of 6)



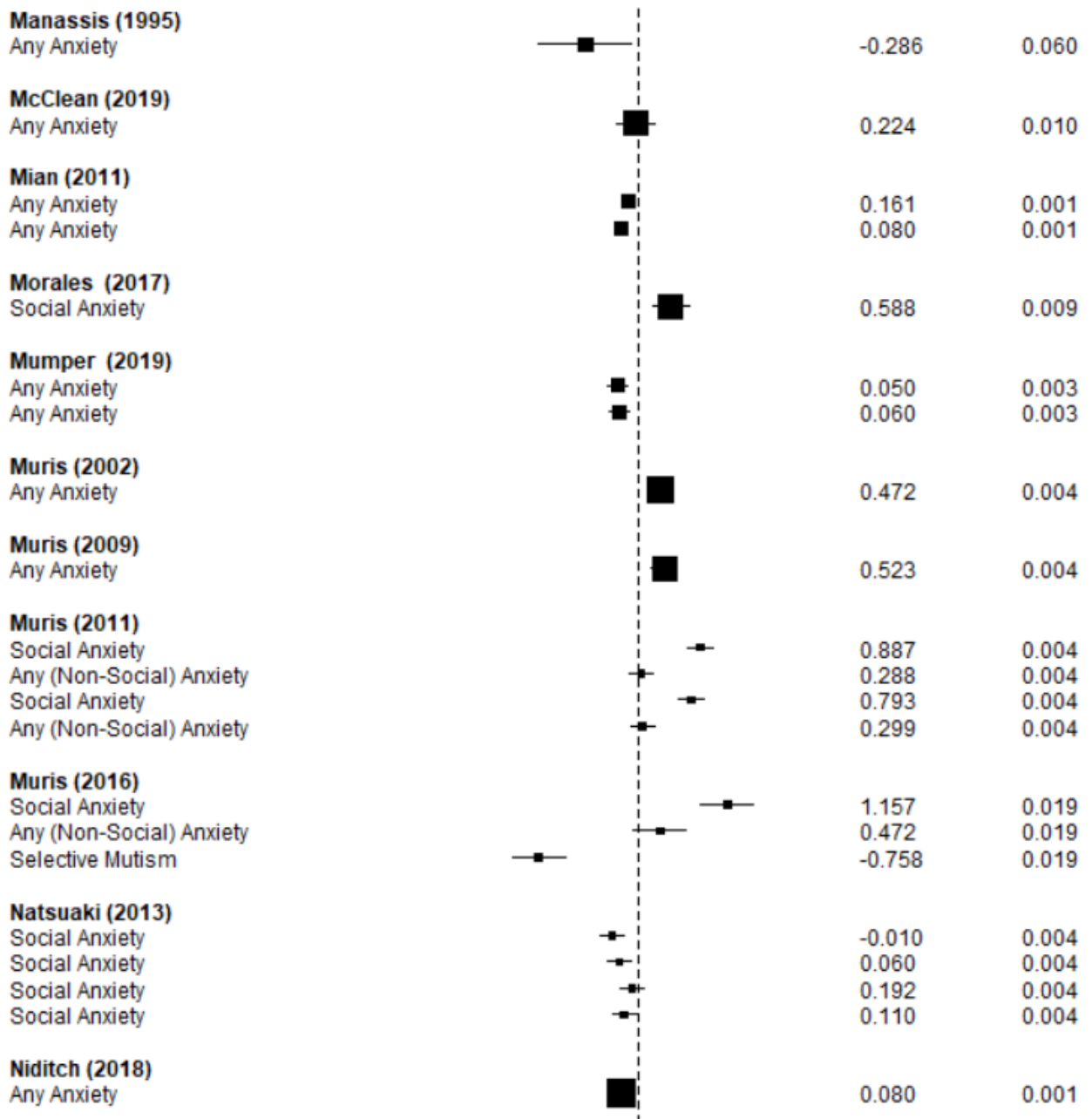
Appendix E – Forest Plot (page 2 of 6)



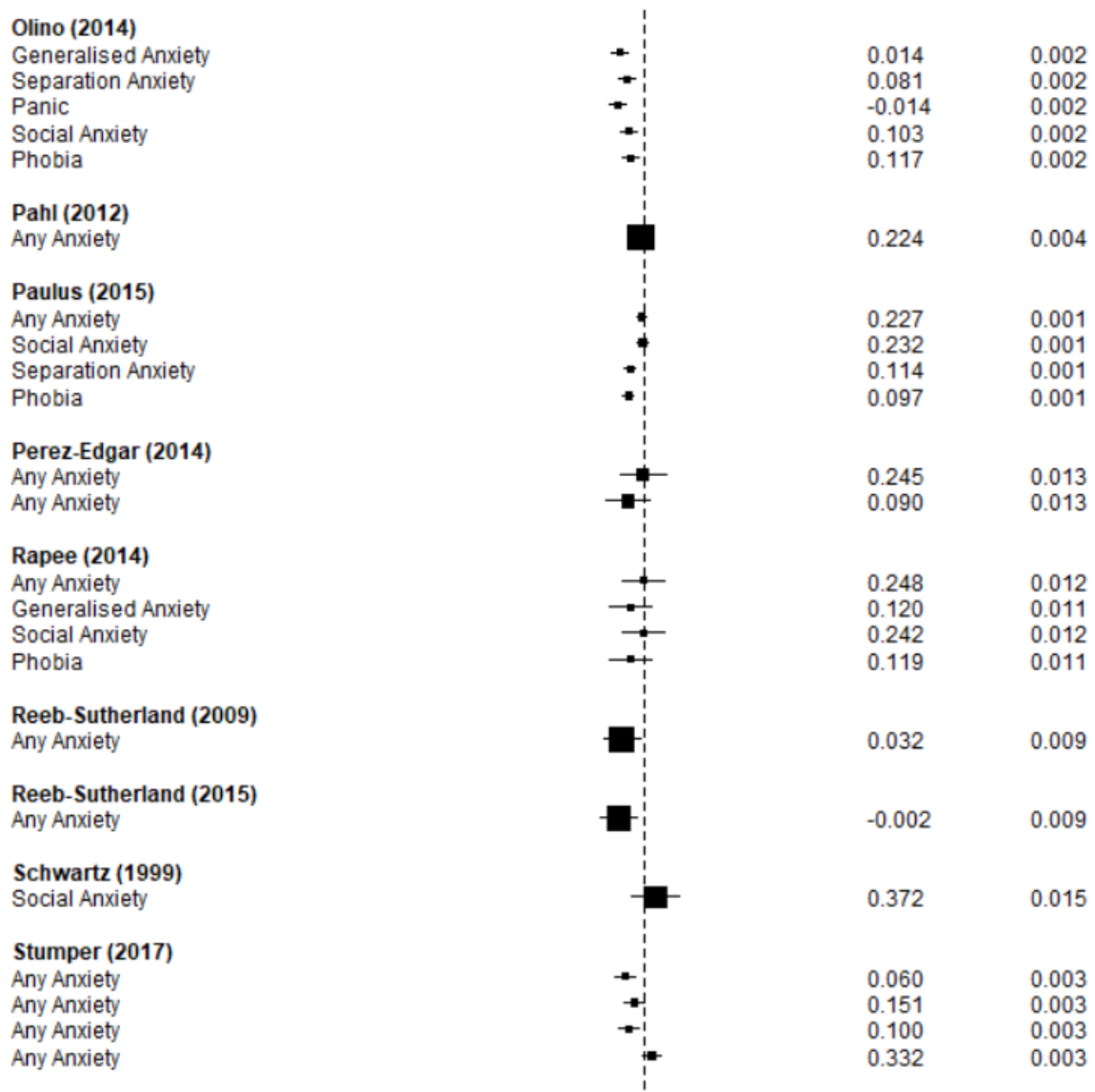
Appendix E – Forest Plot (page 3 of 6)



Appendix E – Forest Plot (page 4 of 6)



Appendix E – Forest Plot (page 5 of 6)



Appendix E – Forest Plot (page 6 of 6)

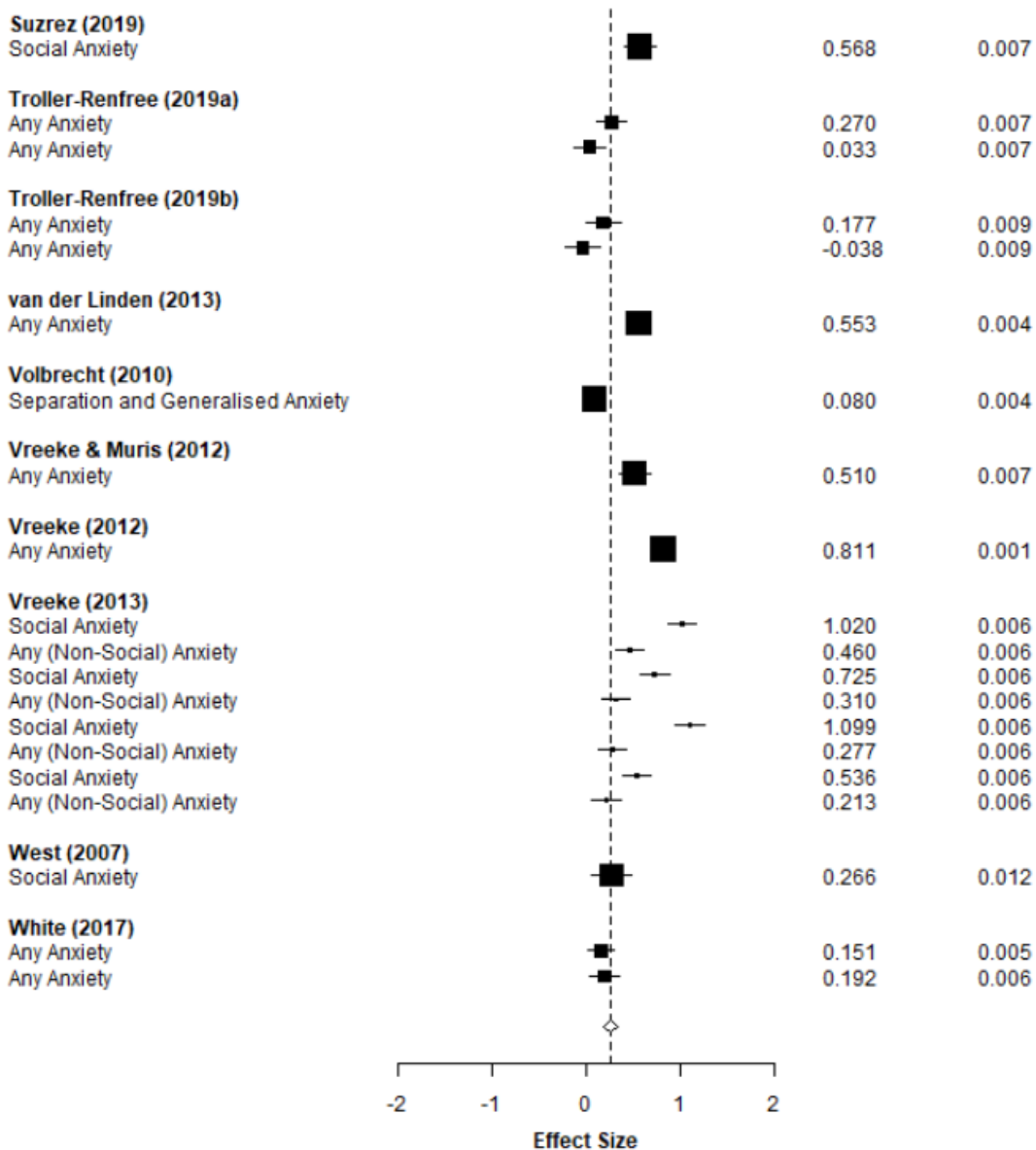


Figure 8. Forest Plot illustrating the effect size (Z) between BI and anxiety from 135 effect sizes from 55 studies.

Note. The position of the black square indicates the effect size, with horizontal lines marking the 95% confidence interval. The size of the black square indicates its weight in the meta-analysis. The name in bold is the first author of the published study along with the publication year. The type of anxiety outcome is listed under each study. The upper and lower 95% confidence intervals are shown in the two columns on the right-hand side. The white diamond with black outline represents the 95% confidence interval of the overall effect size.

Appendix F - Histograms for maternal mood and anxiety questionnaires

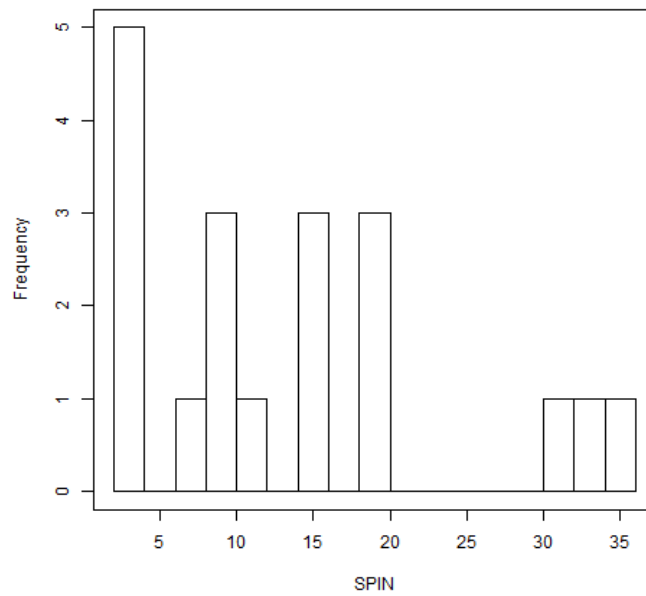


Figure 9. Histogram to illustrate responses to the Social Phobia Inventory (SPIN)

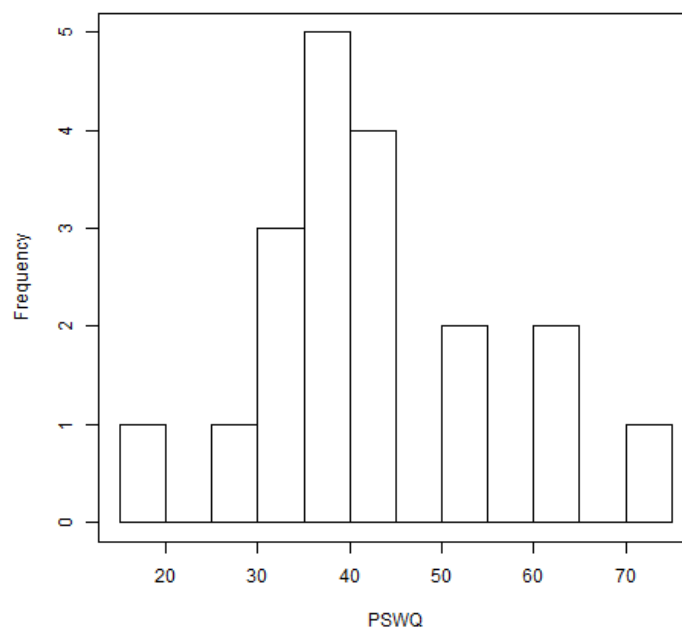


Figure 10. Histogram to illustrate responses to the Penn State Worry Questionnaire (PSWQ)

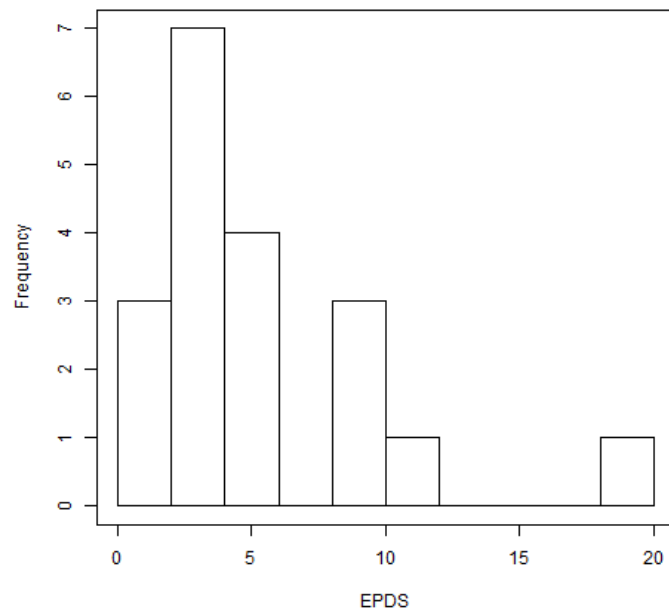


Figure 11. Histogram to illustrate responses to the Edinburgh Postnatal Depression Scale (EPDS)

References

- Ahmadzadeh, Y. I., Eley, T. C., Leve, L. D., Shaw, D. S., Natsuaki, M. N., Reiss, D., . . . McAdams, T. A. (2019). Anxiety in the family: a genetically informed analysis of transactional associations between mother, father and child anxiety symptoms. *J Child Psychol Psychiatry, 60*(12), 1269-1277. doi:10.1111/jcpp.13068
- Aktar, E., Majdandzic, M., de Vente, W., & Bogels, S. M. (2013). The interplay between expressed parental anxiety and infant behavioural inhibition predicts infant avoidance in a social referencing paradigm. *Journal Of Child Psychology And Psychiatry, And Allied Disciplines, 54*(2), 144-156. doi:<http://dx.doi.org/10.1111/j.1469-7610.2012.02601.x>
- Aktar, E., Majdandzic, M., de Vente, W., & Bogels, S. M. (2014). Parental social anxiety disorder prospectively predicts toddlers' fear/avoidance in a social referencing paradigm. *Journal Of Child Psychology And Psychiatry, And Allied Disciplines, 55*(1), 77-87.
- Altman, D. G., & Royston, P. (2006). The cost of dichotomising continuous variables. *BMJ, 332*(7549), 1080. doi:10.1136/bmj.332.7549.1080
- Ashford, J., Smit, F., van Lier, P. A., Cuijpers, P., & Koot, H. M. (2008). Early risk indicators of internalizing problems in late childhood: a 9-year longitudinal study. *J Child Psychol Psychiatry, 49*(7), 774-780. doi:10.1111/j.1469-7610.2008.01889.x
- Association for Video Interaction Guidance UK. (2020). About VIG. Retrieved from <https://www.videointeractionguidance.net/IMH-VIG-publications/>
- Baldwin, J. S., & Dadds, M. R. (2007). Reliability and validity of parent and child versions of the multidimensional anxiety scale for children in community samples. *J Am Acad Child Adolesc Psychiatry, 46*(2), 252-260. doi:10.1097/01.chi.0000246065.93200.a1
- Bates, M. R. J. (2006). Temperament. In R. L. N. E. W. Damon (Ed.), *Handbook of Child Psychology, Sixth Edition: Social, Emotional and Personality Development* (Vol. 3). New York: Wiley.
- Bates, T. (2006). The clinical management of social anxiety disorder. In A. C. M. McNulty (Ed.), *The Handbook of Adult Clinical Psychology: An Evidence-Based Practice Approach*. Hove, East Sussex: Routledge.
- Beidel, D. C., Turner, S. M., & Morris, T. L. (1999). Psychopathology of childhood social phobia. *J Am Acad Child Adolesc Psychiatry, 38*(6), 643-650. doi:10.1097/00004583-199906000-00010
- Bishop, G., Spence, S. H., & McDonald, C. (2003). Can parents and teachers provide a reliable and valid report of behavioral inhibition? *Child Dev, 74*(6), 1899-1917. doi:10.1046/j.1467-8624.2003.00645.x
- Borenstein, M., Hedges, L. V., Higgins, J. P. T., & Rothstein, H. R. (2009). Effect Sizes Based on Correlations. In *Introduction to Meta-Analysis*.
- Bourdon, J. L., Savage, J. E., Verhulst, B., Carney, D. M., Brotman, M. A., Pine, D. S., . . . Hettema, J. M. (2019). The Genetic and Environmental Relationship Between Childhood Behavioral Inhibition and Preadolescent Anxiety. *Twin Research & Human Genetics, 22*(1), 48-55. doi:10.1017/thg.2018.73

- Broeren, S., & Muris, P. (2010). A psychometric evaluation of the behavioral inhibition questionnaire in a non-clinical sample of Dutch children and adolescents. *Child Psychiatry Hum Dev, 41*(2), 214-229. doi:10.1007/s10578-009-0162-9
- Brown, T. A., Antony, M. M., & Barlow, D. H. (1992). Psychometric properties of the Penn State Worry Questionnaire in a clinical anxiety disorders sample. *Behav Res Ther, 30*(1), 33-37. doi:10.1016/0005-7967(92)90093-v
- Chorpita, B. F., Moffitt, C. E., & Gray, J. (2005). Psychometric properties of the Revised Child Anxiety and Depression Scale in a clinical sample. *Behav Res Ther, 43*(3), 309-322. doi:10.1016/j.brat.2004.02.004
- Chronis-Tuscano, A., Degnan, K. A., Pine, D. S., Perez-Edgar, K., Henderson, H. A., Diaz, Y., . . . Fox, N. A. (2009). Stable Early Maternal Report of Behavioral Inhibition Predicts Lifetime Social Anxiety Disorder in Adolescence. *Journal of the American Academy of Child and Adolescent Psychiatry, 48*(9), 928-935. doi:<http://dx.doi.org/10.1097/CHI.0b013e3181ae09df>
- Clauss, J. A., & Blackford, J. U. (2012). Behavioral inhibition and risk for developing social anxiety disorder: a meta-analytic study. *J Am Acad Child Adolesc Psychiatry, 51*(10), 1066-1075 e1061. doi:10.1016/j.jaac.2012.08.002
- Connor, K. M., Davidson, J. R., Churchill, L. E., Sherwood, A., Foa, E., & Weisler, R. H. (2000). Psychometric properties of the Social Phobia Inventory (SPIN). New self-rating scale. *Br J Psychiatry, 176*, 379-386. doi:10.1192/bjp.176.4.379
- Cooper, H. M., Hedges, L. V., & Valentine, J. C. (2008). *The Handbook of Research Synthesis and Meta-Analysis: 2nd edition.*
- Cox, J. L., Holden, J. M., & Sagovsky, R. (1987). Detection of postnatal depression. Development of the 10-item Edinburgh Postnatal Depression Scale. *Br J Psychiatry, 150*, 782-786. doi:10.1192/bjp.150.6.782
- Cox, N. J. (2008). Speaking Stata: Correlation with confidence, or Fisher's z revised. *The Stata Journal, 8*(3), 413 - 439.
- Creswell, C., Apetroaia, A., Murray, L., & Cooper, P. (2013). Cognitive, affective, and behavioral characteristics of mothers with anxiety disorders in the context of child anxiety disorder. *J Abnorm Psychol, 122*(1), 26-38. doi:10.1037/a0029516
- Creswell, C., Cruddace, S., Gerry, S., Gitau, R., McIntosh, E., Mollison, J., . . . Cooper, P. J. (2015). Treatment of childhood anxiety disorder in the context of maternal anxiety disorder: a randomised controlled trial and economic analysis. *Health Technol Assess, 19*(38), 1-184, vii-viii. doi:10.3310/hta19380
- de Rosnay, M., Cooper, P. J., Tsigaras, N., & Murray, L. (2006). Transmission of social anxiety from mother to infant: an experimental study using a social referencing paradigm. *Behav Res Ther, 44*(8), 1165-1175. doi:10.1016/j.brat.2005.09.003
- de Wilde, A., & Rapee, R. M. (2008). Do controlling maternal behaviours increase state anxiety in children's responses to a social threat? A pilot study. *J Behav Ther Exp Psychiatry, 39*(4), 526-537. doi:10.1016/j.jbtep.2007.10.011

- Dodd, H. F., Hudson, J. L., & Rapee, R. M. (2017). Temperament in youth internalising disorders. In M. D., J. S. Abramowitz, & E. A. Storch (Eds.), *Treatments for Psychological Problems and Syndromes* (pp. 504 - 524). West Sussex: John Wiley & Sons Ltd.
- Donovan, C. L., & Spence, S. H. (2000). Prevention of childhood anxiety disorders. *Clin Psychol Rev*, 20(4), 509-531.
- Dyson, M. W., Klein, D. N., Olino, T. M., Dougherty, L. R., & Durbin, C. E. (2011). Social and non-social behavioral inhibition in preschool-age children: Differential associations with parent-reports of temperament and anxiety. *Child Psychiatry and Human Development*, 42(4), 390-405. doi:10.1007/s10578-011-0225-6
- Eley, T. C., McAdams, T. A., Rijdsdijk, F. V., Lichtenstein, P., Narusyte, J., Reiss, D., . . . Neiderhiser, J. M. (2015). The Intergenerational Transmission of Anxiety: A Children-of-Twins Study. *Am J Psychiatry*, 172(7), 630-637. doi:10.1176/appi.ajp.2015.14070818
- Essau, C. A., Lewinsohn, P. M., Lim, J. X., Ho, M. R., & Rohde, P. (2018). Incidence, recurrence and comorbidity of anxiety disorders in four major developmental stages. *J Affect Disord*, 228, 248-253. doi:10.1016/j.jad.2017.12.014
- Ewing, D., Pike, A., Dash, S., Hughes, Z., Thompson, E. J., Hazell, C., . . . Cartwright-Hatton, S. (2019). Helping parents to help children overcome fear: The influence of a short video tutorial. *Br J Clin Psychol*. doi:10.1111/bjc.12233
- Feinman, S. (1982). Social referencing in infancy. *Merrill-Palmer Quarterly*, 28(4), 445 - 470.
- Fisher, Z., & Tipton, E. (2015). robumeta: An R-package for robust variance estimation in meta-analysis. Retrieved from <https://arxiv.org/pdf/1503.02220.pdf>
- Gerull, F. C., & Rapee, R. M. (2002). Mother knows best: effects of maternal modelling on the acquisition of fear and avoidance behaviour in toddlers. *Behav Res Ther*, 40(3), 279-287. doi:10.1016/s0005-7967(01)00013-4
- Gray, J. A. (1970). The psychophysiological basis of introversion-extraversion. *Behav Res Ther*, 8(3), 249-266. doi:10.1016/0005-7967(70)90069-0
- Heckman, J. J. (2000). Policies to foster human capital. *Research in Economics*, 54, 3-56.
- Hedges, L. V., Tipton, E., & Johnson, M. C. (2010). Robust variance estimation in meta-regression with dependent effect size estimates. *Res Synth Methods*, 1(1), 39-65. doi:10.1002/jrsm.5
- Hudson, J. L., & Dodd, H. F. (2012). Informing early intervention: Preschool predictors of anxiety disorders in middle childhood. *PLoS ONE*, 7(8). doi:10.1371/journal.pone.0042359
- Hudson, J. L., Dodd, H. F., & Bovopoulos, N. (2011). Temperament, family environment and anxiety in preschool children. *Journal of Abnormal Child Psychology*, 39(7), 939-951. doi:10.1007/s10802-011-9502-x
- Hudson, J. L., Dodd, H. F., Lyneham, H. J., & Bovopoulos, N. (2011). Temperament and family environment in the development of anxiety disorder: Two-year follow-up. *Journal of the American Academy of Child & Adolescent Psychiatry*, 50(12), 1255-1264. doi:10.1016/j.jaac.2011.09.009

- Hudson, J. L., Murayama, K., Meteyard, L., Morris, T., & Dodd, H. F. (2019). Early childhood predictors of anxiety in early adolescence. *Journal of Abnormal Child Psychology*, 47(7), 1121-1133. doi:10.1007/s10802-018-0495-6
- Kagan, J. (2018). The History and Theory of Behavioral Inhibition. In K. F. Perez-Edgar, N. A. (Ed.), *Behavioral Inhibition: Integrating Theory, Research, and Clinical Perspectives*.
- Kagan, J., Reznick, J. S., Clarke, C., Snidman, N., & Garcia-Coll, C. (1984). Behavioral inhibition to the unfamiliar. *Child Development*, 55, 2212 - 2222.
- Kessler, R. C., Berglund, P., Demler, O., Jin, R., Merikangas, K. R., & Walters, E. E. (2005). Lifetime prevalence and age-of-onset distributions of DSM-IV disorders in the National Comorbidity Survey Replication. *Arch Gen Psychiatry*, 62(6), 593-602. doi:10.1001/archpsyc.62.6.593
- Kim, J., Klein, D. N., Olino, T. M., Dyson, M. W., Dougherty, L. R., & Durbin, C. E. (2011). Psychometric properties of the Behavioral Inhibition Questionnaire in preschool children. *J Pers Assess*, 93(6), 545-555. doi:10.1080/00223891.2011.608756
- Klein, D. N., & Mumper, E. E. (2018). Behavioral Inhibition as a Precursor to Psychopathology. In K. Perez-Edgar & N. A. Fox (Eds.), *Behavioral Inhibition: Integrating Theory, Research, and Clinical Perspectives* (pp. 283 - 307). Switzerland: Springer.
- Kmet, L. M., Lee, R. C., & Cook, L. S. (2004). *Standard quality assessment criteria for evaluating primary research papers from a variety of fields*. Edmonton, AB, Canada.
- Kostyrka-Allchorne, K., Wass, S. V., & Sonuga-Barke, E. J. S. (2020). Research Review: Do parent ratings of infant negative emotionality and self-regulation predict psychopathology in childhood and adolescence? A systematic review and meta-analysis of prospective longitudinal studies. *J Child Psychol Psychiatry*, 61(4), 401-416. doi:10.1111/jcpp.13144
- Landis, J. R., & Koch, G. G. (1977). An application of hierarchical kappa-type statistics in the assessment of majority agreement among multiple observers. *Biometrics*, 33(2), 363-374.
- Lawrence, P. J., Creswell, C., Cooper, P. J., & Murray, L. (2020). The role of maternal anxiety disorder subtype, parenting and infant stable temperamental inhibition in child anxiety: a prospective longitudinal study. *J Child Psychol Psychiatry*. doi:10.1111/jcpp.13187
- Lemery, K. S., Essex, M. J., & Smider, N. A. (2002). Revealing the relation between temperament and behavior problem symptoms by eliminating measurement confounding: expert ratings and factor analyses. *Child Dev*, 73(3), 867-882. doi:10.1111/1467-8624.00444
- Meyer, T. J., Miller, M. L., Metzger, R. L., & Borkovec, T. D. (1990). Development and validation of the Penn State Worry Questionnaire. *Behav Res Ther*, 28(6), 487-495. doi:10.1016/0005-7967(90)90135-6
- Möller, E. L., Nikolić, M., Majdandžić, M., & Bögels, S. M. (2016). Associations between maternal and paternal parenting behaviors, anxiety and its precursors in early childhood: A meta-analysis. *Clinical Psychology Review*, 45, 17-33. doi:10.1016/j.cpr.2016.03.002
- Muris, P., Meesters, C., & Spinder, M. (2003). Relationships between child- and parent-reported behavioural inhibition and symptoms of anxiety and depression in normal adolescents. *Personality and Individual Differences*, 34(5), 759-771. doi:10.1016/S0191-8869(02)00069-7

- Murray, L., Cooper, P., Creswell, C., Schofield, E., & Sack, C. (2007). The effects of maternal social phobia on mother-infant interactions and infant social responsiveness. *J Child Psychol Psychiatry*, 48(1), 45-52. doi:10.1111/j.1469-7610.2006.01657.x
- Murray, L., Creswell, C., & Cooper, P. J. (2009). The development of anxiety disorders in childhood: an integrative review. *Psychological Medicine*, 39(9), 1413-1423. doi:10.1017/S0033291709005157
- Murray, L., de Rosnay, M., Pearson, J., Bergeron, C., Schofield, E., Royal-Lawson, M., & Cooper, P. J. (2008). Intergenerational transmission of social anxiety: The role of social referencing processes in infancy. *Child Development*, 79(4), 1049-1064. doi:10.1111/j.1467-8624.2008.01175.x
- Murray, L., Lau, P. Y., Arteche, A., Creswell, C., Russ, S., Zoppa, L. D., . . . Cooper, P. (2012). Parenting by anxious mothers: effects of disorder subtype, context and child characteristics. *Journal Of Child Psychology And Psychiatry, And Allied Disciplines*, 53(2), 188-196. doi:<http://dx.doi.org/10.1111/j.1469-7610.2011.02473.x>
- Nauta, M. H., Scholing, A., Rapee, R. M., Abbott, M., Spence, S. H., & Waters, A. (2004). A parent-report measure of children's anxiety: psychometric properties and comparison with child-report in a clinic and normal sample. *Behav Res Ther*, 42(7), 813-839. doi:10.1016/S0005-7967(03)00200-6
- NHS Digital. (2018). Mental Health of Children and Young People in England, 2017. Retrieved from <https://digital.nhs.uk/data-and-information/publications/statistical/mental-health-of-children-and-young-people-in-england/2017/2017>
- Ollendick, T. H., King, N. J., & Yule, W. (1994). *International handbook of phobic and anxiety disorders in children and adolescents*. New York: Plenum.
- Pahl, K. M., Barrett, P. M., & Gullo, M. J. (2012). Examining potential risk factors for anxiety in early childhood. *Journal of Anxiety Disorders*, 26(2), 311-320. doi:10.1016/j.janxdis.2011.12.013
- Planalp, E. M., Van Hulle, C., Gagne, J. R., & Goldsmith, H. H. (2017). The Infant Version of the Laboratory Temperament Assessment Battery (Lab-TAB): Measurement Properties and Implications for Concepts of Temperament. *Front Psychol*, 8, 846. doi:10.3389/fpsyg.2017.00846
- Polanczyk, G. V., Salum, G. A., Sugaya, L. S., Caye, A., & Rohde, L. A. (2015). Annual research review: A meta-analysis of the worldwide prevalence of mental disorders in children and adolescents. *J Child Psychol Psychiatry*, 56(3), 345-365. doi:10.1111/jcpp.12381
- Public Health England. (2016). The mental health of children and young people in England. Retrieved from https://assets.publishing.service.gov.uk/government/uploads/system/uploads/attachment_data/file/575632/Mental_health_of_children_in_England.pdf
- Putnam, S. P., Helbig, A. L., Gartstein, M. A., Rothbart, M. K., & Leerkes, E. (2014). Development and assessment of short and very short forms of the infant behavior questionnaire-revised. *J Pers Assess*, 96(4), 445-458. doi:10.1080/00223891.2013.841171

- Ramsawh, H. J., & Chavira, D. A. (2016). Association of Childhood Anxiety Disorders and Quality of Life in a Primary Care Sample. *J Dev Behav Pediatr*, 37(4), 269-276. doi:10.1097/DBP.0000000000000296
- Rapee, R. (2001). The development of generalized anxiety. In M. V. a. M. Dadds (Ed.), *The Developmental Psychopathology of Anxiety*. Oxford, UK: Oxford University Press.
- Rapee, R. M. (2013). The preventative effects of a brief, early intervention for preschool-aged children at risk for internalising: Follow-up into middle adolescence. *Journal of Child Psychology and Psychiatry*, 54(7), 780-788. doi:10.1111/jcpp.12048
- Rapee, R. M., Kennedy, S., Ingram, M., Edwards, S., & Sweeney, L. (2005). Prevention and early intervention of anxiety disorders in inhibited preschool children. *Journal of Consulting and Clinical Psychology*, 73(3), 488-497. doi:<http://dx.doi.org/10.1037/0022-006X.73.3.488>
- Rothbart, M. K., & Bates, J. E. (2006). Temperament. In N. Eisenberg, W. Damon, & R. M. Lerner (Eds.), *Handbook of child psychology: Social, emotional, and personality development*. USA: John Wiley & Sons Inc.
- Ryan, S. M., & Ollendick, T. H. (2018). The Interaction Between Child Behavioral Inhibition and Parenting Behaviors: Effects on Internalizing and Externalizing Symptomology. *Clinical Child & Family Psychology Review*, 21(3), 320-339. doi:10.1007/s10567-018-0254-9
- Sandstrom, A., Uher, R., & Pavlova, B. (2019). Prospective Association between Childhood Behavioral Inhibition and Anxiety: a Meta-Analysis. *J Abnorm Child Psychol*. doi:10.1007/s10802-019-00588-5
- Stein, A., Craske, M. G., Lehtonen, A., Harvey, A., Savage-McGlynn, E., Davies, B., . . . Counsell, N. (2012). Maternal cognitions and mother-infant interaction in postnatal depression and generalized anxiety disorder. *J Abnorm Psychol*, 121(4), 795-809. doi:10.1037/a0026847
- Stein, A., Netsi, E., Lawrence, P. J., Granger, C., Kempton, C., Craske, M. G., . . . Murray, L. (2018). Mitigating the effect of persistent postnatal depression on child outcomes through an intervention to treat depression and improve parenting: a randomised controlled trial. *Lancet Psychiatry*, 5(2), 134-144. doi:10.1016/S2215-0366(18)30006-3
- Swinson, R. P. (1997). Developments in anxiety disorders. *Can J Psychiatry*, 42(10), 1015-1016. doi:10.1177/070674379704201001
- Tandon, M., Cardeli, E., & Luby, J. (2009). Internalizing disorders in early childhood: a review of depressive and anxiety disorders. *Child Adolesc Psychiatr Clin N Am*, 18(3), 593-610. doi:10.1016/j.chc.2009.03.004
- Thirwall, K., , & Creswell, C. (2010). The impact of mental control on children's anxious cognitions, behaviours and affect: an experimental study. *Behaviour Research and Therapy*, 48, 1041 - 1046.
- Towe-Goodman, N. R., Franz, L., Copeland, W., Angold, A., & Egger, H. (2014). Perceived family impact of preschool anxiety disorders. *J Am Acad Child Adolesc Psychiatry*, 53(4), 437-446. doi:10.1016/j.jaac.2013.12.017

- van der Bruggen, C. O., Stams, G. J., & Bogels, S. M. (2008). Research review: the relation between child and parent anxiety and parental control: a meta-analytic review. *J Child Psychol Psychiatry, 49*(12), 1257-1269. doi:10.1111/j.1469-7610.2008.01898.x
- Viechtbauer, W. (2010). Conducting Meta-Analyses in R with the metafor Package. *Journal of Statistical Software, 36*(3), 1 - 48.
- Wei, C., Hoff, A., Villabo, M. A., Peterman, J., Kendall, P. C., Piacentini, J., . . . March, J. (2014). Assessing anxiety in youth with the multidimensional anxiety scale for children. *J Clin Child Adolesc Psychol, 43*(4), 566-578. doi:10.1080/15374416.2013.814541
- Wilson, D. B. (n.d.). Practical Meta-Analysis Effect Size Calculator [Online calculator]. Retrieved from <https://campbellcollaboration.org/research-resources/effect-size-calculator.html>
- Yap, M. B., & Jorm, A. F. (2015). Parental factors associated with childhood anxiety, depression, and internalizing problems: a systematic review and meta-analysis. *J Affect Disord, 175*, 424-440. doi:10.1016/j.jad.2015.01.050