The Hidden Epidemic of Fetal Alcohol Spectrum Disorder: Rates of co-occurring psychological needs of children and clinicians' understanding of assessment and diagnosis.

Emma Heathcote

Candidate registration number: 100373528

Primary Supervisor: Dr Aaron Burgess

Secondary Supervisor: Dr Kenny Chiu

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Norwich Medical School, University of East Anglia

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Thesis Portfolio Abstract

Research shows that the teratogenic effects of alcohol on a developing foetus can have widespread effects, with the potential to develop a range of disorders falling under the umbrella term Fetal Alcohol Spectrum Disorders (FASD). Children with FASD can experience a variety of difficulties, including mental health problems, behavioural and neurodevelopmental difficulties. A meta-analysis was undertaken to understand the prevalence of co-occurring psychological needs among this population. The effects of location of study and method of data collection on prevalence rated were explored with moderator analyses. The highest prevalence rates were found for Attention Deficit Hyperactivity Disorder (ADHD) and behavioural difficulties, although results are considered in the context of high heterogeneity. Another reason for the high prevalence of co-occurring ADHD, is the similar presentations of both disorders. Evidence suggests that FASD is currently missed or misdiagnosed for disorders such as ADHD, for reasons such as a lack of confidence in diagnosing FASD. An online experiment, involving a clinical vignette of a referral about a young person with neurodevelopmental concerns, was conducted to explore the facilitators and barriers associated with clinicians considering FASD as a diagnosis. In particular, the study explored whether additional information regarding prenatal alcohol exposure impacted on the number of clinicians considering a diagnosis of FASD. Overall, clinicians were more likely to consider diagnoses of disorders such as ADHD when presented with the vignette, rather than FASD. This was the case even when the referral contained information of prenatal alcohol exposure, although this significantly increased the number of FASD considerations. Further training in the assessment and diagnosis of FASD is essential for supporting clinicians to hold in mind FASD as a diagnosis, to enable early intervention for children. Clinical implications for both the meta-analysis and empirical study are discussed.

Table of Contents

List of Tables

List of figures

List of Appendices

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A big thank you to my lovely friends and family for their unconditional love and support, including childcare! Thank you to my Mum and Dad, who have always encouraged me to chase my dreams. Mum, the chocolate muffins every study day were the boosts I needed! A huge thank you to my husband for all your support throughout this journey, you had to fill multiple roles during this time including being practically a single parent, chef, cleaner, and my biggest cheerleader. Your sacrifice has not gone unnoticed, and I am eternally grateful. Lastly, the biggest thank you to my special little girl, whose energy and affection were the perfect antidotes to many a stressful day! I was often asked how I was coping with the competing demands of the doctorate and being a Mum. The truth is, having a mischievous toddler helped to keep everything in perspective and was the best distraction I could wish for!

Chapter 1: Introduction to Thesis Portfolio

The number one cause of non-genetic learning disabilities in the United Kingdom, are a collection of neurodevelopmental disorders which fall under the umbrella term, Fetal Alcohol Spectrum Disorders (FASD; British Medical Association, 2016). These entirely preventable conditions are caused by a developing fetus being exposed to alcohol in the womb, and the effects are incredibly widespread. A systematic review by Popova et al. (2016) found that over 420 conditions can co-occur with FASD, due to the potential impact on every system in the body.

The umbrella term encompasses a spectrum of disorders which vary in the severity of prenatal alcohol effects. Fetal Alcohol Syndrome (FAS), which was first recognised in 1973 (Jones & Smith, 1973; Jones et al., 1973), within the United States of America, is the most severe form and is characterised by central nervous system (CNS) problems, growth problems, distinct facial features, problems with memory, communication, learning and social interaction. Following FAS are a range of other disorders which present with a continuum of effects across many of these areas, as a result of prenatal alcohol exposure (PAE): Partial FAS (PFAS), Alcohol-Related Neurodevelopmental Disorder (ARND), Neurodevelopmental Disorder Associated with Prenatal Alcohol Exposure (ND-PAE), and Alcohol Related Birth Defects (ARBD) (Hoyme et al., 2016). In more recent years, these terms have been simplified to FASD with sentinel facial features (short palpebral fissures, a smooth or flat philtrum, and a thin upper lip, see Figure 1) or FASD without sentinel facial features (Department of Health and Social Care, 2021). This change has been accepted by the Scottish Intercollegiate Guidelines Network (SIGN) 156 (SIGN, 2019), the guidelines which have shaped the National Institute for Health and Care Excellence (NICE) quality standards for the United Kingdom (NICE, 2022). These standards document the support and advice that should be given to pregnant women, and what an assessment should include for an appropriate diagnosis of FASD (see Chapter 4 for further information), and are expected to accept the changes to diagnosis in line with the SIGN 156.

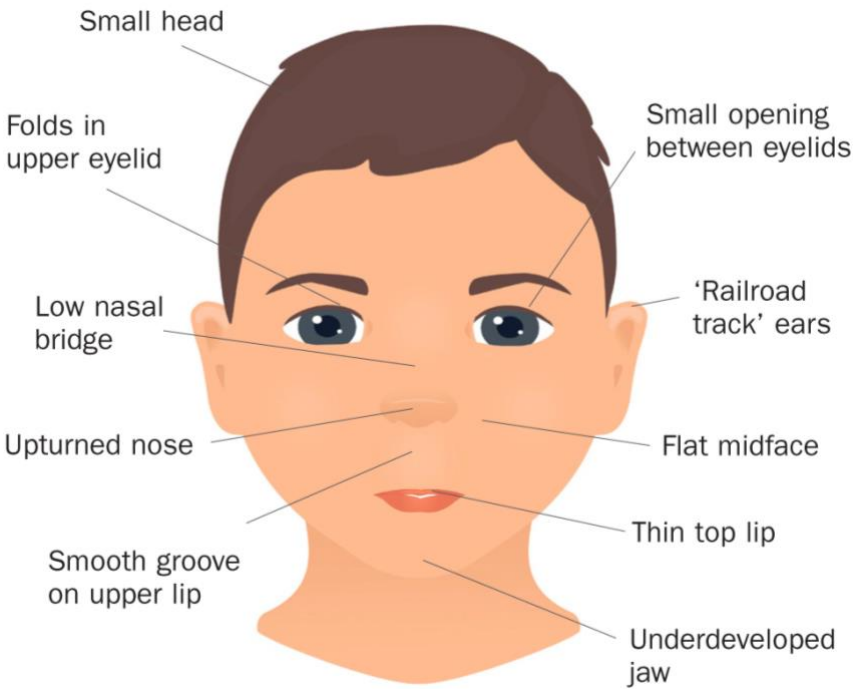

Figure 1: Representation of FASD with sentinel facial features (Kruithof & Ban, 2021).

Whilst the disorder is preventable, there are a range of reasons for its existence. Firstly, research has indicated that there are no safe limits of alcohol consumption during pregnancy, which is reported in government guidance documents (NICE, 2022). However, focus groups held by Mukherjee et al. (2015) found that common perception amongst health professionals is that safe consumption levels are unknown. Because of this, pregnant women are often told conflicting advice, or maybe not advised at all. Mukherjee and colleagues (2015) reported that that 72.5% of health professionals did not feel that they had enough information to advise patients about safe alcohol consumption. Interestingly, this study also found that a number of health professionals who may be involved within the perinatal period, felt that FASD was not relevant to their role.

Moreover, the prevalence of FASD in particular settings indicate that low socioeconomic status plays an important role in the risk of FASD. A review by Abel (1995) found that FAS rates were 10 times higher for low SES populations when compared to middle/upper class populations. This is an interesting finding, due to research indicating that women from upper SES groups are more likely to drink alcohol during their pregnancy. This has led to the "multiple hit" hypothesis, by which the combination of PAE as well as factors associated with SES groups, such as poverty and trauma, are at higher risk of poor outcomes (Yumoto et al., 2008), and FASD (Moritz et al., 2023). Other risk factors which are associated with FAS include higher gravidity (total number of pregnancies) and parity (total number of births of pregnancies which have reached viable gestational age), living with or

associating with others who drink heavily, older maternal age, and the physical attributes of the mother including low body mass index (BMI) (May et al., 2008).

With preventability also comes the shadow of guilt and self-blame by mothers. This in turn results in the disorder having an attached stigma, further preventing families coming forward for support (British Medical Association Board of Science, 2016), making the true prevalence of FASD difficult to determine. Among this, May et al. (2009) reports that other reasons include the variance in methodologies, lack of complete data sets and the lack of screening which is accurate and routine in antenatal clinics has resulted in FASD being undetected in children, leading to Clarren and Lutke (2008) describing it as the "hidden epidemic".

A further complication contributing to diagnosis and prevalence difficulties, is the variety of different classification standards that are adopted. For example, worldwide, diagnostic clinics utilise different FASD diagnostic systems to assess for FASD. These include systems such as Hoyme et al. (2016), the 4-Digit Code (Astley, 2004), and the Canadian Guidelines (Cook et al., 2016). Whilst there are similarities between the systems, Astley et al. (2017) reported that only 38% of participants received the same diagnosis, when the 4-Digit code and Hoyme et al. (2016) classifications were compared. This poses the question as to the true prevalence of FASD, which has been debated in literature and is still relatively unknown (see Chapters 2 and 4), when compared to other neurodevelopmental disorders such as Autism Spectrum Disorders (ASD).

As stated, the effects of FASD are widespread and thus is it imperative that the research field dedicates resources to understanding how to support with early diagnosis to ensure early intervention, which are dependent on the needs of children with FASD.

This thesis portfolio[1] explores the needs of children diagnosed with FASD and investigates the practice of diagnosing the disorder. Within the second chapter, a meta-analysis is presented which studies the prevalence of neurodevelopmental, behavioural and mental health difficulties reported by children with FASD, including those with and without official diagnoses within these domains. For the purpose of this thesis, these domains will be named psychological needs. Moderator analysis explored the impact of different methods of data collection (active versus passive methods) on the reported rates of prevalence. Due to a large number of studies occurring in particular geographical locations, an exploratory moderator analysis also took place to investigate whether this had any impact on the reported prevalence rates.

Following this, a bridging chapter introduces the five quality standards issued by NICE (2022) and the possible implications of their introduction, including an increase in referrals to neurodevelopmental assessment centres. With this in mind, Chapter 4 presents an empirical project

[1] Material from the first author's ClinPsyD Thesis Proposal has been used throughout this portfolio.

exploring the decision making around diagnosis and assessment of FASD by a range of clinical professionals. Within Chapters 5 and 6, additional methodology and results are presented from the empirical project and meta-analysis. The final chapter summarises the findings from both projects and concludes the thesis, offering suggestions for future research and clinical implications. Appendices for both projects are provided at the end of the portfolio. A list of abbreviations which are commonly used throughout this portfolio is also provided at the end for the reader's convenience.

Chapter 2: Meta-analysis

The prevalence of mental health, behavioural and neurodevelopmental difficulties in children with Fetal Alcohol Spectrum Disorder: A Meta-Analysis

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Emma Heathcote

Abstract

Background: Fetal Alcohol Spectrum Disorders (FASD) are a group of neurodevelopmental conditions caused by prenatal alcohol exposure to a developing fetus. Given the limited exploration of internalising disorders which co-occur with FASD, in addition to the limited use of non-clinic samples within the literature base, this study investigated the prevalence of psychological needs which fell under the following umbrella terms: depression, anxiety, behavioural difficulties, Attention Deficit Hyperactivity Disorder (ADHD) and Autism Spectrum Disorder (ASD). This was inclusive of self-report measures, broadening the investigation.

Methods: Searches were conducted across four databases, which yielded 50 (*N*=39071) studies after exclusion criteria had been applied. Moderator analyses were conducted to explore the impact of the location of study and the method of data collection (active versus passive) on prevalence rates. Sensitivity analysis was also conducted to account for risk of bias.

Results: Pooled prevalence rates and confidence intervals (CI) were found to be similar or higher than previously reported: depression=13.3% (95% *CI*: 8.7-18.6), anxiety=18.9% (95% *CI*: 10.8-28.5), behavioural difficulties=22.8% (95% *CI*: 13.2-34.1), ADHD=56.6% (95% *CI*: 49.9-63.1), ASD=14.8% (95% *CI*: 5.4-27.4), with no statistically significant differences found from either moderator analysis. These results are considered within the context of high heterogeneity.

Conclusions: High prevalence rates were found for children with FASD and co-occurring psychological needs, indicating that screening for these needs is important for support and treatment to be accessed.

Keywords: FASD, alcohol, externalising, internalising, prevalence

Introduction

The teratogenic effects of alcohol when consumed during pregnancy, has the ability to impact on any organ or system in the body. The results of this are incredibly widespread; Popova et al. (2016) found that there are over 420 conditions which can co-occur with Fetal Alcohol Spectrum Disorder (FASD), an umbrella term for a range of disorders experienced by individuals who were prenatally exposed to alcohol. Most commonly, individuals with FASD can present with a range of externalising behaviours, including aggression, destruction of property and violation of social norms (Lange et al., 2018). Unsurprisingly, due to these behaviours being considered challenging for society in terms of management and cost, much of the research base for FASD focuses on these behaviours, adding to the stigma associated with FASD (Corrigan et al., 2018). A focus on externalising behaviours can also lead to misdiagnosis and missed diagnosis. Chasnoff et al. (2015) found 86.5% of 156 children and adolescents meeting the criteria for FASD had never received a diagnosis previously, or were misdiagnosed, typically with the externalising disorder, Attention Deficit Hyperactivity Disorder (ADHD). This is likely to impact certain populations greater than others, such as African American children, who are more likely to be diagnosed externalising disorders such as Conduct Disorder (CD) or Oppositional Defiant Disorder (ODD), rather than neurodevelopmental diagnoses which are often missed (Baglivio et al., 2016).

Moreover, because of the perceived impact on society that externalising behaviours bring, the proportion of FASD individuals entering the justice system is high. Streissguth et al. (2004) reported that 60% of individuals assessed to meet the diagnostic criteria for Fetal Alcohol Syndrome (FAS) or had Fetal Alcohol Effects (FAE), had had contact with law enforcement services, with 35% also incarcerated. These difficulties are also seen in childhood; Bower et al. (2018) reported the prevalence of 36% of all children within a detention centre in Western Australia, met the diagnosis for FASD. With this trajectory, it is also likely for individuals with FASD to experience further secondary effects, including unemployment (Rangmar et al., 2015) and homelessness (Temple et al., 2020). In order to better understand these potential long term effects, investigations have been made into the early needs of this population. In particular, Tsang et al. (2016) explored the behavioural needs of children with FASD, compared to children without FASD. This systematic review focused specifically on studies where the Achenbach System of Empirically Based Assessments (ASEBA; Achenbach & Rescorla, 2004) were used. These assessments include a range of validated measures, including the Child Behaviour Checklist (CBCL; Achenbach, 1999), the Teacher Rated Form (TRF; Achenbach & Rescorla, 2001) and the Youth Self Report (YSR; Achenbach & Rescorla, 2001). When comparing T-scores, Tsang and colleagues (2016) found that higher scores were reported on the CBCL for internalising and externalising difficulties for children with FASD than for children

without FASD. The most common problems documented for children with FASD were found to be thought problems, behavioural problems such as rule-breaking, aggression, attention difficulties and social problems. This review also found that scores for both the anxiety/depression and withdrawn/depressed subscales were usually within the 'normal' range.

This study is a great introduction to the differences in needs between those with and without FASD. By including only studies utilising ASEBA, there is greater assurance of the quality of included studies. However, there are some drawbacks to this approach; other assessment methods of difficulties were not included which are equally validated measures, such as the Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997), The Schedule for Affective Disorders and Schizophrenia for School-Age Children-Present and Lifetime (K-SADS-PL; Kaufman et al., 1997), or clinical interviews with clinical professionals. Whilst the study only aimed to investigate research utilising ASEBA, it is possible that measures such as these may differ in sensitivity and specificity in regards to identifying psychological needs, thus the results of this meta-analysis may have differed with their inclusion. The authors of this review also report that significant problems in areas such as anxiety and depression were not found, which is incongruent with studies utilising other assessments, leading them to suggest that the ASEBA items may not be sensitive enough to recognise these difficulties.

Further to this research, a systematic review by Weyrauch et al. (2017) explored the prevalence of 15 different comorbid mental health presentations found in children with FASD, within which they included neurodevelopmental disorders such as ADHD. Whilst this study was able to provide evidence for high rates of ADHD (50.2%) as well as ODD (16.3%), Anxiety Disorder (7.8%) and Depression (14.1%), there were an insufficient number of studies to provide pooled estimates for Autism Spectrum Disorder (ASD), Mood Disorder and CD.

Finally, Lange et al. (2018) explored the prevalence of ADHD, ODD, CD and ASD in children diagnosed with FASD. The meta-analysis included studies published up until 2016, and found that 52.9% of children with FASD had ADHD, 12.9% had ODD, 7% had CD and 2.6% had ASD. This study adds further weight to the growing evidence of behavioural difficulties exhibited by children with FASD. However, without exploring the internalising difficulties of this population, the full needs of a child with FASD may be unknown and as previously mentioned, underreported. The authors of this study also recognised that research within the FASD field typically utilises samples which are clinically referred, which may impact the generalisability of results given that children with noticeable externalising behaviours are more likely to be referred to such clinics. They conclude that research is needed in samples which are non-clinically referred, such as population based samples. Interestingly, this study also investigated the method of data collection in each study, reporting what studies used

active methods (new data collected for the purpose of the study) versus passive methods (previously collected data utilised for the purpose of the study). Sub-analysis of data collection found that passive methods yielded slightly higher pooled prevalence estimates for ADHD and ODD when compared to active methods. This suggests the methodology used in the literature base could impact on our understanding about the true prevalence of comorbidities.

In recent years there have been several changes in the way FASD is diagnosed and categorised, with the most recent updates coming from the introduction of clinical standards set by the National Institute for Health and Care Excellence [NICE]), which document what an assessment of FASD should include (NICE, 2022). Moreover, conceptualisation of other disorders, such as ASD, have developed over time, which has led to more awareness and ultimately higher prevalence estimates (Zeidan et al., 2022). Due to this, it is possible that the estimates given in previous meta-analyses may be understated and an updated analysis may be beneficial, to include more recent studies.

Current meta-analysis

Along with the new NICE clinical standards (NICE, 2022), it is hoped that more awareness is given to individuals diagnosed with FASD, and thus it is essential to get an up to date picture of the needs of this population, to ensure that appropriate support can be offered. Therefore, the current study aims to take a broader view of the needs of children with FASD, by exploring the mental health, behavioural and neurodevelopmental needs reported, via a proportional meta-analysis. By considering the limitations of previous research, this meta-analysis aims to include studies where needs are reported rather than just diagnoses, to develop a broader picture of the difficulties experienced. This will ensure that those who have mental health, behavioural and neurodevelopmental needs but have not been able access an assessment, (i.e., those not from clinic-referred samples) can be included. This approach aims to help with the clinical relevance of the results. This meta-analysis also aims to put further emphasis on internalising behaviours such as low mood and anxiety, given the proportion of studies which focus on disorders with externalising behaviours, such as ADHD and CD.

Materials and Methods

There were no similar meta-analyses registered on PROSPERO, therefore, utilising the PRISMA guidelines (Shamseer et al., 2015), a systematic review protocol was developed and registered (January 2023, CRD42023387449)

Search Strategy

Systematic searches were performed in a total of four electronic databases, chosen by those with the largest results during initial scoping searches. These databases were Pubmed, PsycINFO, Medline and CINAHL. The initial scoping searches supported with developing the search terms, which included three elements: (1) the disorder of interest (FASD), (2) the population of interest (children) and (3) the comorbidities of interest (mental health needs, behavioural needs and neurodevelopmental needs). Due to FAS first being described in a journal article in 1973 (Jones & Smith, 1973; Jones et al., 1973), all articles published between January 1973 and February 2023, which were peer reviewed and written in English Language, were considered. The full search terms can be found in Table 1.

Table 1: Search terms used for database searches

Disorder	Population	Comorbidities
"alcohol related neurodevelopmental disorder" OR "ARND" OR "FAE" OR "FAS" OR "FASD" OR "fetal alcohol syndrome" OR "fetal alcohol spectrum disorder" OR "foetal alcohol syndrome" OR "foetal alcohol spectrum disorder" OR "partial fetal alcohol syndrome" OR "partial foetal alcohol syndrome"	“child*” or “adoles*” or “juvenile” or “delinquent” or “teenager*” or “youth*” or “camh*” or “young adult*” or “paed*”	"ADD" OR "ADHD" OR "attention deficit disorder*" OR "attention deficit hyperactivity disorder" OR "autism" OR "autism spectrum disorder" OR "ASD" OR "conduct disorder" OR "externalizing disorder*" OR "internalizing disorder*" OR anxious OR anxiety OR depress* OR "withdraw*" OR "low mood" OR aggress* or behav* OR "mood disorder" OR "mood problem" OR difficul* OR problem*

Note: ARND = alcohol related neurodevelopmental disorder. FAE = fetal alcohol effects. FAS = Fetal Alcohol Syndrome. CAMH = child and adolescent mental health. ADD = Attention deficit Disorder. ADHD = Attention Deficit Hyperactivity Disorder. ASD = Autism Spectrum Disorder.

Eligibility Criteria

In order to be included in the review, each study needed to meet the following criteria: (1) the sample were human participants, with the average age of the sample below the age of 18 years old (2) participants must have had a diagnosis of FASD, including subtypes, (3) the study clearly reported co-morbid mental health, behavioural or neurodevelopmental needs. To support with the study aim of widening the sample beyond clinical populations, these co-morbid needs did not need

an official diagnosis in order to be included. Studies were excluded if (1) there were no clear diagnosis of FASD or subtype, (2) comorbidity data was not reported clearly in the form of n or percentage (e.g. studies just reporting *t*-scores were excluded), (3) the study was a case study, research dissertation/thesis, qualitative study, systematic review/meta-analysis, (4) if the sample was used in another included study (if this was unclear, they were excluded). Following an abstract review, the first reason for any study excluded was recorded. Figure 2 illustrates this process within a PRISMA flowchart.

Risk of bias

A range of Joanna Briggs Institute (JBI) Quality Appraisal tools (Joanna Briggs Institute, 2020) were selected to rate the quality of each included study (see Appendix B). These are common tools used within meta-analyses exploring mental health (for example, Righy et al., 2019), and offer a selection of different tools for each methodology. This was important for the current meta-analysis, due to there being a variety of designs used within the included studies. The JBI tools comprised of 8-13 questions assessing the methodological quality, to explore the potential of bias. Questions assessing inclusion criteria, the descriptions of the samples used, and the validity and reliability of the measures used are included. Each criterion is answered with either 'Yes', 'No', 'Unclear' or 'Not Applicable'. Whilst the tools do not stipulate cut-off scores for overall ratings, the following cut-offs have been utilised by previous research (for example, Valesan et al., 2021): studies in which up to 49% of questions were scored as "yes" were rated as having a high risk of bias, 50-69% as a moderate risk of bias, and over 70% as a low risk of bias. Thus, these cut-off points were used within the current study to inform overall risk of bias. A total of 13 (26%) were inter-rated by the first author and a researcher who was not involved in this present research. If there were discrepancies in the overall rating of individual studies, then both raters discussed the specific questions for these studies which had resulted in different ratings. If no agreement could be met, then the study in question was taken to the primary supervisor for this project for further consideration.

Coding of studies

Based on the current literature, the following variables were considered: depression, anxiety, behavioural conditions, ADHD and ASD. These variable names acted as umbrella terms for a variety of reported conditions, needs or disorders. Under depression, all studies reporting prevalence for depression, low mood, mood disorders, withdrawn behaviour and suicidal ideation were included. For anxiety, all studies reporting prevalence for anxiety, generalised anxiety disorder and panic were included. For behavioural conditions, behavioural disorder, CD, ODD, rule breaking behaviour,

aggressive behaviour, disobedient behaviour, and scores above the borderline range for problem behaviours on the CBCL were included. For ADHD, Attention Deficit Disorder (ADD), Attention Deficits and ADHD were included. Finally, for ASD, Autism, Childhood Autism, Atypical Autism, Asperger's syndrome, social communication disorder and ASD were included. Due to the scope of the study and the need to limit what was investigated, other conditions which fell under these umbrella terms, such as Post Traumatic Stress Disorder (PTSD), Obsessive Compulsive Disorder (OCD), and phobias (falling under anxiety disorders), were not considered.

Data was extracted from all remaining studies by the lead researcher, using a data extraction table which was designed specifically for this project. This included: author, year of publication, location, study design, sample size, methods of assessment, FASD diagnoses and the reported prevalence of comorbidities. Due to the various methodological differences which were identified throughout, several rules were created to standardise the data extraction phase:

- Some studies included samples which spanned childhood and adulthood; if the reported average age was under 18 then they were included within the current study.
- If a study reported prevalence rates for multiple diagnoses which fell under the same umbrella term, only one of these was considered. This occurred on several occasions for behavioural conditions, under which studies reported prevalence rates for CD as well as ODD. For these studies, if it was unclear whether participants could have had both diagnoses, rates of conduct disorder took priority and were included in the data, due to this disorder appearing to have a broader diagnosis criterion. In contrast, if it was clear which participants had which disorder, and thus there were no concerns of participants having both disorders, then the prevalence rates for both were grouped together by summing the number of participants with each disorder together.
- If a study utilised more than one data collection method, then only one was considered. This occurred in one study within which both the TRF and CBCL were used. In this instance, the CBCL data took priority as it was rated by parents, who could be considered to have greater insight into their child's difficulties.
- Due to the limited amount of data available for children with FASD, many studies utilised a retrospective case review method, using clinic assessment data. This resulted in the same sample being used in multiple studies. The authors took great care in identifying these studies, by searching each included study and cross referencing the available data, including the timeframe data was collected, location

of the study and the clinic the data was extracted from. If the same sample had been used, the only the most recent study was included.

Data synthesis

The meta-analyses were performed using the statistical software, STATA 18 (StataCorp, 2023). For each study, the prevalence of each variable (depression, anxiety, behavioural, ADHD and ASD) were taken and pooled together to provide a weighted, overall estimated prevalence.

The included studies varied in numerous domains, including statistically and methodologically, which indicated that there would be high levels of heterogeneity. To support with this, an inverse Freeman-Tukey's Transformation Proportion Random Effects model was utilised, which ensures that confidence intervals stay above zero and is a commonly used approach (Higgins et al., 2023). To assess for heterogeneity, forest plots were created and explored. Two statistical tests were also inspected: the I^2 statistic (Higgins & Thompson, 2002), which explores how much variation in the studies are due to heterogeneity rather than chance, and the Cochran's *Q* test (Cochran, 1954) which tests the significance of heterogeneity within the studies.

Moderator Analysis

Inspired by Lange et al. (2018), information was also collected on the method of data collection in each study, to see if this impacted on the prevalence of each need. Active methods involved the researchers collecting new information about the participant's needs within the study, such as through screening questionnaires (for example the Conners Rating Scales-Revised; Conners, 1999), or through clinical assessment. Passive methods involved the researchers collecting this information from elsewhere, such as through file reviews or reports. This information was collected to see if there were any disparities in the pooled prevalence when comparing current needs (via active data collection) versus historical needs (via passive data collection). A further justification for this comparison was that passive methods such as file reviews were more likely to only present data for children who have presented in clinics and completed measures, whereas active methods could, for example, include studies where self-reported data through nationwide surveys, thus providing a broader scope of clinical need. To explore the effects of active versus passive methods of data collection, moderator analysis was conducted, utilising a random effects model with Freeman Tukey Inverse Transformation applied.

Sensitivity Analysis

To explore whether overall prevalence rates were skewed by individual studies which were rated to have a high risk of bias, sensitivity analysis was completed. These studies were removed from the meta-analysis and then the pooled prevalence was statistically compared to the original dataset.

To explore the potential of publication bias in each study, funnel plots were explored.

Results

The first stage of selection included all studies that were identified by the search strategy. This yielded a total of 6759, which reduced to 2355 once filters were applied. The titles and abstracts of all remaining studies were screened by the lead researcher, based on the aforementioned inclusion criteria. A cautionary approach was taken whereby studies were included if it was unclear from the title or abstract whether they met the criteria, so that they could be read in full. A total of 1954 records were excluded during this process, with a further 159 duplicate studies being excluded following this. This resulted in 242 records to be considered in full. Again, these remaining studies were screened in line with the inclusion criteria, which resulted in 192 being excluded. This left 50 studies eligible for this current meta-analysis. This process is detailed in a PRISMA flowchart (Moher, Liberati, Tetzlaff & Altman, 2009) found in Figure 2.

Across the studies, sample sizes ranged from 4 to 9382, with a total sample size of 39071. A number of studies reported prevalence for more than one clinical need (for example, depression as well as anxiety), and also utilised different collection methods (for example, use of a screening assessment as well as file review) thus the 50 studies yielded a total of 125 prevalence rates.

Study Characteristics

Characteristics for each study can be found in Table 2, with further details found in Appendix C.

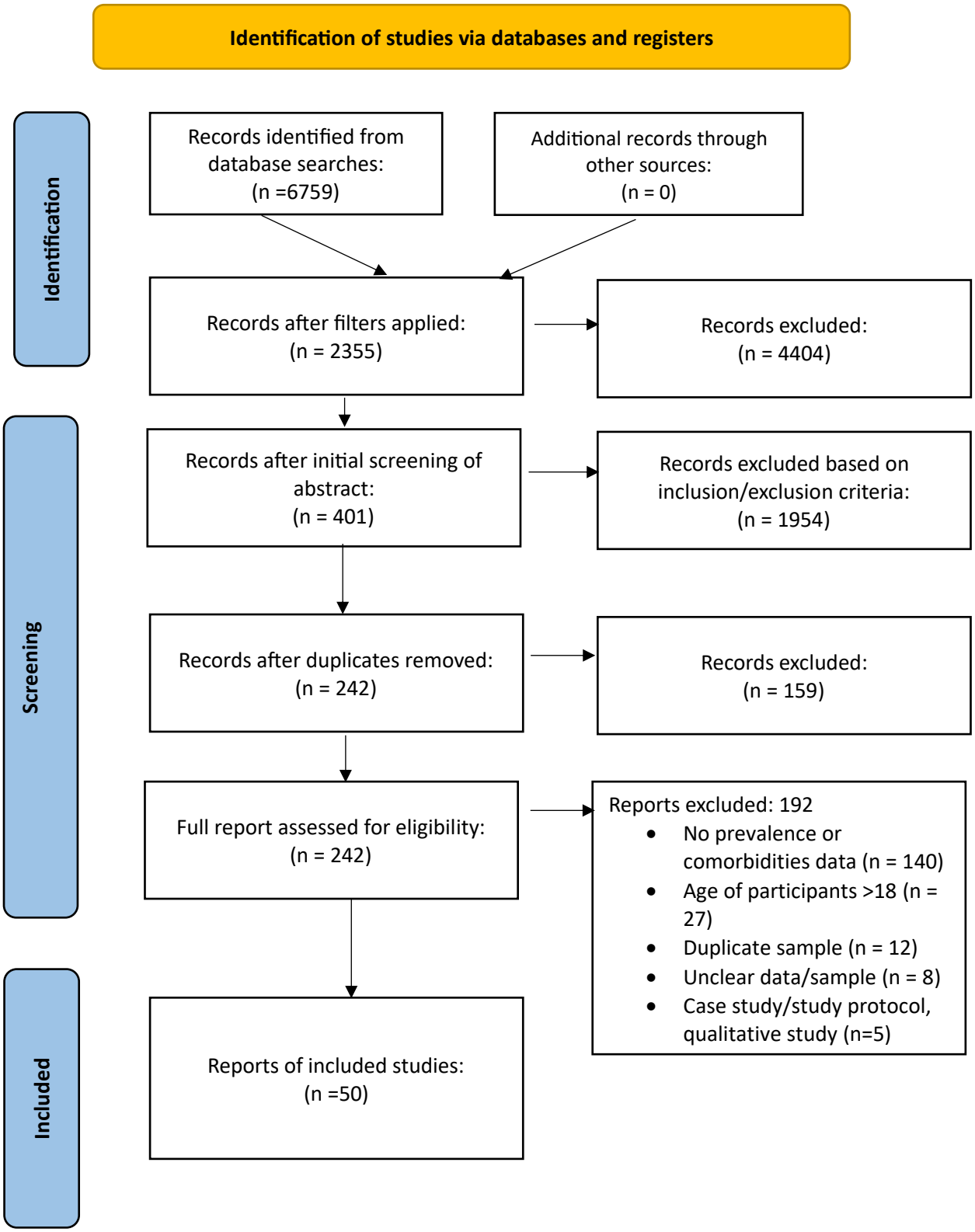

Figure 2: PRISMA flowchart

Table 2: Included studies and extracted data included within meta-analysis

Study	Sample size	Psychological need									
		Depression		Anxiety		Behavioural		ADHD		ASD	
		n	*%*	*n*	*%*	*n*	*%*	*n*	*%*	*n*	*%*
Agnihotri et al. (2019)	41	1	2.4	3	7.3	3	7.3	26	63.4		
Banerji et al. (2017)	39					39	79.6	21	42.9		
Breiner et al. (2013)	17					15	88.2				
Burd et al. (2003)	303	29	9.6			53	17.5	119	39.3		
Burns et al. (2021)	665	86	12.9	87	13.1	53	8			27	4.1
Chasnoff et al. (2015)	156	12	7.7	15	9.6	4	2.6	88	56.4	8	5.1
Chen et al. (2012)	33							25	75.8		
Connor et al. (2020)	199	11	5.5	50	25.1	13	6.5	83	41.7		
Elgen et al. (2007)	47							42	89.4		
Fagerlund et al. (2011)	73	8	11	2	2.7			44	60.3		
Flannigan et al. (2019)	38	0	0	0	0	21	55.3	8	21.1		
Franklin et al. (2008)	44	15	34.1			8	18.2	23	52.3		
Geier & Geier. (2022)	321							166	51.7	22	6.9
Green et al. (2009)	89	10	11.2	15	16.9	19	21.4	53	59.6		
Green et al. (2014)	52					19	36.5	31	59.6		
Greenbaum et al. (2002)	28							9	32.1		
Greenbaum et al. (2009)	33	1	3	2	6.1	1	3	20	60.6		
Hayes et al. (2020)	163	20	12.3	71	43.6	51	31.3	107	65.6	37	22.7
Herman et al. (2008)	36							18	50		
Ipsiroglu et al. (2019)	40	17	42.5	23	57.5	8	20	27	67.5	2	5

Study	Sample size	Psychological need									
		Depression		Anxiety		Behavioural		ADHD		ASD	
		n	*%*	*n*	*%*	*n*	*%*	*n*	*%*	*n*	*%*
Kambeitz et al. (2019)	98	22	22.5	40	40.8			84	85.7	4	4.1
Kooistra et al. (2010)	28							27	96.4		
Landgren et al. (2010)	37					15	40.5	21	56.8	2	5.4
Lane et al. (2014) (Active)	14							10	71.4		
Lane et al. (2014) (Passive)	14							9	64.3		
Lange et al. (2019)	21							5	23.8	3	14.3
Lidstone et al. (2020)	17							15	88.2		
Malisza et al. (2012)	23	1	4.4			2	8.7	11	47.8		
Mattson et al. (2013)	79							19	24.1		
Montag et al. (2022)	15							1	6.7	10	66.7
Mughal et al. (2020)	91			67	73.6						
Mukherjee et al. (2019)	97							72	74.2	62	68.1
O'Conner et al. (2019)	54	20	37								
Oesterheld et al. (1998)	4			1	25	3	75	4	100		
Okulicz-kozaryn et al. (2017)	50							19	38		
Palmeter et al. (2021)	9382	3087	32.9	4156	44.3	6530	69.6				
Paolozza et al. (2013)	27	3	11.1	3	11.1	4	14.9	11	40.7		
Paolozza et al. (2014)	72	4	5.6	9	12.5	10	13.9	45	62.5		
Rai et al. (2017)	52	1	1.9	1	1.9	2	3.9	12	23.1		
Rasmussen et al. (2010)	52							33	63.5		

Study	Sample size	Psychological need									
		Depression		Anxiety		Behavioural		ADHD		ASD	
		n	*%*	*n*	*%*	*n*	*%*	*n*	*%*	*n*	*%*
Rasmussen et al. (2013)	32	2/32	6.3	3/32	9.4	2/32	6.3	18	56.3		
Reid et al. (2017)	31							19	61.3	4	12.9
Stromland et al. (2015)	16					1	6.3	6	37.5		
Tsang et al. (2017)	18	7	38.9	4	22.2	9	50.0	4	22.2		
Uecker et al. (1996)	15							2	13.3		
Webster et al. (2020)	15							11	73.3	1	6.7
Wells et al. (2012)	78	14	18	5	6.4	6	7.7	55	70.5		
Williams et al. (2014)	31							27	87.1		
Yu et al. (2022)	26							11	42.3		
Zhou et al. (2011)	20							10	50.0		

ADHD = Attention Deficit Hyperactivity Disorder. ASD = Autism Spectrum Disorder

Assessment of Risk of Bias

The overall proportion of studies rated as at low, moderate or high risk of bias through the JBI tools, can be seen in Figure 3. To ensure reliability, 13 (26%) studies were inter-rated. Initial inter-rater agreement was 76.92%, Cohen's Kappa coefficient (k = 0.53), indicating moderate agreement (Landis & Koch, 1977).. For the three studies that were not rated the same by both raters, a discussion was held whereby the individual appraisal questions which were rated differently were explored. Following this, consensus was met, which adjusted the final overall agreement to 100% (k = 1). This high level of consistency across the subset of studies indicates that the ratings were reliable. A total of 32 (64%) of studies were rated as low risk of bias, 12 (24%) as moderate, and 6 (12%) as high.

Prevalence

Overall, the included studies resulted in a pooled prevalence of 13.3% (95% *CI* 8.7 – 18.6%) for depression, 18.9% (95% *CI* 10.8 – 28.5%) for anxiety, 22.8% (95% *CI* 13.2 – 34.1%) for behavioural difficulties, 56.5% (95% *CI* 49.9 – 63.1%) for ADHD, and 14.8% (95% *CI* 5 – 29.9%) for ASD. However, as predicted, all of these results were found to be significantly heterogenic. Additional statistical information can be found in Table 3.

Table 3: Meta-analysis outcomes

Psychological Need	Number of studies	N	Pooled prevalence estimate	95% Confidence interval		I^2	*Q*	*p*
				Lower	Upper			
Depression	22	11680	13.3%	8.7%	18.6%	95.29	622.2	<0.001
Anxiety	20	11351	18.9%	10.8%	28.5%	97.79	856.73	<0.001
Behavioural	24	2248	22.8%	13.2%	34.1%	96.85	424.2	<0.001
ADHD	47	12139	56.5%	49.9%	63.1%	95.15	604.14	<0.001
ASD	12	1653	14.8%	5.4%	27.4%	96.81	258.67	<0.001

ADHD = Attention Deficit Hyperactivity Disorder. ASD = Autism Spectrum Disorder

Moderator Analysis

Data collection

The method of data collection was explored through sub-group analysis. For this, any studies which used mixed methods (active and passive), or the methods were unclear, were removed from analysis due to there being an insufficient number of studies for a meaningful comparison. Moreover, due to only one study using active methods for ASD, this psychological need was removed from the subgroup analysis. Overall, a test of group differences found that prevalence rates from active measures of data collection were not statistically higher than passive methods (depression: $\chi^2(1) = 2.93, p = 0.09$; behavioural: $\chi^2(1) = 0.51$, p=0.47; ADHD: $\chi^2(1) = 0.03$, p = 0.87; anxiety: $\chi^2(1) = 0.02$, p = 0.89). Table 4 presents these results.

Table 4: Sub-group analysis for method of data collection

Psychological Need	Data collection method	Number of studies	N	Pooled prevalence estimate	95% Confidence interval		I^2	*Q*	*P*-value
					Lower	Upper			
Depression									
	Active	4	223	23.8%	11.3%	38.9%	81.41	15.48	0.001
	Passive	16	11249	12.2%	7.2%	18.2%	95.75	519.81	<.001
Anxiety									
	Active	6	316	16.4%	3%	43.8%	95.63	170.85	<0.001
	Passive	13	10879	20.1%	12%	30.2%	97.67	542.51	<0.001
Behavioural									
	Active	6	185	31%	3.4%	67%	94.49	72.25	<0.001
	Passive	16	1974	21%	10%	34%	97.31	334.36	<0.001
ADHD									
	Active	16	584	58.8%	42.0%	74.6%	93.56	204.01	<0.001
	Passive	27	11270	57.1%	50.3%	63.8%	93.28	336.47	<0.001

ADHD = Attention Deficit Hyperactivity Disorder. ASD = Autism Spectrum Disorder

Location of study

Following extraction of the data, it became apparent that there were more studies which took place in Canada and the USA, compared to other places worldwide. Due to this, exploratory analysis took place on the location of study to investigate whether this factor impacted on the prevalence of psychological needs. Sub-group analyses did not find any significant differences in the reported prevalence rates for any of the psychological needs between Canada, USA and studies taking place in the rest of the world (ROW). Further details of this analysis can be found in Chapter 6.

Sensitivity Analysis

Studies rated as high risk of bias were removed to conduct sensitivity analysis. Chi Square analysis found no statistically significant differences when comparing the full data set and when high risk of bias studies were removed (depression: $\chi^2(1) = 0.36$, $p = 0.55$; anxiety: $\chi^2(1) = 0.24$, $p = 0.63$; behavioural: $\chi^2(1) = 0.20$, p=0.66; ADHD: $\chi^2(1) = 0.01$, $p = 0.93$; ASD: $\chi^2(1) = 0.02$, $p = 0.88$).

Publication Bias

Funnel plots of the prevalence data were explored to assess for publication bias (see Appendix D). Because prevalence rates cannot go below zero, publication bias can be hard to assess within meta-analyses of proportion. Asymmetry was present in all funnel plots across the psychological needs, indicating that small study effects may be present and thus the results should be interpreted with caution.

Discussion

The studies included in this meta-analysis summarised the neurodevelopmental, behavioural and mental health needs of children with FASD. For depression, prevalence (13.3%) was found to be similar to previous research (14.1%, Weyrauch et al., 2017). This was also the case for ADHD (56.6%), reported to be between 50.2% - 52.9% previously (Weyrauch et al., 2017; Lange et al., 2018). The prevalence of behavioural problems (22.8%) was higher than previously reported when compared to rates of ODD (16.3%, Weyrauch et al., 2017; 12.9%, Lange et al., 2018) and CD (7%, Lange et al., 2018). This was similar for ASD (14.8%), which was reported to have a prevalence of 2.6% previously (Lange et al., 2018). The prevalence of anxiety (18.9%) was higher than previously reported (7.8%, Weyrauch et al., 2017). The included papers varied in terms of quality; 64% were rated as low risk of bias, 24% as moderate, and 12% as high.

Overall, there was a high level of heterogeneity. One factor which may have exacerbated this was the method of data collection. The majority of studies opted for a passive method of data collection, often through medical note reviews. Some studies were able to utilise and report results from previous standardised questionnaires, such as SDQ or CBCL. However, for many studies the original method of data collection was unknown, often with only the prevalence stated. Passive data collection methods result in research being cheaper and quicker to run, due to not having to invest time and money into collecting new data. However, concerns could be raised over the validity of secondary data, with many studies not reporting how comorbidity data was originally collected, or when it was collected. Whilst the secondary difficulties of FASD can be life long, it is also recognised that they can change over time. Interestingly, for all the psychological needs explored within this study except anxiety, pooled prevalence estimates were higher when active data collection methods were used, although these differences were not found to be significant. Higher prevalence rates from active methods may be reflective of the changing needs of individuals with FASD, which may be more intense as individuals face more challenges as they grow older. This indicates that support plans for children with FASD should be dynamic and adaptive to address the impact of maturity. Services should also be equipped to support with changing needs, offering a streamlined pathway to support

those with an FASD diagnosis as and when they arise. This would serve as a protective factor against the risk of secondary disabilities.

From the 50 studies included, 23 were based in Canada and 13 were based in the USA, with a small number of studies taking place elsewhere (Australia = 4, UK = 2, international multi centre sites = 2, and one study each from Brazil, Poland, Sweden, Finland, South Africa and Norway). Due to this, location of study was also explored, although no significant results were found between Canada, USA and studies from the rest of the world (ROW). Conclusions from this are limited, however, due to a lack of studies included from other parts of the world to compare with those from Canada and USA, which resulted in the category of ROW. Thus, pooled prevalence rates may differ within the countries of the ROW category, and ultimately more research regarding prevalence is needed.

Despite no statistically significant results, it is clear that whilst FASD is being investigated more overall, there is greater investment in certain parts of the world. Also reflective of this, is the origination of some of the guidelines adopted, for example the Canadian FASD guidelines (2005). Thus, the current study may not be reflective of how FASD presents globally, and may be biased towards a western ethnocentric population. This is concerning, given the prevalence of FASD amongst low socio economic populations, which are found in western countries, but may be more prevalent in other parts of the world. South Africa, for example, is documented as having the highest prevalence of FASD with a rate of 222.2 per 1000 population (Lange et al., 2017). The reasons for this are proposed to be due to the complex cultural, social and political history the nation has with alcohol consumption, as well as a lack of government intervention to announce FASD as a major public health concern (Olivier et al., 2016). Despite this, within this meta-analysis, only three studies included samples from South African populations. This is not to say that research is not taking place; for example, a series of studies investigating the epidemiology of FASD within South African communities were completed by May and colleagues (for example May et al., 2007). However, due to these studies focusing on developmental tests and only providing *T*-scores for behavioural difficulties, rather than prevalence, they were excluded from analysis within the current meta-analysis. This indicates that whilst we know that the prevalence of PAE is high in some areas, we know less about how the prevalence of psychological needs of these children present. This means that many children with FASD, may have needs which are unmet, contributing to the risk of secondary disabilities (Streissguth et al., 2004).

Another factor which was not explored, but may have contributed towards heterogeneity, was the assessment methods used for the diagnosis of FASD. Across the studies, more than five different assessment and classification methods were utilised by the studies, including the 4-Digit Code (Astley, 2004), Hoyme et al. (2016), and the Canadian FASD Guidelines (Chudley, 2005). Lange

et al. (2018) notes that the numerous clinical guidelines for assessing FASD can make the distinction between FASD and other neurodevelopmental conditions difficult to understand, often leading to an overstated relationship between the two. The result of this means that many children may be treated for a disorder other than FASD, such as ADHD, which may not meet the needs of the child. Further to this, a child may be given multiple diagnoses, which may bring its own difficulties. For example, in the landscape of Integrated Education (IE), whereby education in the classroom is tailored to each child's needs, multiple diagnoses lands further demand on educators to understand how these needs interplay and how to effectively teach the curriculum (Warnes et al., 2021). This is concerning, given that there is already ambivalence and inconsistent practice among teachers in relation to IE and complex needs (Parey et al., 2019). This runs the risk that children will have their needs unmet in schools, which may add to their feelings of frustration and isolation through punitive treatment (Office for National Statistics, 2022). This highlights the pressing need for a universal method of assessment to be adopted globally, so that a diagnosis of FASD can be made with clarity without the complexity of multiple diagnoses or indeed, misdiagnosis.

Strengths

The current study has a number of strengths which are worth noting. Firstly, Lange et al. (2018) documented that majority of the research utilises clinically referred samples, indicating that sample bias may be present. The current study sought to overcome this issue by including studies by which self-reporting of needs were included, thus less likely to be clinic samples. This meant that children who are less likely to present at clinics for psychological needs also had the chance to be represented within the current study. This enabled the current study to take a broader look at the needs of children with FASD, rather than relying on only those with psychiatric disorders. In line with this aim, the decision was used to create categories of psychological needs, rather than singular diagnoses, for example, looking at behavioural needs rather than the prevalence of ODD or CD independently. This ensured that any studies utilising measures which indicates areas of difficulties, for example the CBCL, were included. Whilst this was the aim, it is recognised that the majority of the studies (72%) included were still samples which derived from clinic or hospital settings, with official diagnoses. This reflects the research that is currently undertaken within FASD populations and highlights that there is still a need for more research within non-clinically referred samples.

Further to this, a number of studies utilised the same sample of children with FASD. The current study took great care in excluding duplicate samples in order to get a representative prevalence estimate.

Due to the increase of research into FASD and neurodevelopmental conditions overall, the current study was able to provide pooled prevalence estimates for ASD, which were not able to be provided previously.

Limitations

Due to the nature of the current study, the data that needed extracting from studies was typically found in the participant information/demographic section of the paper, rather than in the results section. Possibly because of this, there was a consistent lack of detail about how the data was collected. Many papers stated the documented comorbidities of the sample without explaining where the information had come from. Some studies reported the demographics of the full sample, but then only reported data for a subsection due to missing data, resulting in difficulties in understanding the demographics of just the subsection. Rejected studies also included papers which documented that comorbidities were present, then failed to fully document these. Finally, a large number of studies were rejected due to the prevalence of needs not being documented, despite using measures to explore these. These studies typically presented *T*-scores, comparing the needs of FASD children to typically developing children. Whilst this is supportive of the aims of their research, if the prevalence of children falling with high T-scores were reported, pooled prevalence studies would be benefited greatly.

Another limitation of this piece of research is the small numbers of studies for some of the psychological needs. For example, only 12 studies were included for ASD. This could have resulted in the prevalence of ASD not being representative for an FASD population. In addition to this, as the majority of studies utilised passive methods of data collection, there was limited data to compare active and passive methods of data. For example, for ASD, 11 out of 12 studies used passive methods. This means that the analysis may have been underpowered and therefore caution must be taken when interpreting the results, as small effects may not have been identified.

In order to get an overall view of psychological needs without a reliance on diagnoses, the needs were grouped together into categories under umbrella terms. This required a level of subjectivity from the authors, to decide what data fit into each category, and did result in decisions where some data took prevalence over others. It could be argued that the categories may misrepresent some of the needs they encompass. Thus the validity of the categories needs to be taken into consideration when drawing conclusions. Despite these drawbacks, this approach is still felt to have advantages, as noted above.

Finally, whilst great care was taken to identify duplicate samples across the studies, the authors cannot guarantee that all were identified and removed. This particularly applies to studies

which used national databases recording data from various clinics which were included; attempts were made to ascertain whether a sample had been used before via contacting database administration teams, however due to confidentiality, the clinics the data were collected from could not be named. The authors took the decision to include such studies due to the large sample size, and complete sensitivity analysis to explore the impact of these studies when included.

Clinical implications

The pooled prevalence estimates for each psychological need were found to be similar to previous meta-analyses completed, or even higher for some of the needs reported. This could be reflective of the inclusion criteria of the current meta-analysis, with any need falling under the umbrella terms being counted. Alternatively, this could also be reflective of how the knowledge and awareness of FASD and co-occurring conditions has developed since previous meta-analyses have been published. Moreover, the current meta-analysis indicates that much of the research is still utilising clinic samples, however, research utilising active case ascertainment has indicated that the prevalence of FASD is higher than reported, and is often undiagnosed (McCarthy et al., 2021). This suggests that co-occurring conditions could also be underrepresented by the use of clinic samples. Further research, possibly utilising school samples to screen wider populations, would be beneficial to understand the prevalence of co-occurring conditions further. Regardless of the reasons for this, the current meta-analysis indicates that the needs of a child with FASD can cover many domains. Much focus has been given to externalising disorders of children with FASD, most likely due to the economic impact of these, however, the current study indicates that when mood is actively explored, 22.8% may report difficulties. Greater emphasis needs to be given as to how FASD can impact on these domains, particularly as low mood has been found to be strongly associated with behavioural problems (Maasalo et al., 2016). Supporting children with FASD with the issues which may contribute to low mood, such as social interaction and sleep, may support with the behavioural issues which are seen (Hayes et al., 2020). Further to this, adopting a more formulaic approach towards documenting the needs of children with FASD, by focusing on needs rather than diagnoses, may have its own advantages. Firstly, individuals who fall below thresholds for disorders can still have symptoms which are clinically impairing (Lewinsohn et al., 2004), thus focusing on needs may result in more children being identified as requiring support. Secondly, formulaic approaches typically incorporate strengths; focusing on strengths may support with the stigma that is attached to an FASD diagnosis. Despite this, it is recognised that services are currently not operationalised in a way which favours formulaic approaches, most requiring a diagnosis to allow access.

As many studies into the FASD have recognised, the requirement for a universal FASD diagnostic approach is clear. One clear system adopted by all, would increase the clarity between FASD and other neurodevelopmental conditions, supporting with earlier diagnosis and intervention.

Future Research

Whilst the results between active and passive methods were not found to be significant, possibly due to sample size, it is still important to consider the impact of using retrospective data when documenting the co-occurring needs of children with FASD, due to the disorder being lifelong and thus the challenges children face changing over time. Therefore, future research should focus on collecting new data to ensure that the prevalence rates of co-occurring psychological needs are accurately captured.

An area which was not considered due to being beyond the scope of this meta-analysis, is the interaction between ethnicity and the co-occurring needs of children with FASD. Research suggests that African American males, who present with neurodevelopmental needs, are often misdiagnosed with externalising disorders such as CD, resulting in a delay in effective intervention and longitudinally, poorer outcomes (Baglivio et al., 2016). Given that 18% of individuals living in England and Wales are from Black, Asian and other ethnic minority groups (Office for National Statistics, 2022), exploring the available data on co-occurring needs and the moderating effects of ethnicity, would help to highlight whether this issue is present within FASD populations.

Conclusion

The current meta-analysis provided prevalence rates for a range of psychological needs, including depression, anxiety, behavioural difficulties, ASD and ADHD, co-occurring alongside FASD in children. The prevalence of ADHD was particularly high, corroborating with previous findings. Many of the studies included took place within Canada and the USA, with no significant differences found between location of study. Moreover, no significant differences were found between active and passive data collection methods, although conclusions drawn from both moderator analyses should be carefully considered due to small sample sizes.

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Chapter 3: Bridging chapter

Summary of Meta-analysis

Chapter 2 presented a meta-analysis illustrating the high prevalence of neurodevelopmental, mental health and behavioural difficulties experienced by children with Fetal Alcohol Spectrum Disorder (FASD). In particular, the prevalence of Attention Deficit Hyperactivity Disorder (ADHD) was reported to be 56.6%, which is corroborative with previous research and is the most reported co-occurring disorder, with the most extensive research base. Further to this, research has also noted the high rate of missed or misdiagnosis of FASD. Children presenting in assessment clinics have typically previously received a range of diagnoses including ADHD and behavioural issues (Chasnoff et al., 2015). This does highlight the issue of whether the co-morbidities discussed do exist alongside FASD, or whether they are part of FASD. Regardless of this, there remains an issue rooted in the detection, assessment, and diagnosis of FASD.

Some of the reasons for this have been discussed in previous chapters. These include the lack of routine antenatal assessment of alcohol intake, the stigma attached to the disorder which prevents families from seeking assessment, and the heterogeneity in assessments.

Given the high societal cost of FASD, more attention has recently been given to this area, which led to the development of the National Institute for Health and Care Excellence (NICE) quality standards (NICE, 2022), shaped by the Scottish intercollegiate guidelines Network (SIGN) 156 (SIGN, 2019). These quality standards are detailed below (NICE, 2022):

1) Advice on avoiding alcohol in pregnancy: this statement documents that midwives and other healthcare professionals should give “clear and consistent advice on avoiding alcohol throughout pregnancy” (p. 6), stating that there is “no known safe level of alcohol consumption during pregnancy” (p. 6). This statement highlights the discrepancies often reported by healthcare professionals and attempts to standardise the advice given. The guidance also states that pregnant women who are worried about already having drunk alcohol are reassured and offered further help. This part of the statement reflects the stigma attached to the disorder and attempts to support women instead of inflict shame.

2) Fetal Alcohol exposure: this statement involves the routine discussion around alcohol intake throughout pregnancy, in a “sensitive, non-judgemental way” (p. 10). Asking about the frequency and pattern of drinking can also support with early diagnosis and intervention for children with FASD.

3) Referral for assessment: this statement reports that all “children and young people with probable prenatal alcohol exposure and significant physical, developmental or

behavioural difficulties are referred for an assessment" (p. 14). This highlights that prenatal alcohol exposure is "often not considered as a probable cause" (p. 14), particularly if the child does not present with the sentinel facial features. This is despite the prevalence of sentinel facial features only being present for 10% of those with FASD. Thus, the statements aim to increase assess to a referral pathway for an assessment for FASD.

4) Neurodevelopmental assessment: "Children and young people with confirmed prenatal alcohol exposure or all 3 facial features associated with prenatal alcohol exposure have a neurodevelopmental assessment if there are clinical concerns" (p. 19). This should involve an assessment by a multidisciplinary team exploring the following areas: motor skills, neuroanatomy/neurophysiology, cognition, language, academic achievement, memory, attention, executive function (including impulse control and hyperactivity), affect regulation, and adaptive behaviour, social skills or social communication. The statement also mentions that a diagnosis of FASD requires ruling out genetic factors. Statement 4 attempts to standardise the practice of assessing and diagnosing FASD, given that the evidence base suggests that this can be varied.

5) Management plan: "Children and young people with a diagnosis of fetal alcohol spectrum disorder (FASD) have a management plan to address their needs" (p. 23). This statement attempts to ensure that children and their families are supported in the short term and long term with a "staged management plan" (p. 23), tailored to the individual based on their assessment.

These guidelines appear to suggest an assessment and diagnosis process that appear to be helpful in supporting many children who would usually go undetected. However, the evidence base reports that whilst many clinical guidelines that are issued, many fail to make any behavioural change in clinical practice due to factors such as lack of familiarity, lack of awareness, and lack of agreement with the guidelines (Cabana et al., 1999). Thus, there is the possibility that despite clear guidance, there may still be high levels of missed and/or misdiagnosis of FASD. Moreover, the steps towards an assessment are dependent on the information gathered regarding alcohol intake. This bodes the question as to whether alcohol intake would still be considered later in the pathway, if not mentioned previously, particularly if the sentinel facial features are not present.

Overview of Empirical Paper

The next chapter reports an empirical project which attempts to explore this question: given the new quality standards, can healthcare professionals keep FASD as a possible diagnosis in mind

amongst other conditions, and is this dependent on the referral received including information about prenatal alcohol exposure? The study also explores the confidence and attitudes towards FASD, in order to see whether these impact on the detection of the disorder.

Chapter 4: Empirical Paper

Do clinicians consider FASD? Exploring decisions made when assessing and diagnosing in neurodevelopmental child services.

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Emma Heathcote

Abstract

Background

Previous research suggests that missed and misdiagnosis of Fetal Alcohol Spectrum Disorder (FASD) is high due to factors such as stigma and confidence in diagnosing. Following the introduction of specific FASD Clinical Standards by The National Institute for health and Care Excellence (NICE), this study aimed to explore the barriers and facilitators which are associated with clinicians considering FASD as a diagnosis.

Methods

Participants included 139 clinical professionals who were randomised into one of two conditions. Both conditions received a vignette of a referral letter documenting the neurodevelopmental concerns of a 12 year old girl. Within condition A, the referral contained information about prenatal alcohol exposure (PAE),whereas this was omitted for group B. Participants recorded their initial thoughts about potential diagnoses and the assessments they thought were relevant. Following this, they were presented with information regarding the FASD NICE clinical standards, and then completed a survey to document their confidence, attitudes, and opinions in relation to FASD diagnosis

Results

ADHD was the most popular potential diagnosis within both experimental conditions, although the inclusion of PAE information significantly increased the amount of FASD considerations. Most participants in both conditions considered elements of a neurodevelopmental assessment, but only one participant documented elements more specific to FASD. The majority of participants reported to feel confident in identifying FASD, but less so in selecting appropriate assessment tools. Further to this, the majority felt that they had not received enough training about FASD.

Conclusions

Most clinicians do not consider FASD as a potential diagnosis when reviewing referrals with neurodevelopmental concerns. Information of PAE may support clinicians to hold FASD in mind, highlighting the importance of collecting and reporting this information. Finally, further training, in particular regarding assessment, may be beneficial for all clinicians who may come into contact with pregnant women and neurodiverse children.

Keywords: FASD, diagnosis, assessment, vignette

Introduction

Fetal Alcohol Spectrum Disorders (FASD) relates to a spectrum of disorders, caused by the teratogenic effects from consuming alcohol during pregnancy. The developing fetus can experience a diverse range of adverse health effects, including cognitive, behavioural and emotional deficits (Wozniak et al., 2019). In addition to this, Popova et al. (2016) discovered 428 comorbid conditions which can occur alongside FASD, due to its potential impact on any organ or system in the body. These include cardiac and respiratory problems, as well as other neurodevelopmental (ND) disorders such as Autism Spectrum Disorder (ASD) and Attention Deficit Hyperactivity Disorder (ADHD) (Popova et al., 2016). Secondary conditions are also common, with those effected often experiencing mental health problems, school disruptions, substance use problems and trouble with the law (Streissguth et al., 1996).

Prevalence

Despite the wide ranging and severity of the effects of FASD, the prevalence is still relatively unknown. This is due to differences in methodologies and incomplete data sets, meaning that only conservative estimates can be supplied (May et al., 2009). These estimates have been more accessible recently, due to more representative data of FASD in specific settings and prenatal alcohol exposure (PAE), which suggests a prevalence of 7.7 per 1000 children globally (Lange et al., 2017). Furthermore, May et al. (2018) estimated that 1.1% to 5% of first graders in four communities in the United States (US) were affected by FASD. However, with specific settings, comes a lack of generalisability, which the authors acknowledge. The first United Kingdom (UK) prevalence study of FASD utilised an active case ascertainment methodology, by assessing children in primary schools in Manchester. This study found that 1.8% of 220 children met the criteria for FASD, which increased to 3.6% when including possible cases (McCarthy et al., 2021). This indicates that FASD is likely to be more common than ASD, thus it Is understandable that Aiton (2021, p. 1) would conclude that “The condition is not rare, just rarely diagnosed”.

Diagnosis

When completing an anonymous online questionnaire, 28.5% of women in the UK reported drinking alcohol during pregnancy (Mårdby et al.,2017). In addition to this, global estimates suggests that 1 in 67 women who drink alcohol will give birth to a baby with FASD (Popova et al., 2017). Despite these statistics, many families struggle to access an assessment and obtain a diagnosis. In fact, it is estimated that of all the babies born with FASD each year, less than 1% will be diagnosed (Burd & Popova, 2019). Extensive research has explored the reasons for this, which has highlighted

various system-level barriers (Petrenko et al., 2014; May et al., 2009) and the stigma associated with a diagnosis of FASD (May et al., 2009).

System-level barriers

May et al. (2009) also report that one of the difficulties with accessing an FASD assessment and diagnosis lies within the varying knowledge base of clinical professionals involved. Further to this, a systematic review by McCormack et al. (2022) found that most participants within the studies, felt that the health professionals involved with the assessment of FASD, were not sufficiently aware of the disorder. Interestingly, this review also found that specific knowledge of FASD and associated standardised tools and guidelines was poor. Confidence identifying FASD varies across different health professionals, which is likely to be reflective of the volume of training received, ranging from none to at least some (McCormack et al., 2022). From these results, it is concerning, yet not surprising, that missed diagnosis rates can be as high as 80.1% (Chasnoff et al., 2015), with children often being diagnosed with other ND conditions such as ADHD (Wozniak et al., 2019).

Research has suggested that reasons for a lack of knowledge in associated standardised tools may be due to the lack of distinctive criteria within diagnostic classification manuals. Instead, a selection of 11 different guidelines have been developed and adopted by various countries, resulting in a lack of standardisation (National Institute on Alcohol Abuse and Alcoholism, 2019). This lack of standardisation is felt to be the most crucial factor in limiting understanding of prevalence and symptomology (Popova et el., 2020). One particular problem which comes with this, is the reoccurring practice of PAE being overlooked in children presenting with developmental problems, particularly if they do not present with the sentinel facial features (NICE, 2022), which less than 10% of those living with FASD have (Department of Health and Social Care, 2021). This in turn can be related back to the issue of adequate training received by all disciplines, which are stated to be important for the assessment and diagnosis of FASD within guidance documents.

Stigma

Research has indicated that families, as well as professionals in various settings, express concerns about the stigmatizing effect of receiving a FASD diagnosis and the misperceptions of FASD (McCormack et al, 2022). When compared to mothers with serious mental illness, jail experience and substance use disorder, Corrigan et al. (2017) found that birth mothers of children with FASD are viewed as more different to others, valued less than others, and more to blame for their child's disorder, ultimately putting them more at risk of discrimination. Thus, the self-disclosure of prenatal

alcohol consumption is often a barrier to accessing an assessment for FASD, which is likely to be dependent on the prior determination of PAE (Popova et al., 2020).

Towards Standardisation within the United Kingdom

FASD places significant financial burden on society; a report from the British Medical Association (BMA) reported that the estimated annual cost overall for FASD in the UK is over £2 billion (BMA, 2016). Moreover, the mean annual cost for a child with FASD, including special education and effects of secondary difficulties such as criminal justice involvement, is estimated to be $22,180 (Greenmyer et al., 2018), which is approximately £16,615[2]. One of the most consistent protective factors against secondary difficulties is an early diagnosis (Streissguth et al., 2004). Thus, it is within the societal and individual interest to diagnose and support those affected early. In knowledge of this, the UK government and National Institute of Health and Care Excellence (NICE) quality standards were introduced in March 2022 (NICE, 2022), shaped by the Scottish Intercollegiate Guidelines Network (SIGN) 156 (SIGN, 2019), to acknowledge and support these difficulties with diagnosis. These guidelines indicate the need for referral, assessment, and support for any child with probable/definite PAE, facial features and deficits in at least three or more ND areas. The quality standards state that an assessment should include the ruling out of other genetic disorders, a full family, social and medical history, physical examination, and a full ND assessment. The full guidelines can be found here: www.sign.ac.uk/media/1092/sign156.pdf (SIGN, 2019).

The quality standards are a positive step towards standardising a pathway for an assessment of FASD. However, it does not address the high missed diagnosis rate noted in the literature, as well as the associated stigma of disclosing PAE. Successful diagnosis of FASD is likely to be problematic if there are barriers associated with a clinician feeling competent and confident in recognising FASD in ND services. Howlett et al. (2019) took a step towards understanding these barriers within the UK, by investigating the perceived knowledge, confidence and attitudes towards FASD and alcohol use in pregnancy in UK health care professionals. This research indicated a need for further training and highlighted the associated stigma of FASD. This study, however, did not include psychologists or Speech and Language Therapists (SLTs) who can be crucial in the identification and treatment of FASD (NICE, 2022). Whilst this study gave an interesting insight into the attitude towards FASD diagnosis in the UK, it could be argued that it is limited due to the sample being self-selected; participants who opted to take part were aware that it was about FASD and thus may have had prior experience or interest in the area, resulting in possible sample bias. This means that it may not be reflective of how all clinicians will respond to the new NICE quality standards.

[2] Calculated using the average exchange rate in 2018 (£1 = $1.3349; exchangerates.org.uk)

Rationale for current study

As the quality standards hope to increase awareness for FASD and support with increasing access to assessment services, professionals are likely to be faced with an increase in referrals of individuals with potential FASD. Therefore, it is important to ascertain the knowledge, confidence and views towards FASD held by professionals receiving referrals. As well as this, gaining an understanding of what factors are associated with the likelihood of considering and/or assessing for FASD, will support with clinical training for all professionals. This includes understanding what information is most relevant within referrals, and ensuring this information is collected as early as the first contact with services, such as the first antenatal appointment. Based on these issues, the current study aims to investigate whether clinicians will consider FASD as a potential diagnosis and if they will select appropriate assessments for FASD, when presented with a referral of a child with neurodevelopmental difficulties. The researchers are also interested in what factors act as facilitators or barriers, such as clinician factors and referral information, when considering potential FASD from a referral. Due to the literature cited, we predict that clinicians are less likely to consider potential FASD unless PAE is mentioned. Exploring these factors will support with the current difficulties of under-diagnoses, making the project clinically applicable. The researchers aim to investigate the likelihood of a clinician recognising FASD features in a way that is more in line with how they will work 'in real life' and therefore not primed. It is also hoped that by not advertising the study as research investigating FASD, it will be less susceptible to self-selection bias in regard to only attracting participants who are interested or knowledgeable within this area.

Research Questions

In line with the aforementioned aims of the current study, the research questions are:

1) How does the inclusion of PAE information in a case vignette impact whether a clinician considers a potential diagnosis of FASD and appropriate assessment methods?

2) What factors are associated with an increased likelihood that clinicians will opt for a potential diagnosis of FASD and select appropriate assessment methods?

3) How confident do clinicians feel and what are the current attitudes towards recognising and assessing for FASD in children?

Method

Participants

Ethical approval was granted by the Faculty of Medicine and Health Sciences Research (FMH) Ethics Committee, at the University of East Anglia (ETH2223-0111). Clinicians with current or prior experience within the last two years of working with neurodivergent children in the UK were invited to participate in an online study. This timeframe was chosen to ensure that the experiences reported on were reflective of recent clinical practice, since the introduction of the FASD clinical standards in 2021. The project was advertised as a study investigating decision making by clinicians in ND services. Omitting information regarding the FASD focus of the study was deemed necessary, in order to gain results representative of all clinicians who may come into contact with possible FASD presentations, and not just clinicians who may have entered the study due to their interest or experience in FASD presentations. Participants were required to had trained and were currently registered within the UK, as one of the following disciplines: Clinical Psychologist, Psychiatrist, Occupational Therapist, Social Worker, Nurse or Speech and Language Therapist. Whilst NICE cite that further clinical professions may be involved in an assessment of FASD, the scope of the current project limited how many professions could be included in terms of recruitment and statistical analysis, thus only these six professions were included. The exclusion criteria included professionals who had trained or were practising outside of the UK due to the study investigating the current training needs within the UK. Participants were recruited through social media, through contact with professional groups for each profession, and through universities. All participants were given the option to opt into a raffle to win a voucher, to acknowledge the time and effort taken to complete the survey. For more information, see Chapter 5.

Measures

Demographic, employment and education information

Participants completed a survey for the following information to be collected: age, gender, current profession, amount of experience in neurodevelopmental settings in years, and level of education. Participants answered with the use of tick boxes, except for gender for which an open text box was supplied. Research suggests that options other than a binary male/female choice avoid making assumptions about gender, thus making the survey more accessible (Lindqvist et al., 2018). The purpose of this phase was to collect clinician information which may predict whether a participant considers FASD as a potential diagnosis and selects appropriate assessment methods. This survey can be found in Appendix E.

Clinical vignettes

Two vignettes of referral letters were created for the purpose of the study. Vignette studies are viewed as a "hybrid" option involving survey and experimental methods. These are cited to be highly generalisable and offer high external validity, and can be used to examine clinical judgements and decision-making processes (Evans et al., 2015). This methodology is common in clinical practice research, including studies exploring ND conditions (for example, Whitlock et al., 2020). These were created with the involvement of National FASD, a charity dedicated to increasing awareness and supporting people with FASD, to ensure that the referral contained information typical of a real referral received by ND services. The referrals contained information about a 12-year-old girl with cognitive, behavioural and emotional difficulties. In condition A, this referral letter contained information regarding PAE, whereas this information was excluded in condition B (see Appendix F). Aside from this, both of the referrals were identical. The purpose of this manipulation was to see if this information influenced the clinician's plans around assessment and diagnosis.

Clinical decision survey

This survey asked participants to document their initial thoughts about potential diagnoses and assessment plans via open text boxes (see Appendix G). The purpose of this survey was to gather data relating to the number of participants who consider FASD as a potential diagnosis and consider appropriate assessments that they would use.

Confidence, practice and views survey

This survey involved a series of questions relating to confidence in assessing and diagnosing FASD, current practice regarding FASD, and views on the amount of training received. Questions were answered using a five-point Likert scale ranging from 1 (strongly disagree) to 5 (strongly agree) (see Appendix H). Likert scales were chosen to reduce the amount of burden to the participant. Although this survey was researcher designed and therefore an unvalidated measure, it was developed in collaboration with National FASD, to ensure the quality of information being retrieved is suitable and useful for the FASD research community. This organisation was consulted regarding the content and style of the survey questions and response options, prior to the project going live. The purpose of survey was to gather data which would hopefully give an insight into what factors act as facilitators or barriers to clinicians considering FASD as a potential diagnosis.

Procedure

Please see Appendix I for a flowchart of the procedure.

Following the 'Demographic, employment and education information' questionnaire, participants were randomised to either condition A or B and presented with the corresponding clinical vignette. After, participants documented their initial thoughts around potential diagnoses and assessment tools within the 'Clinical decision survey'. All participants were then presented with an information sheet advising them of the new NICE standards about FASD assessment (see Appendix J). Finally, participants were invited to answer the 'Confidence, practice and views survey'. Participants were presented with a debrief and an opportunity to enter a raffle. See Chapter 5 for further details.

Data analysis

Prior to statistical analysis, data from the 'Clinical decision survey' were extracted and transformed into codes. To do this, for question one ("From reading the case vignette, what potential diagnosis/diagnoses come to mind that you might consider and wish to further assess for?"), the following categories were created based on the responses given: FASD (incorporating any associated disorder which would fall under this umbrella term), ASD, ADHD, Language disorders, Learning needs/cognitive disorders, emotional disorders, behavioural disorders, sensory processing disorders, executive functioning disorders, and 'other' (incorporating responses which did not fit into other categories, see Chapter 6 for more information). All responses were assessed and coded against each of the diagnoses categories, with 0 representing that they did not report a particular diagnosis, and 1 representing that the particular diagnosis was reported. Participants were able to record more than one diagnosis. For question two ("What would your full assessment include"), categories were created by including all the criteria documented within the quality standards, plus common responses made by participants. These included an ND assessment, plus the 10 individual domains of an ND assessment (motor skills, neuroanatomy/neurophysiology, cognition, language, academic achievement, memory, attention, executive function including impulse control and hyperactivity, affect regulation, and adaptive behaviour/social skills/social communication), medical/physical assessment, sentinel facial features, genetic testing, alcohol use, developmental history, observations, interviews/reports, and screening tools. Again, responses were coded as either 0 (not reported) or 1 (reported).

To account for ambiguity, all responses were reviewed by the research team who all came to a consensus on the appropriateness of the response before coding.

Research questions were explored with descriptive statistics and a series of logistic regressions.

Descriptive statistics were utilised to explore the range of diagnoses and assessment methods considered by all participants. Chi Square analysis was performed to investigate the

difference in FASD diagnosis considerations and appropriate assessment methods, between the PAE and non PAE condition. Researchers were also interested in what other possible diagnoses/disorders were reported by the sample, and how this differed based on profession.

To explore the assessment tools which were considered by the participants, two methods were utilised. Firstly, the core criteria for an assessment of FASD, documented within the SIGN guidelines, were used to code the responses against. These included three core areas: the 10 sections of a neurodevelopmental assessment, a medical assessment and an assessment of sentinel facial features. Secondly, other criteria relevant to prenatal alcohol disorders, as well as general neurodevelopmental assessments, were included and the data were coded against. These included: genetic testing, alcohol use, developmental histories, observations, interviews/reports and screening tools).

If participants recorded that they would assess all the three areas of the SIGN guidance, they were coded to have provided a full assessment (coded as 2). If participants mentioned one or more areas of the SIGN guidance, there were coded to have provided a partial assessment (1). If there were no mention of any of the SIGN guidance criteria, they were coded to have not provided an appropriate assessment (0). For the purpose of this project, these responses were described as inappropriate assessments, to reflect how the guidance criteria were not included in their response, rather than the nature of the responses they provided. Descriptive statistics were used to explore these results.

To explore what predicted the consideration of a potential FASD diagnosis and the selection of appropriate assessment methods, a series of logistic regressions were completed for each participant group.

For the logistic regression exploring for FASD diagnosis, the dependent variable (DV) was the inclusion/exclusion of FASD as a potential diagnosis. For the logistic regression exploring appropriate assessment methods, responses previously coded as full or partial assessments (described above), were collapsed into one category: appropriate assessment. The DV for this regression was the type of assessment reported by the participants, either appropriate or inappropriate. Predictor variables for all regressions were age, gender, qualification, profession, years' experience in ND services, and experimental condition: PAE information versus no PAE information.

Descriptive statistics were used to explore the breakdown of clinician self-ratings, in relation to confidence in recognising and assessing for FASD, their views about diagnosing FASD, the amount of training or teaching they have received about FASD, and their current practice (see Appendix H). This data was explored descriptively to gather an understanding about how these factors may act as

facilitators or barriers to assessing for FASD. The results were also explored by profession, to investigate differences in training and confidence between the different roles.

Additional information regarding statistical assumptions, power and sample size, and sensitivity analysis can be found in Chapters 5 and 6. All statistical analyses were performed utilising SPSS statistical package, version 28, with an α of .05 used to determine statistical significance.

Results

During data collection, the survey was infiltrated by suspected survey bots; automated programs which are designed to complete online surveys typically for financial gain. The survey was closed, and stringent criteria was placed against the available data in an effort to remove hypothesised false entries, based on commonalities within the data. The data reported within this project is the remaining, cleaned data. Full details of this process can be found in chapter 5.

Sample Characteristics

A total of 139 participants completed the online study, with 65 (46.8%) randomly allocated to the PAE information group and 74 randomly allocated to the non PAE information group, via the online platform. A greater proportion of females completed the survey (74.8%) and the majority of participants fell within the 31-40 age category (52.5%). Most of the participants held a Master's degree (30.9%) or higher level qualification (doctoral level; 28.8%). The most common profession was Clinical Psychologist (30.2%) followed by Speech and Language Therapist (24.5%). Finally, most participants held between 6-10 years' experience of working with neurodivergent people (37.4%). Table 5 gives an overview of the sample characteristics.

Table 5: Sample Characteristics

Sample Characteristics		*n*	%
Age	21-30	25	18
	31-40	73	52.5
	41-50	29	20.9
	51-60	9	6.5
	61-70	3	2.2
Gender	Male	35	25.2
	Female	104	74.8
Qualification	Cert. of higher education	5	3.6

	Foundation degree	13	9.4
	Bachelors	38	27.3
	Masters	43	30.9
	Doctoral	40	28.8
Profession	Clinical Psychologist	42	30.2
	Psychiatrist	9	6.5
	Speech & Language Therapist	34	24.5
	Occupational Therapist	32	23
	Social worker	5	3.6
	Nurse	17	12.2
Experience (Years)	<1	5	3.6
	1-5	47	33.8
	6-10	52	37.4
	11-15	13	9.4
	16+	22	15.8

The impact of PAE information on diagnosis and assessment tools

Diagnosis

Descriptive statistics found that the most commonly considered diagnosis was ADHD with a total of 98 responses, followed by ASD (83) and then cognition/learning needs (41). Please see Figure 3 for an overview of diagnoses considered by participants. From this, it can be seen that FASD was the 5th most popular diagnosis.

For participants who received PAE information within the referral, 26 (40%) stated that they would consider FASD as a potential diagnosis to explore, compared to 2 (2.7%) of participants not receiving this information. A Chi-Square test of Association found this difference to be significant ($\chi^2(1) = 29.93$, p <.001), indicating that there was an association between PAE information within the referral and considerations of FASD as a diagnosis. Table 6 displays these results.

Table 6: Diagnoses considered by professionals

Diagnosis	PAE group		Non-PAE group		Total	
	n	%	*n*	%	*n*	%
ADHD	49	75.4	49	66.2	98	70.5
ASD	41	63.1	42	56.8	83	59.7
Cognition/learning needs	14	21.5	27	36.5	41	29.5
Emotional	11	16.9	18	24.3	29	20.9
FASD	26	40	2	2.7	28	20.1
Other	5	7.7	11	14.9	16	11.5
Language	8	12.3	6	8.1	14	10.1
Sensory	1	1.5	9	12.2	10	7.2
Executive functioning	0	0.0	3	4.1	3	2.2
Behavioural	1	1.5	0	0.0	1	0.7

PAE: Prenatal alcohol exposure. ADHD: Attention Deficit Hyperactivity Disorder. ASD: Autism Spectrum Disorder. FASD: Fetal Alcohol Spectrum Disorder.

The consideration of diagnosis was also explored by profession via descriptive statistics. The results revealed that Clinical Psychologists were most likely to consider FASD as a diagnosis from all of the professions, particularly when PAE information was given in the referral (10, 53%) compared to when this information was missing (1, 4%). FASD was not considered by psychiatrists or social workers; psychiatrists were most likely to consider emotional disorders, whereas social workers were most likely to consider ADHD, ASD or a learning/cognition disorder. These results are described in more detail in Table 7.

Table 7: Percentage of diagnoses by profession, comparing PAE and non PAE conditions

		Profession							
		Clinical Psychologist		SLT		OT		Nurse	
Diagnosis	Condition	*n*	*%*	*n*	*%*	*n*	*%*	*n*	*%*
FASD	PAE	10	52.6	5	31.3	8	50.0	3	33.3
	Non PAE	1	4.3	0	0.0	0	0.0	1	12.5
ADHD	PAE	17	89.5	12	75.0	8	50.0	5	55.6
	Non PAE	20	87	10	43.5	12	75.0	8	100.0
ASD	PAE	14	73.7	8	50.0	11	68.8	5	55.6
	Non PAE	19	82.6	7	38.9	7	43.8	5	62.5
Language	PAE	1	5.3	7	43.8	0	0.0	0	0.0
	Non PAE	2	8.7	3	16.7	1	6.3	0	0.0
Cognition	PAE	7	36.8	2	12.5	3	18.8	2	22.2
	Non PAE	12	52.2	4	22.2	6	37.5	2	25.0
Emotional	PAE	2	10.5	4	25.0	1	6.3	1	11.1
	Non PAE	2	8.7	9	50.0	4	25.0	0	0.0
Behavioural	PAE	0	0.0	0	0.0	0	0.0	1	11.1
	Non PAE	0	0.0	0	0.0	0	0.0	0	0.0
Sensory	PAE	0	0.0	0	0.0	0	0.0	1	11.1
	Non PAE	3	13.0	3	16.7	1	6.3	1	12.5
Executive Functioning	PAE	0	0.0	0	0.0	0	0.0	0	0.0
	Non PAE	2	8.7	1	5.6	0	0.0	0	0.0

Note: Psychiatrists and Social Workers not included due to low sample size. FASD = Fetal Alcohol Spectrum Disorder. ADHD = Attention Deficit Hyperactivity Disorder. ASD = Autism Spectrum Disorder. SLT = Speech & Language Therapist. OT = Occupational Therapist. PAE = Prenatal alcohol exposure

Assessments

Overall, only one participant stated they would assess the three core criterion included within the SIGN guidance. Within the PAE group, 51 participants recorded partial criteria for an FASD assessment, compared to 55 participants within the non PAE group. In the PAE group, 14 participants did not provide appropriate assessment tools, compared to 18 of the non PAE group.

Moreover, 75.4% of participants in the PAE group stated that they would perform an ND assessment or mentioned one or more sections within an ND assessment, compared to 67.6% within the non PAE group. In the PAE group, 14% of participants mentioned a medical/physical assessment, compared to 19% within the non PAE group. Finally, none of the participants within the PAE group stated they would assess for sentinel facial features, compared to 1% of participants within the non PAE group. Further details can be found in Appendix R.

Factors which predict clinicians considering FASD and appropriate assessment methods

FASD Diagnosis

A binary logistic regression was performed to explore what predicted participants to consider FASD as a potential diagnosis. A preliminary analysis exploring the assumption of multicollinearity suggested that this assumption was met. Standardised residual values were inspected which found four outliers within the data (Std residual: 1.4, 1.89, 1.98, 4.81). As the logistic model was statistically significant χ^2 (19, N = 139) = 57.909, p < 0.001, Nagelkerke R^2 = 53.8%, correctly classifying 84.9% of the cases, the four outliers were kept within the data set.

From the six predictor variables included (age, gender, qualification, profession, experience and condition: PAE information versus no PAE information), only one variable was found to significantly increase the likelihood of participants considering FASD: the condition they were placed in. Participants within the PAE group were 34.4 times more likely to consider FASD compared to the non PAE group. Further details can be found in Appendix S.

FASD appropriate assessments

A binary logistic regression was also performed to explore what improved the likelihood of participants reporting appropriate assessment methods or tools based on the referral information they received. For this analysis, the one participant recorded to state a ‘full’ assessment was included within the partial group, thus the model predicts the likelihood of a partial assessment method being stated compared to an inappropriate assessment method. The predictor variables remained the same as the diagnosis regression and thus met the necessary assumptions. Whilst the model was

able to correctly classify 78.4% of the cases, it was found to not be a statistically significant model (χ^2 (19, N = 139) = 24.59, p = 0.174, Nagelkerke R^2 = 24.6%). This suggests that the model was not able to distinguish between those selecting partial appropriate assessment tools or not selecting any appropriate assessment tools. Further details can be found in Appendix T.

Confidence and attitudes towards recognising and assessing for FASD

Descriptive statistics were performed to explore the participants ratings in relation to a survey exploring perceived confidence, attitudes, and opinions towards FASD diagnosis. Overall, most participants selected 'Agree' when asked "I am confident in my ability to recognise FASD" (38.8%). They were less confident in selecting appropriate assessments to assess FASD, with the majority of participants selecting 'disagree' (30.2%). A total of 38.2% of participants 'Strongly disagreed' that they had received adequate teaching about FASD during their training, although 32.4% 'Agreed' that they had received further training post qualification.

Most participants 'strongly agreed' that they were confident in asking about alcohol intake during pregnancy, with 32.4% 'agreeing' that they regularly ask about this during their assessments. 36.7% of participants 'strongly disagreed' when asked if they would only ask about alcohol intake if the child presented with sentinel facial features, however the majority 'agreed' that they would only consider FASD as a potential diagnosis if prenatal alcohol exposure was mentioned the referral (33.1%). Finally, 44.6% of participants 'strongly agreed' that there is still merit in diagnosing a condition for which there is no cure. See Table 8 for an overview of these results.

Table 8: Ratings from the Confidence, Attitudes and Opinions survey

Question	Strongly disagree		Disagree		Neutral		Agree		Strongly agree	
	n	%	n	%	n	%	n	%	n	%
I am confident in my ability to recognise FASD	8	5.8%%	34	24.5%	25	18%	54	**38.8%**	18	12.9%
I am confident in my ability to select appropriate assessments when querying FASD	20	14.4%	42	**30.2%**	21	15.1%	31	22.3%	25	18%
I received adequate training about FASD during my training	54	**38.8%**	29	20.9%	7	5%	37	26.6%	12	8.6%
I have received further training about FASD since qualifying	30	21.6%	23	16.5%	12	8.6%	45	**32.4%**	29	20.9%
I am confident in asking about alcohol intake during pregnancy	3	2.2%	18	12.9%	19	13.7%	45	32.4%	54	**38.8%**
I regularly ask about alcohol intake during assessments	16	11.5%	17	12.2%	19	13.7%	45	**32.4%**	42	30.2%
I would only ask about alcohol intake if a child presents with the associated facial features	51	**36.7%**	28	20.1%	27	19.4%	25	18%	8	5.8%
I would only consider FASD if prenatal alcohol exposure is mentioned in the referral	29	20.9%	36	25.9%	15	10.8%	46	**33.1%**	13	9.4%
I believe there is still merit in diagnosing a condition/disorder for which there is no cure	2	1.4%	3	2.2%	14	10.1%	58	41.7%	62	**44.6%**

Percentages in bold are the most common responses for each question. FASD = Fetal Alcohol Spectrum Disorder.

The responses to the confidence, attitudes and views survey were also explored in terms of profession. The results indicated that Psychiatrists were most confident in their ability to recognise FASD, with 100% selecting 'agree' or 'strongly agree'. All other professions selected 'agree' or 'strongly agree', 53% or less of the time. In terms of only asking about alcohol intake if sentinel facial features were present, psychiatrists were more ambivalent, with 67% selecting neutral, whereas 55% of psychologists disagreed with this statement. Psychiatrists were also more likely to only consider FASD as a potential diagnosis if PAE was mentioned in the referral, with 77% agreeing or strongly agreeing with this statement. In comparison, the majority of other professions (nurses, social workers, OTs, and clinical psychologists) were more likely to disagree with this statement, with up to 76% selecting 'disagree' or 'strongly disagree'. Tables 9 and 10 illustrate these results.

Table 9: Clinician's rating for the question 'I am confident in my ability to recognise FASD'

	I am confident in my ability to recognise FASD				
Profession	Strongly disagree	Disagree	Neutral	Agree	Strongly agree
Clinical Psychologist	4.8%	28.6%	16.7%	**40.5%**	9.5%
Psychiatrist	0.0%	0.0%	0.0%	44.4%	**55.6%**
SLT	11.8%	20.6%	17.6%	**38.2%**	11.8%
OT	6.3%	25.0%	15.6%	**43.8%**	9.4%
SW	0.0%	**40.0%**	20.0%	**40.0%**	0.0%
Nurse	0.0%	29.4%	**35.3%**	23.5%	11.8%

Note: Bold font represents highest percentage. SW = Social worker. OT = Occupational Therapist. SLT = Speech & Language Therapist.

Table 10: Clinician's rating for the question 'I would only consider FASD as a potential diagnosis if prenatal alcohol exposure is mentioned in the referral'

	I would only consider FASD as a potential diagnosis if prenatal alcohol exposure is mentioned in the referral				
Profession	Strongly disagree	Disagree	Neutral	Agree	Strongly agree
Clinical Psychologist	26.2%	**50.0%**	7.1%	14.3%	2.4%
Psychiatrist	0.0%	0.0%	22.2%	33.3%	**44.4%**
SLT	5.9%	5.9%	17.6%	**58.8%**	11.8%
OT	25.0%	25.0%	9.4%	**37.5%**	3.1%
SW	20.0%	**40.0%**	20.0%	20.0%	0.0%
Nurse	**41.2%**	17.6%	0.0%	23.5%	17.6%

Note: Bold font represents highest percentage
SW = Social worker. OT = Occupational Therapist. SLT = Speech & Language Therapist

Discussion

This study investigated the decision making process made by a range of health professionals documented to be important in the process of assessment and diagnosis of FASD (SIGN, 2019), when presented with an online survey. The results indicated that based on the referral information, ADHD was most likely to be considered by the professionals overall, within both conditions. This is in line with previous research which indicates that ADHD is one the most common misdiagnoses of FASD (Wozniak et al., 2019). Research also indicates that externalising disorders such as ADHD are less stigmatising than FASD (Lange et al., 2017), which was the fifth most common response, indicating that this may influence a clinicians decision process.

Regarding factors that affect this decision making process, clinical psychologists were most likely to consider FASD, closely followed by OTs. Psychiatrics and social workers did not consider FASD at all, posing the question as to what makes this difference, although sample sizes were small for these professions thus conclusions are limited.

The inclusion of PAE information within the referral was shown to significantly increase the amount of FASD consideration, indicating that this information is key for professionals receiving referrals. Whilst probable or confirmed prenatal alcohol exposure is necessary for a diagnosis of FASD, missing PAE information in a referral does not necessarily mean that it was not present and

does not need enquiring about. As a comparison, a diagnosis of ASD requires a developmental history to be taken to ensure that difficulties have been present from a young age. Despite this information being missing, clinicians were still able to consider ASD as a potential diagnosis. This suggests that clinicians can consider potential diagnoses even when there is information missing from a referral, but FASD is a diagnosis that is more difficult to hold in mind, despite being the most common, non-genetic cause of learning disability within the UK (British Medical Association, 2016). One possible reason for this could be the higher levels of awareness that exists for other ND disorders which have similar phenotypes, which again leads to misdiagnoses or diagnostic overshadowing.

As there was a significant impact on FASD considerations when additional referral information was given, future research would benefit from exploring the impact of other pieces of referral information. For example, Abel (1995) reported that low SES populations have a significantly higher proportion of FAS diagnoses than middle/upper class populations. Moreover, other risk factors for poor outcomes such as poverty and trauma (Yumoto et al., 2008), are also associated with an increased risk of FASD (Moritz et al., 2023). Understanding whether factors such as these impact on a clinician's consideration of diagnosis may support with training to finetune the information captured in referrals.

The results indicated there were no predictors which were significantly stronger at predicting whether a clinician would select appropriate or inappropriate assessment methods. This is a particularly interesting finding, as this suggests that regardless of how long a clinician has been working with neurodiverse individuals, the level of qualification they hold or their profession, they will still struggle to identify the necessary components of an FASD assessment. A total of 106 participants were able to consider a partial assessment, which indicates that whilst many clinicians feel comfortable with assessing neurodiversity, they are less inclined to consider how to assess for the specificities of FASD. This is not necessarily surprising given the findings, as to consider appropriate FASD assessment tools would also depend on considering FASD as a diagnosis initially.

The 'confidence, practice and views' survey was able to explore these intricacies in more detail, by offering an insight into the potential barriers to assessing and diagnosing FASD, experienced by clinicians. From this, clinicians indicated that whilst they agreed they felt confident to identify FASD, they disagreed that they felt confident in selecting appropriate tools, corroborating the results found previously. In addition to this, most participants felt that they would only consider FASD if PAE was mentioned in the referral. Despite this, even when PAE was included in the referral, only 26 (40%) included FASD within their considered diagnoses. It would be interesting to investigate why participants reported that they would consider FASD but did not do this in practice. One

possibility could be that clinicians have many diagnoses to consider when reviewing referrals, which would understandably make it very difficult keep all diagnoses in mind, particularly when more commonly diagnosed disorders present very similarly. However, when they are reminded of FASD, such as within the information given just before the confidence, practice and views survey, this can support clinicians to hold FASD in mind. This study purposely did not give any information about FASD prior to the vignettes as to not prime participants, however, it would be interesting to see whether giving information, whether specifically about FASD or a list of all ND disorders, would have improved clinicians rate of FASD consideration. If so, this would indicate that more investment is needed to increase awareness of FASD to ensure that clinicians can hold the disorders in mind when reviewing referrals. Another possibility for the disparity between the self-reported views and the experimental data is response bias. There is a chance that clinicians responded to the confidence, practice and views survey in a way which felt more favourable, given their experience and nature of their work.

The survey indicated that most professions did not feel that they received adequate pre-qualification training about FASD, which is possibly one of the reasons why clinicians did not hold it in mind when considering diagnoses. This suggests a universal deficit found in most clinical profession training courses. Whilst there is a limit to how much can be taught within a fixed period of training, given the prevalence, vast range of difficulties as well as the financial burden of FASD, it can be argued that the disorders should receive the same amount of attention as other neurodevelopmental conditions. In contrast to the majority, all psychiatrists felt that they had received adequate teaching, but were also most likely to indicate that they had received further training since qualifying.

Whilst the majority of participants indicated that they strongly disagreed that they would only consider FASD if the sentinel facial features were present, 43.2% were neutral or agreed with the statement. In particular, 78% of psychiatrists answered ‘neutral’ or ‘agree’ to this statement. As previously stated, with only 10% of individuals with FASD presenting with sentinel facial features, this would leave 90% of individuals undiagnosed (NICE, 2022). This is another indicator of misdiagnosis and a probable reason for the high rates of ASD and ADHD considered within the study. This also indicates that there has not been a shift as of yet in behaviour since the introduction of the quality standards. The success of system or treatment level approaches is dependent on the clinicians who work within these systems, and research suggests that change in clinical practice can take time due to barriers such as internal motivations as well as service delivery characteristics (Riemer et al., 2005). Given the time taken for change to be witnessed, if the study were to be repeated in five

years, we may see an increase in FASD considerations. However, currently, the results indicate that again, more awareness about the disorders is needed.

Clinical implications

Overall, the survey indicated that there are several barriers in place which limit the chances of clinicians holding FASD in mind when considering potential diagnoses and assessments. These include the amount of training on pre-qualification courses, for all professions. This indicates that FASD should be given greater consideration when academic syllabuses are revised. Overlooking this, professional bodies should ensure that training centres meet the necessary requirements in terms of how much FASD is covered.

Another element of training refers to the amount of information given within referrals. The evidence suggests that clinicians are more able to consider FASD when there is PAE information in the referral, thus the more this is explicitly stated, the more likely FASD will be held in mind. This requires this information to be collected from individuals whenever they are in contact with clinical services, antenatally and beyond. Therefore, specific training regarding FASD awareness, as well as collecting and reporting alcohol usage information, should be available for midwives, as well as those completing referrals. The NICE quality standards already give guidance on how this information should be collected (see Chapter 3), however how this has been developed practically is unknown. Thus, exploring this would be beneficial to ensure that midwives feel supported within the pathway.

Ensuring that necessary information is shared with the right clinicians, depends on effective multi-disciplinary team (MDT) working. Again, the necessity of MDTs has been highlighted within the clinical standards, however, how achievable this is may be questioned. How and where children are diagnosed with ND conditions such as ASD and ADHD can differ nationally, with some accessing assessments within Child Development Teams (CDTs) and others through Child and Adolescent Mental Health Services (CAMHS) (Male et al., 2020). Increasing awareness for conditions such as ASD have led to long waiting lists, for centres for which budgets are not necessarily related to caseloads (Galliver et al., 2017). With strained services, access to all of the information required for a diagnosis of FASD may be hard to come by. Anecdotal evidence reported from The National Organisation for FASD (www.nationalfasd.org.uk), suggests that many services do not have clinical psychologists within the team in order to for cognitive assessments to be completed. With this is mind, it is unsurprising that there can be delays to diagnosis of FASD and clinicians may opt to provide a primary diagnosis for disorders which are more readily diagnosable than FASD (Brown et al., 2011). This suggests that services should be resourced in a way which the clinical standards align with, particularly as services may see an increase in referrals for FASD referrals as awareness increases.

Limitations

Whilst this study was the first known experiment to explore the clinical decision making of clinicians in relation to FASD, there are some limitations which are worth noting. Firstly, as the survey was the target of bot activity, the data had to be manually cleaned which made subjectivity unavoidable. It is possible that responses created by bots were still present in the reported data, or that responses made by real participants were removed. Substantial efforts were made to avoid this as much as possible, with a selection of rules created by the researchers to screen each response against (see Chapter 5 for more information). Moreover, sensitivity analysis was also completed for responses that the researchers were unsure about, to explore the impact on the overall results (see Chapter 5). These results indicated that responses which were 'at risk' of being made by a bot, did not alter the significance of the models or the predictors they identified as being significant. Overall, the researchers were confident that the remaining dataset was reflective of a 'real' sample.

Secondly, there were substantially smaller numbers in some of the professional groups compared to others, for example 42 Clinical Psychologists compared to five social workers, and due to this reason, analyses exploring profession may be underpowered and conclusions drawn are limited. A larger sample may have given more opportunity to see more considerations of FASD. In addition to this, the study included six key professions involved in the assessment and diagnosis of FASD, however there are other professions which were not included due to the scope of the study, which may have provided greater insight. These include paediatricians, educational psychologists, clinical geneticists, general practitioners (GPs), health visitors, midwives and obstetricians. Further to this, it could be argued that some of the clinicians included would not have access or knowledge of what an appropriate assessment should include, due to the scope of their profession. However, responses were included if clinicians mentioned further referrals to specific disciplines, and information about what should be assessed can be found publicly.

Thirdly, in order to capture the necessary information for data analysis for the assessments, responses were coded to have included a neurodevelopmental assessment even if they had only mentioned one of the areas of an ND assessment. This means that clinicians would have been able to appear as though they documented a 'full assessment', even without the knowledge of what a full ND assessment should cover. Whilst this had the potential to limit the results, only one participant did appear to document a 'full assessment', thus the impact was minimal.

Conclusion

The majority of clinicians do not consider FASD as a diagnosis when receiving a referral with neurodevelopmental concerns. This is significantly improved when PAE is mentioned in the referral,

and is most likely to be considered by Clinical Psychologists. This indicates that including information about PAE within a referral, possibly even if exposure is unknown, will support clinicians to keep FASD as a diagnosis in mind. The results indicate that the ability to select appropriate assessment tools to explore FASD is limited, regardless of profession, experience, and qualification, warranting further training in this area for all professions. Overall, greater awareness of FASD would support clinicians, antenatally and beyond, to consider alcohol intake and subsequent related disorders.

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Chapter 5: Additional methodology

The purpose of this chapter is to provide further information regarding the methodology utilised within the empirical paper. This includes additional information about power calculations, statistical assumptions, and the process of data cleaning. Ethical considerations will also be provided.

Empirical paper

The recruitment process occurred in two phases: social media drive and organisational drive. Social media recruitment commenced on 9th February 2023 and took place on Facebook and X (formally known as Twitter). A copy of the recruitment poster can be found in Appendix K.

Due to slow uptake with recruitment through social media, the organisational recruitment drive was introduced and commenced on 27th March 2023. Contact was established with 22 UK universities, randomly chosen from an online list of all UK universities, via email. The head of each professional department was identified from each university website. A copy of this email can be found in Appendix L. As well as this, contact was also made with the national professional bodies for each discipline, requesting that the study was publicised via their social media channels (Appendix M).

Sample size and power calculations

Currently, there is no gold standard in regard to how to approach a power analysis and sample size calculation for a logistic regression (Demidenko, 2006). A widely used guideline is the 10 Events Per Variable (EPV) ratio, however, this has been found to be unpredictable (van Smeden et al., 2016). Due to this, power analysis and sample size calculations were approached in a number of ways, documented below.

Software to perform power analyses on for logistic regressions typically require pilot raw data in order to define the parameters of the variables. Unfortunately, such data was not available for the current study, so a priori power analyses were not conducted. In order to gain some understanding of an appropriate sample size, a survey sample calculator (Raosoft, 2004) was used. This calculator requires the population sample to be entered in. Estimates of the number of clinical professions who have worked with neurodiverse populations were also very difficult to obtain. The following data was found:

- 276 registered speciality CAMHS psychiatrists (Royal college of Psychiatrists, 2021)
- 57024 registered children's nurses (Nursing and Midwifery Council, 2023)
- 32,502 registered children's social workers (Institute for Government, 2023)

- 41318 registered Occupational Therapists (speciality unknown; Health & Care Professions Council, 2021)
- 26000 registered practitioner psychologists (speciality unknown; Health & Care Professions Council, 2023)
- 17,240 registered Speech and Language Therapists (speciality unknown - Health & Care Professions Council, 2021)

Totalling these numbers estimated a population of 174,360 clinical professionals, however, this is only an estimate, due to more specific data relating to specialities, or the number of clinicians who may have worked with neurodiverse children, not being available. Because of this, sample sizes for each profession individually were not calculated, and instead, an overall sample size was considered. The survey sample calculator documents that sample numbers do not vary significantly for populations over 20,000. The calculator indicates that with a confidence interval of 95%, with a margin of error inputted as 5%, a total sample size of 73 would be required. This was found to be true when both estimates, 20,000 and 174,360, were inputted.

Post hoc power analysis indicated that, with an odds ratio of 34.4, a sample size of 139, the study was powered at 94%. However, this analysis utilises a specific odds ratio for one of the statistical analyses which took place and does not take into consideration the number of cases per level of each variable. Overall, as sufficient sample and power analysis were unable to take place, there is a possibility that the study is underpowered and thus, conclusions drawn are limited.

Ethical considerations

The research was approved by the University of East Anglia's Ethics Committee (ETH2223-0111) (Appendix N). To assure confidentiality and anonymity, participants were not required to enter their name to take part in the study. Although they entered some demographic information, participants were not identifiable as they were not required to enter the department or trust they work in. As the research was conducted online via a hyperlink, the risk of coercion was reduced as the participants were not known to the research team. It was deemed that a prize draw of a £20 Amazon voucher to acknowledge clinician's time felt appropriate within this research context. It was not anticipated that the project would cause any distress, however participants were invited to contact the researchers if they had any issues as part of the debrief.

The collection and storage of data were carried out in accordance with the General Data Protection Regulations (GDPR) and Data Protection Act (2018), which the chosen platform PsyToolkit complied with. Electronic data was stored with encryption on OneDrive, of which only the lead

researcher and primary supervisor had access to. Data will be stored in accordance with the Good Clinical Practice Guidelines, which states that research data must be kept for 10 years.

Recruitment

Participants were invited to take part in the study advertised through professional membership bodies or social media. A hyperlink and QR code were provided for those wishing to find out more information or enter the survey, which directed them to the survey platform.

Consent process

Participants were presented with an electronic participant information sheet (see Appendix I), providing the purpose of the study as well as ethical considerations. They were presented with a consent statement (see Appendix O) and were then invited to continue with the study, by checking a tick box and clicking 'Continue to survey', assuming informed consent. Consent was also assumed by participants completing the survey.

Debrief

Finally, participants were presented with debriefing information, signposts to FASD resources and information about how to contact the researcher for further information (see Appendix P).

Although participants were informed of the general study aims and methodology at the start of the study ensuring informed consent, they were blind to the specific topic of interest; FASD. The strengths and limitations of this approach were fully and deeply considered in line to balance the ethical factors and clinical applications of the study. Not informing the participants of the full aims of the study, could be perceived as deception and there is a possibility that participants may have felt blindsided. Following participation, participants may have felt they would have been less likely to participate had they known that the study was about FASD. However, in order to ascertain how clinicians currently react when faced with potential FASD presentations without prior priming, it was felt that this is the most appropriate experimental design. To support with possible feelings of deception, information regarding the full aims of the study were given at the earliest possible time in the procedure, following the clinical decision survey. Participants were also informed of their right not to continue and right to withdraw their data, as well as details on how to contact the researchers to discuss the project.

A full debrief was given at the end of the experiment. This included signposting to FASD resources and details on how to contact the researchers. Participants also had the opportunity to enter a prize draw for a £20 gift voucher to acknowledge the time taken to complete the survey. If a

participant wanted to enter this draw, they were informed that their email address would be needed, however this was not linked to their data or shared with anyone other than the researcher. Additional information about ethical considerations is available in Chapter 5.

Data collection

During the data collection phase of this project, it became apparent that there was a problem with the responses that had been submitted. There was a significant increase in the number of responses to the survey within a short amount of time, from 91 responses on 25th May 2023, to 887 on 20th June 2023. It was unlikely that this was a genuine response to the recruitment drive, given the sharp increase, therefore the survey was closed for the data to be inspected. Organisations who had supported with recruitment were also notified that the survey was closed. Figure 3 illustrates this increase in responses.

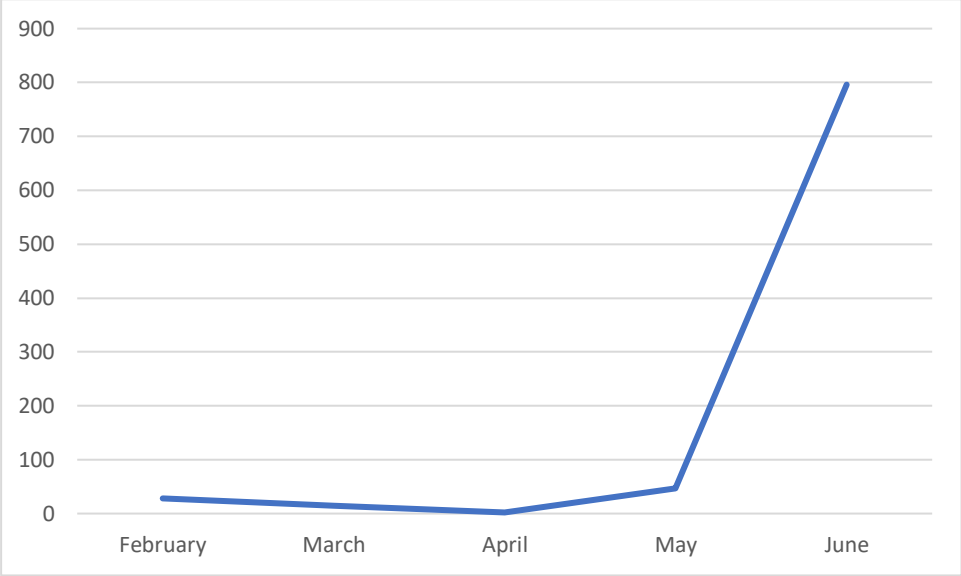

Figure 3: Number Of Responses to Empirical Study by Month

Upon inspection, it became clear that some of the responses were at odds with what would be considered typical responses. For example, there were many duplicates of the same answers, many empty response boxes, answers which were irrelevant to the question, and answers with odd characters, for example '~'. Following a discussion via supervision, it was deemed that the survey had been the target of 'bots'; individuals who utilise computerised programmes to complete surveys automatically on their behalf, in order to potentially gain access to something, such as a monetary prize. As participants were able to enter into a raffle for a £20 Amazon voucher, it felt likely that this was the reason for the 'invasion'.

Following this discovery, the research team discussed and evaluated the options moving forward. It was felt that there were two options: the first involved deleting the data already collected, and opening the survey back up again, with the inclusion of survey bot protection strategies. These include using Captcha technology or including questions within the survey which would indicate highly as to whether a participant was unlikely to be genuine, i.e., a bot. This option would support with collecting a new set of data that was less likely to be affected by bots, allowing for better data integrity. However, due to the element of deception involved in the survey, in which the participants were blind to the aim of the study, this approach would require a brand new sample who had not completed the survey before. This posed two problems. Firstly, it was evidently difficult to recruit participants for the survey, so recruiting a new sample of the same amount would be extremely tough given the limited timeframe for this thesis project. Secondly, assuring that the participants had not completed the survey the first time round, would have been impossible to control for. Conversely, research suggests that even with the inclusion of extra measures to protect against bot activity, "there is no sure way to conduct a bot-free study" (Simone, 2019, para 18).

The second approach which was considered, was to clean and use the original data collected. This approach avoided the methodological difficulties of finding a new sample, however, again had some notable flaws. There was no completely accurate way of knowing which responses were real and which were of bot activity. Cleaning the data would depend on human interpretation and thus open the door to sources of bias.

In this unfortunate situation, both approaches had strengths and limitations. However, given the lack of assurance that even with protective measures in place, the survey would not be fully protected, plus the methodological difficulties with finding a new sample, the decision was made to clean the original data. This approach was felt to be the strongest by all members of the research team, as well as the Ethics board who were notified of the problem (see Appendix Q). To maximise data integrity, a set of criteria were created by the whole research team, of which the responses were rated against. If a response met any of the criteria, they were excluded. These criteria and the reasons for their inclusion are documented below:

1) The response contains '~' at the end: This was included due to all responses with this symbol appearing to be incongruent with the topic or question.
2) The gender open text box contained the following: boy, girl: This was included due to the assumption that all clinical professionals would use a more age-appropriate term.
3) The time taken to complete the survey was recorded as one minute or less: This was included due to the unlikelihood of a participant being able to read all of the information and answer each question within one minute or less.

4) Not a complete data set: Any responses which had missing data were discarded.
5) Duplicate responses: Any responses which had the exact same responses to each question to another participant, were discarded. This was due to the unlikelihood of two different participants entering the exact same information.
6) The response was not written in the English Language: This criterion was included due to the inclusion criteria for the study stating that the participants needed to have trained and practiced within the UK, thus the unlikelihood of a participant meeting this criterion and not using English Language was high. As well as this, translations of the free text boxes may not have been accurate.
7) The response indicated that the participant was a Clinical Psychologist/Psychiatrist but held less than a Master's qualification: This was included due the lowest level of qualification needed to become a Clinical Psychologist was a Master's before 1994, which then evolved to a doctoral qualification in 1995. Psychiatrists were included within this due to the profession also needing the highest levels of qualification.
8) The response indicated that the participant held less than a certificate level qualification: This was included due to a higher-level qualification being needed for every profession included.
9) The response included repeated information for both the diagnosis question and the assessment question: This was included because the responses were not congruent with the questions asked.
10) No clear diagnosis or assessment reported: This was included due to the responses not being congruent with the questions asked.
11) Survey was completed at the same time and took the same duration as another participant in addition to the same quantitative responses on the Likert scales. Both criteria were included together because it was felt that there was a significantly small likelihood that either phenomenon could have happened naturally, but the chance of them happening together was felt to be minimal.

As a result from cleaning the data, the overall responses fell from 888 to 139. Whilst this was a large decrease and resulted in the study possibly being underpowered, it was felt that this was the safest way to keep the integrity of the data.

Chapter 6: Extended Results

Meta-Analysis

Moderator analysis: location

Given the high number of studies taking place in certain parts of the world, exploratory analysis was completed to see if there were any differences in the prevalence reported for each psychological need between countries. The studies were categorised into three groups: studies from Canada, studies from the United States of America (USA), and studies from the rest of the world (ROW). The last category was created due to there being minimal studies from other parts of the world when compared to Canada and USA.

There is currently no universally accepted rule about how many studies is considered 'sufficient' when completing moderator analysis, although Fu et al. (2011) advised a minimum of four. Therefore, this rule was adopted and any category containing less than four studies was excluded. Therefore, Autism Spectrum Disorder (ASD) was not included in the sub analysis due to there being less than four studies within each category. The results indicated that whilst prevalence rates for each psychological need varied across the location categories, however there were no statistically significant differences found for Depression ($\chi^2(2)$ =2.79, *p* = 0.25), Anxiety ($\chi^2(2)$ = 1.51, *p* = 0.47), behavioural difficulties ($\chi^2(2) = 0.72$, *p* = 0.70), or ADHD ($\chi^2(2) = 0.37$, *p* = 0.83). Table 11 displays further statistical information.

Table 11: Sub-group analysis by location of study

Psychological Need	Location	Number of studies	N	Pooled prevalence estimate	95% Confidence interval		I^2	*Q*	*P*-value
					Lower	Upper			
Depression									
	Canada	12		9.7%	4.2%	17%	95.7	354.01	<0.001
	USA	6		19.4%	10.5%	30.1%	90.01	44.16	<0.001
	ROW	4		13%	4.1%	25.4%	88.97	15.70	0.001
Anxiety									
	Canada	11		13.9%	5.7%	24.7%	97.43	578.05	<0.001
	USA	5		18.4%	6.1%	34.1%	88.74	44.23	<0.001
	ROW	4		31.4%	3%	69.1%	98.39	130.60	<0.001

Behavioural								
	Canada	14	22.4%	9.8%	37.9%	96.09	236.45	<0.001
	USA	5	13.8%	3.1%	29.3%	93.03	39.05	<0.001
	ROW	5	16.4%	3.6%	34.8%	92.78	54.36	<0.001
ADHD								
	Canada	22	58.2%	48.4%	67.6%	94.75%	199.34	<0.001
	USA	12	57.3%	41.2%	72.6%	94.97%	134.08	<0.001
	ROW	13	53.6%	49.9%	63.1%	84.82%	118.87	<0.001

Empirical Paper

Diagnoses considered by clinicians

For data analysis of the diagnoses considered by clinicians, all responses which did not fit into the categories created, were amalgamated into an 'other' category. Included under this category were the following responses: wider health needs, attachment difficulties, trauma and personality disorders.

Assessments considered by clinicians

For data analysis of the diagnoses considered by clinicians, all responses which were vague or did not fit into the categories created, were amalgamated into an 'other' category. Included under this category were the following responses: assessment of adverse childhood experiences (ACEs)/trauma, risk assessment, the Health of the Nation Outcome Scale (HoNOS), educational psychology assessment, assessment of attachment, history of previous interventions, psychological assessment, Coventry grid assessment, family assessment, behaviour assessment, symptom assessment, laboratory tests, childhood psychiatric assessment and sleep assessment.

Sensitivity Analysis

A selection of the entries were included in sensitivity analysis. These were responses which were considered to be 'at risk' of being bot responses, due to having the same start and end date/time and the same completion durations. As mentioned, it was felt that there was a small chance that these could still be real responses. Thus, they were not discarded, but a sensitivity analysis was performed to see if including these responses skewed the overall results. A total of 28 responses were removed, resulting in 111 included for analysis. The statistical tests were repeated to check if significance levels changed (for example a non-significant result became significant, or vice

versa); if they were to change, then further analyses were planned to examine whether there was a statistical difference between the significance levels.

The impact of PAE information on diagnosis

A Chi-Square test of Association was conducted to explore whether the condition of receiving PAE information within the referral impacted on the number of FASD diagnosis considerations. With the 'at risk' responses, this difference to be significant ($\chi^2(1) = 29.93$, p <.001). With the 'at risk' group removed, the difference was still found to be significant ($\chi^2(1) = 26.92$, p <.001). This indicates that the 'at risk' responses did not impact on the significance of the model.

Factors predicting clinicians considering FASD

A binary logistic regression was performed to explore what predicted participants to consider FASD as a potential diagnosis. The logistic model was statistically significant χ^2 (19, N = 139) = 57.909, p = <0.001, Nagelkerke R^2 = 53.8%, correctly classifying 84.9% of the cases. Only the condition (PAE group versus non PAE group) was found to be significant (OR = 34.37, p<0.001). With the 'at risk' responses removed, the results remained similar χ^2 (19, N =1 11,) 50.350, p<0.001, with still only condition being significant (OR = 42.06, p<0.001). This indicates that the 'at risk' responses did not impact on the significance of the model or its predictor variables.

Factors predicting clinicians considering appropriate FASD assessment methods

A binary logistic regression was also performed to explore what improved the likelihood of participants reporting appropriate assessment methods or tools based on the referral information they received. Whilst the model correctly classified 78.4% of the cases, it was found to not be a statistically significant model, (χ^2 (19, N = 139) = 24.59, p = 0.174, Nagelkerke R^2 = 24.6%). With the 'at risk' group removed, the results remained similar; the model continued to be non-significant (χ^2 (19, N =111) = 26.383, p = .120).

Overall, the sensitivity analysis indicated that the 'at risk' group of responses did not pose a substantial risk to the models utilised. As the preliminary results indicated no changes regarding significance of the models, no further analyses were conducted.

Chapter 7: Discussion and Critical Evaluation

Overview of chapter

Within this chapter, the main findings from meta-analysis and empirical research project, reported in Chapters 2 and 4 respectively, are summarised and brought together. The strengths and limitations of each paper will be explored within a critical evaluation, followed by ideas for future research and clinical implications. Reflections as a post graduate researcher completing this thesis portfolio has been included, which will be followed by an overall conclusion.

Main findings

Meta-analysis

The meta-analysis included within this portfolio, aimed to explore and identify the prevalence of psychological needs of children presenting with Fetal Alcohol Spectrum Disorder (FASD). For the purpose of this study, psychological needs encompassed mental health needs, behavioural needs and neurodevelopmental conditions. From the available research available, five overarching themes were created for which prevalence data were collected: depression, anxiety, behavioural difficulties, Autism Spectrum Disorder (ASD) and Attention Deficit Hyperactivity Disorder (ADHD). Moderator analyses were utilised to explore the effect of data collection type used within each study (active versus passive), and the location of the study (Canada, United States of America (USA), and Rest of the World (ROW)) on prevalence reported. Sensitivity analysis was also completed to account for any biases within the data.

Overall, 50 papers were included which yielded 125 prevalence rates across the different psychological needs. The weighted pooled prevalence of children with FASD presenting with each psychological need was as follows: depression = 13.3%, anxiety = 18.9%, behavioural difficulties = 22.8%, ADHD = 56.5%, and ASD = 14.8%. Moderator analysis found that for depression, behavioural difficulties and ADHD, active measures of data collection yielded higher prevalence rates than passive methods, whereas for anxiety and ASD, passive methods yielded higher prevalence rates. However, these results were not found to be statistically significant.

High rates of heterogeneity were found across the prevalence rates, which should be considered when interpreting the findings. Reasons for this include the different methods of data collection which were used in each study, the different methods of FASD classification and methodological differences.

Prior to the current study, several meta-analyses exploring co-occurring conditions had taken place. Due to the increase in research into the prevalence of FASD and the introduction of the NICE clinical standards, the current study sought to build on these previous results, by reporting updated

prevalence rates which may have been impacted by these factors. The current study also aimed to address some of the methodological issues faced by previous papers, such as the tendency for samples to be all populations from neurodevelopmental assessment clinics, by utilising broader inclusion criteria. This enabled papers to be included which did not report particular diagnoses, but rather needs which fell under the overarching themes.

The results from this study corroborated some findings from earlier meta-analyses, with similar prevalence rates found for ADHD (50.2%, Weyrauch et al., 2017; 52.9%, Lange et al., 2018) and depression (14.1%, Lange et al., 2018). Higher rates of ASD were found in the present study compared to previous research (2.6%, Lange et al., 2018). For the present study, behavioural difficulties were utilised as an umbrella term for studies reporting any behavioural need, thus the prevalence rate of 22.8% can be compared to the rates of Oppositional Defiant Disorder (ODD; 16.3%) reported by Weyrauch et al. (2017), and the rates of ODD (12.9%) and Conduct Disorder (7%) reported by Lange et al. (2018). Higher rates were also found for anxiety (18.9%) compared to previous reports (7.8%, Weyrauch et al., 2017). The results indicate that the needs of children with FASD can be widespread and are likely to contribute to secondary difficulties seen within the FASD population. FASD is documented to be misdiagnosed as ADHD, due to a similar neurodevelopmental profile, but also due to the stigma attached to FASD, thus the relationship between the two conditions continues to gather necessary research interest.

Empirical Paper

The empirical research study aimed to explore the clinical decision making of various clinicians involved in the assessment and diagnosis of FASD. This included Clinical Psychologists, Psychiatrists, Speech and Language Therapists, Occupational Therapists and Nurses. The study saw clinicians presented with one of two vignettes of a referral of a 12 year old girl presenting with neurodevelopmental needs. The only difference between the referrals was the inclusion of prenatal alcohol exposure (PAE) information, which was included in one of the referrals and formed the PAE condition. Overall, 139 clinicians read the vignettes and then answered a survey regarding their thoughts about possible diagnoses and assessment methods/tools.

The survey found that clinicians were most likely to consider ADHD as a diagnosis, with FASD being the fifth most common answer. FASD was statistically more likely to be considered when clinicians received PAE information within the referral, indicating its level of importance. Whilst Clinical Psychologists were most likely to consider FASD as a diagnosis, binary logistic regressions found that PAE information was the only significant predictor of an FASD diagnosis consideration.

Following this, clinicians self-rated their confidence, views and practice towards FASD on 5-point Likert scales. The findings from this survey reflected disparity between assessment and diagnosis; the majority of participants felt confident in identifying FASD, whereas the majority were not confident in selecting appropriate assessment methods/tools.

Portfolio in the context of literature base

The meta-analysis within this portfolio documented the needs of children with FASD, which include difficulties with mood and anxiety, behavioural issues, and further neurodevelopmental needs. These needs may be present because of the different systems that alcohol can affect within a developing fetus, or they could be secondary effects due to living with a disorder which is typically undiagnosed or misdiagnosed and therefore misunderstood. The empirical project found that despite new clinical standards around the assessment and diagnosis of FASD, clinicians are more likely to consider disorders such as ASD and ADHD before FASD, illustrating the difficulties of obtaining an FASD diagnosis. Barriers to this process were identified which included a perceived lack of adequate training (including sentinel facial features) and confidence in assessment methods. Supporting clinicians with these barriers may help to increase early access to FASD assessments, which in turn may help reduce the secondary effects experienced by children with FASD.

Both studies highlighted the co-morbidity and misdiagnosis of ADHD. The impact of being misdiagnosed with ADHD can be detrimental. For example, children with FASD are less responsive to classical ADHD medications such as methylphenidate (O'Malley et al., 2000). Thus misdiagnosis of ADHD can result in symptoms associated with ADHD not being effectively treated. Individuals with FASD are documented to experience similar symptoms to those diagnosed with ADHD, such as inattention, impulsivity and hyperactivity (Bhatara et al., 2006), so misdiagnosis could be seen as understandable, given the diagnostic ambiguity between the disorders. The overlap in symptoms poses the question whether ADHD is in fact co-occurring, or rather the symptoms are part of an FASD presentation (Rasmussen et al., 2010). Thus, research has been focused on how to differentiate between diagnoses to support with ambiguity. For example, another core deficit seen in both conditions is executive functioning, which encompasses broad domains such as working memory, response inhibition and set shifting (Miyake et al. 2000). A meta-analysis conducted by Khoury and Milligan (2016) found that children with FASD presented with significantly larger deficits within executive functioning when compared with children with ADHD, which were exacerbated by IQ; lower scores on cognitive functioning psychometrics were associated with more pronounced executive functioning deficits. Interestingly, differences in executive functioning are also seen in individuals with ASD, with much research suggesting a broad impairment which can look different for

each person (Demetriou et al., 2019). Given that 14.8% of children within the current meta-analysis were also diagnosed with ASD, research which aids clinicians to explore the nuances between how constructs such as executive functioning, present between different neurodevelopmental disorders, is imperative in in order to effectively support children with FASD. This would not only allow for earlier detection and diagnosis of FASD, but also for specialised support plans which are highly individualised, and person centred.

Awareness and stigma

The stigma surrounding FASD is well reported and has been discussed throughout this portfolio. This is also reported to be one of the reasons contributing to the underdiagnosis of FASD, due to women feeling unable to present to primary care with concerns for fear of societal judgement (British Medical Association Board of Science, 2016). Another reason which has been less discussed, is the overall awareness of FASD within the general population. The empirical study within this portfolio indicated that FASD is not a disorder that clinicians typically keep in mind when reviewing referrals. One question which could be posed is if FASD would be more routinely considered if individuals presented in services with specific concerns of FASD or alcohol use. This has been seen to be influential with ASD; one of the factors associated with the rising prevalence of ASD, is the increased awareness of individuals who then seen assessment (Matson & Kozlowski, 2011).

In order to develop public health strategies to increase awareness, assessing what is known by the public is important to consider beforehand. Mukherjee et al. (2014) explored the level of knowledge of FASD within the general public through questionnaires and focus groups. Whilst 86.7% had heard of FASD, the study found that there was a general lack of knowledge, suggesting people are less aware of the features of FASD. The study also found that 59.9% did not know the government guidelines regarding alcohol consumption, illustrating the need for clearer and more consistent messages. Overall, this suggests greater awareness within the general public is required.

Increasing awareness has been proven to support with increased assessment and diagnoses for other neurodevelopmental conditions. In more recent years, awareness campaigns have been implemented for FASD. Following recognition of FASD as a serious public health concern in the UK, Drymester, a digital campaign, was initiated. This project sought to identify how a marketing campaign using digital formats could help reduce alcohol exposed pregnancies (AEP). The campaign took place between May 2018 and March 2021 in Greater Manchester, and was successful in reaching and engaging the general public, with 53% responding positively to a survey about their thoughts towards the campaign (Reynolds et al., 2021). This was part of a larger project called the Greater Manchester AEP Programme; a project with six core aims categorised into Prevention,

Support and Knowledge. Overall, this campaign saw impressive progress within all three areas. Results from a post-campaign survey indicated differences between those who did and did not see the campaign, in relation to attitudes towards drinking whilst pregnant (Reynolds et al., 2021). For example, when presented with the statement "It's ok to occasionally drink a small amount of alcohol when pregnant", 20% of participants who saw the campaign agreed, compared to 30% of participants who did not see the campaign. Further to this, following the AEP programme, 18% of individuals identified as at risk of an AEP reduced their alcohol consumption (Greater Manchester Combined Authority, n.d.). The outcomes of this project suggest that campaigns which deliver unambiguous, clear messages, delivered in formats which are most accessible, have the power to shift the attitudes and behaviours of its recipients. Moreover, equipping individuals with the knowledge presented in this format, may support with individuals recognising the potential impact of their alcohol intake during pregnancy, and advocate for specific FASD assessments. Given that the empirical project within this thesis portfolio saw that clinical professionals are unlikely to consider FASD, having individuals present with specific FASD concerns may help professionals keep this information in mind when assessing, allowing for earlier diagnosis and support.

Despite these campaigns and changes regarding awareness of FASD, it remains a significant public health issue. Barker et al. (2011) wrote that awareness of the incidence and prevalence of disorders are no longer the most important factors when it comes to how people respond to disorder specific information. Instead, how society relates to particular disorders is most instrumental, which inevitably invites the issue of stigma back into the playing field. How to improve a society's relationship with disabilities can be started at a grass-roots level; education and inclusive attitudes towards FASD through schools has the capacity to impact on the surrounding stigma (Barker et al., 2011). Moreover, representation of FASD through channels which are most accessed and influential for our younger population will be instrumental in supporting children to feel seen. This is an area requiring expansion; Barker et al. (2011) explored the descriptions of characters in North American youth fiction novels. This systematic review found that only two books contained characters with a diagnosis or characteristics of FASD, compared to 14 books containing characters with ASD. More recently, Aspler et al. (2022) explored characters with neurodevelopmental disorders within television shows. This study found that that FASD is the least commonly represented, with only one character known to the authors knowledge. However, the character's storylines were mostly violent with "sociopathic tendencies", playing into stereotyped representations. Media, therefore, has a larger part to play in the representation of FASD to support with stigma, particularly when it is evidently beneficial in raising awareness for other neurodevelopmental conditions; for example, 41% of pre-service teachers reported to have first learned about ASD through television (Chansa-Kabali et

al., 2019). Further to this, as part of the AEP programme, an interactive performance targeted at young people called 'Birthday', saw 93% report that information learnt from the performance would inform future choices, which again highlights the success of campaigns through accessible media.

Taken together, all the documented research exploring awareness, stigma and the role of media, indicate that whilst FASD is underrepresented, when accessible formats of information delivery are utilised for specific purposes, they can be advantageous. Increasing awareness whilst reducing stigma, may support with individuals seeking FASD specific assessments, and thus support clinicians to bear this diagnosis in mind. This may protect against missed and misdiagnosis of other disorders, such as ADHD or conduct disorder (Chasnoff et al., 2015). Given that 18% of individuals within England and Wales belong to Black, Asian and other ethnic groups (Office for National Statistics, 2022), this is particularly significant for African American children, who are more likely to have neurodevelopmental concerns missed in favour of behavioural disorders (Baglivio et al., 2016). Further to this, earlier diagnosis, particularly before the age of six, has been identified as a protective factor against the development of secondary disabilities (Streissguth et al., 2004). This is due to enabling individuals to access support tailored to their specific needs, for example poor social skills and lower intelligence quotient (IQ) (Walthall et al., 2008). This may potentially impact on the rates of co-occurring psychological needs documented in the meta-analysis. Whilst all of the positive effects of awareness and stigma reduction are seen to be beneficial to individuals with FASD and their families, it should also be noted that earlier diagnosis, and thus reduced secondary disabilities, would also be financially beneficial to society.

Critical Evaluation

The studies within this portfolio were able to add to the growing literature base for FASD, which is required for a disorder which is thought to be more prevalent than ASD. The meta-analysis was able to utilise weighted pooled prevalence to gain overall estimates of psychological needs from 50 studies, incorporating 125 prevalence rates. The methodology sought to overcome weaknesses found in previous meta-analyses by broadening the inclusion criteria, to include studies where needs were reported (for example through self-report) as well as diagnoses. This is, to the author's knowledge, the first meta-analysis within the field of FASD to do this. The study also endeavoured to explore another weakness found in many research papers investigating FASD, the method of data collection. Much of the research utilises a retrospective case review methodology, possibly due to budget and time constraints. The meta-analysis explored whether this passive method of data collection yielded different prevalence rates compared to when new data was actively collected. As well as this, it was identified that many of the published studies included were completed in Canada

and the USA, which was then also explored within a moderator analysis. Moreover, sensitivity analysis explored whether the overall prevalence rates were impacted by the inclusion of studies rated as low quality. The efforts taken to explore the different ways prevalence rates could have been impacted on is seen as a strength for the meta-analysis.

The empirical paper was, to the researcher's knowledge, the first study to explore the decision-making process of clinicians reviewing referrals, within an FASD framework. Utilising a clinical case vignette methodology aimed to increase ecological validity, meaning that the findings are able to offer an insight into the decisions that would be made, as close to a real life setting as possible. With an aim to recruit a sample which was both representative as well as sufficient for the study to have sufficient power, multiple recruitment methods were used, again supporting with ecological validity. Finally, collaborating with National FASD enabled the study to capture the perspective of clinicians who work closely with individuals with FASD, allowing us to tailor the study in a meaningful way. It is hoped that by doing so, the study will be valuable and informative for this community.

However, there are some limitations of both studies that should be addressed. Firstly, the design of the empirical project unfortunately left it susceptible to survey bots, threatening the integrity of the data. Whilst great efforts were made to reduce the impact of the bots, documented in Chapter 5, there is still a possibility that the included data set includes bot responses, or responses made by 'real' participants were discarded. However, this is also a possibility in all survey studies, thus a strength can be found in the way that the problem was identified and managed.

In relation to this, the sample size could be considered small and there is a possibility that the study is underpowered. This means that smaller effects were not found. This could have affected investigations into how profession affects the decision making process, as there were many more Clinical Psychologists who participated compared to other groups, such as psychiatrists and social workers. In addition to this, there were many clinical professions who are instrumental within an FASD diagnosis pathway, which were unable to be included within this current study, due to the scope of the thesis portfolio. Clinicians from these professions may have had differing views in terms of their perceptions of training and confidence regarding FASD and may have responded differently towards the vignette. Due to these sample limitations, the results should be interpreted with caution.

The second part of the empirical study included a researcher-developed survey to explore the confidence, views and practice of clinicians in relation to FASD. This was created with input from National FASD, and allowed for the questions to be tailored to what was felt to be relevant to the researchers, as well as the FASD community. However, the use of a non-validated measure does

mean that results need to again be taken with caution, as there is a possibility that they were interpreted by clinicians in different ways, and thus measure different concepts. Furthermore, self-report response can result in demand characteristics, and Likert scales can be prone to particular response styles which can threaten the validity of the measure (Liu et al., 2017). Despite these limitations, the methodology was felt to be the most efficient option which placed the least burden on participants.

The meta-analysis included a comprehensive search of articles relating to the co-occurring conditions of children with FASD, across three databases. This identified 6759 articles, however, as no grey literature was explored, there may have been unpublished data which was not included. Secondly, many of the papers examined utilised *T*-scores from various psychometric measures to describe the psychological needs of children. Due to selecting a pooled prevalence methodology, many of the papers were discarded due to not providing the data required. The majority of the data included in the meta-analysis, came from the descriptions of the samples, rather than specific results from each study. This meant that the origin of the needs reported was not always known, reducing the quality of the studies. Therefore, whilst a pooled prevalence methodology is helpful to gain an overall picture, this did mean that due the current research base, the overall data set was smaller, of reduced quality, and not inclusive of the data which many studies are currently collecting.

Clinical Implications

Taken together, the studies included within this portfolio have a number of implications for clinical service delivery and practice. The meta-analysis highlighted the high prevalence rates for many psychological needs for children with FASD, indicating that screening for such difficulties should be facilitated as and when children with FASD present within services. Moreover, FASD is a lifelong disorder and as such the co-occurring difficulties a child may experience may change, as they encounter different challenges throughout their lives. Thus, this should be considered and entry into services as and when required should be made accessible for the FASD population.

Alongside this, both papers highlighted several training needs, indicating that greater emphasis is needed about FASD in training programmes, to ensure that clinicians feel supported. These training needs include understanding the similarities of FASD and other neurodevelopmental conditions, the co-occurring conditions mentioned within the meta-analysis, and how FASD can be assessed. Given some of the intricacies documented between FASD and ADHD such as greater executive functioning deficits (Khoury & Milligan, 2016), assessment training should also ensure that all 10 areas of an ND assessment are considered. As stated by the authors, exploring all areas would enable a more well-rounded picture of a child's abilities to problem solve and the difficulties they

may face, academically, socially and functionally. This in turn would enable tailor-made support plans to be created to give a child the best chance at success.

More training would also support with the well documented issue of FASD only being considered if sentinel facial features are present, which is 10% of the FASD population. Furthermore, training about the necessary and appropriate information required in referrals would support clinicians to consider all relevant diagnoses, as well as streamline the pathway as much as possible. For example, if referrers documented prenatal alcohol exposure information within their referral, even if this is unknown, this may support clinicians to hold this in mind and explore within their assessment. Finally, further training on how multi-disciplinary teams can work together to assess, diagnose, and support a young person with FASD would be beneficial. This would ensure that all clinicians know where referrals should be made to ensure that all areas of a full FASD assessment are covered.

Future Research

As one of the limitations of previous research is the use of retrospective data from clinically referred samples, future research should focus on identifying the needs of children with FASD who do not just present in services. This would help to find more generalisable findings. Further to this, collecting new information regarding children's needs would ensure that the findings are more reliable and current, rather than relying on previously collected data which may not be accurate at the time of the study. Given the impact an FASD diagnosis can have on the entire family system (Olsen et al., 2009), it would also be important to understand whether there are any differences in the prevalence of different psychological needs when reported from caregivers compared to self-reported needs from the child.

Moreover, whilst the empirical study indicated some interesting findings, the sample size and unbalanced clinician groups limit its conclusions. Thus, repeating this study with a larger sample, with more balanced profession groups and also representation from other clinicians important to the FASD assessment and diagnosis pathway, would be beneficial.

As well as this, the empirical study aimed to explore the likelihood of clinicians considering FASD, without being primed to do so. Some of the responses to the survey, did not corroborate with what was seen practically in the vignette experiment. For example, some clinicians reported that they are confident in identifying FASD, but did not consider this diagnosis when reviewing the referral. Future research exploring this discrepancy could be beneficial. For example, if clinicians are more able to consider FASD when they have been given information about FASD beforehand, or they

could choose from a list of diagnoses, this would suggest that methods which help to keep specific diagnoses in mind would be supportive.

Reflections

During the data collection phase for the meta-analysis, I was struck by how much data was available in some of the studies, but presented in a way which was not appropriate for the study. For example, lots of studies reported T-scores for scales reporting difficulties such as anxiety or attention problems. These scores are usually compared to a 'typically developing' sample to emphasis any differences. However, these are reported separately from the prevalence of anxiety disorders or ADHD. In a society by which access and treatment to services is mostly dependent on diagnoses, I wondered whether these individuals, despite having recognisable difficulties in a range of areas, were able to access support services without diagnoses. Moreover, I felt frustrated that I could not use the data presented, as it felt as though the voices of those individuals were not heard.

As a researcher, I am interested in conducting research that will add to the evidence base and help to support individuals who face daily difficulties, such as those affected by prenatal alcohol exposure. Therefore, I was excited for the empirical study to go live and to see the response number increase over time was encouraging. However, once the numbers started to increase drastically, scepticism set in and after discussions with supervisors, it was evident that the survey needed to close due to bot activity. I was disheartened by this, as I was doubtful that research which has been compromised in such a way would be considered for publication, which meant that I could not contribute to the research base and thus the FASD community. Upon researching survey bot activity and following the rigorous data cleaning process devised in Chapter 5 however, I became more optimistic about the future of the study. I was surprised to learn about how common these attacks are, and I was proud that I had been diligent enough to identify the problem and intervene when I did. It is likely that published research has been unknowingly comprised in this way previously; I was aware and able to create solutions which resulted in 'real' responses being saved, and thus the time and effort of clinicians who participated not being wasted. I am now hopeful that this important piece of research will be supportive for the FASD community.

Overall Conclusions

The meta-analysis covered in Chapter 2 outlined the weighted pooled prevalence estimates for a range of psychological needs including depression, anxiety, behavioural difficulties, ADHD and ASD, with the highest rates being identified for ADHD. Moderator analysis did not find any differences in terms of data collection methods or location of study.

ADHD was also most considered diagnosis within the empirical paper, with FASD being considered more when PAE information was included within the referral. Both studies highlight that greater awareness needs to be given to FASD, so training needs can be fulfilled, and the clinical standards can be brought into action. Together, these two studies contribute to the knowledge of FASD within children and the practice of clinicians who assess and diagnose.

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Appendices

Appendix A – Author guidelines for Alcohol: Clinical & Experimental Research

Alcohol, Clinical and Experimental Research is the official publication of the Research Society on Alcohol and the International Society for Biomedical Research on Alcoholism. It is an online, peer-reviewed, multidisciplinary journal that publishes original research that contributes substantially to our understanding of the etiology, treatment, and prevention of alcohol-related disorders. The journal accepts both full-length papers and critical reviews. Papers that the editors consider to be of cross-disciplinary interest and significance may be highlighted as Feature Articles. The journal also publishes editorial commentaries.

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Please indicate in the online system that the manuscript is to be considered a Critical Review and whether it was invited or not. The title, Abstract, and Introduction of your Critical Review should indicate that the manuscript is either a narrative or systematic review.

Clinical Trials All clinical trials to be reported in the journal must adhere to the principles of the Consolidated Standards of Reporting Trials (CONSORT), as embodied in the CONSORT Checklist (http://www.consort-statement.org/checklists/view/32-consort/66-title) and the CONSORT Flow Diagram (http://www.consort-statement.org/consort-statement/flow-diagram). For a clinical trial to be reviewed for publication in ACER, both the completed checklist and diagram must accompany the manuscript at the time of its initial submission. The diagram should be uploaded as Figure 1 during submission, and the checklist as an additional file. In addition, the title of your manuscript must indicate that the data being reported are part of a clinical trial.

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Structured abstract and key words: The article should be briefly summarized or abstracted in a short paragraph (approximately 300 words) at the beginning of the text on a separate page. It should contain 4 elements labelled: Background, Methods, Results, and Conclusions. At the end of the paragraph, provide no more than 5 key words or phrases.

Abbreviations used in the abstract and the text should be written out in full the first time they are used, followed by the abbreviation in parentheses (if they are used again), unless they are commonly used abbreviations. Citations should not be included in the abstract.

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Abbreviations: For a list of standard abbreviations, consult the Council of Biology Editors Style Guide or other standard sources. Write out the full term for each abbreviation at its first use unless it is a standard unit of measure.

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Citation in text: Cite references in the text by name and year in parentheses. Several publications by the same author or group should be listed in chronological order; those that appeared in the same year should be distinguished by a, b, c, etc. Where there are two authors, both should be named, but with three or more authors only the first author's name plus "et al." should be given.

Appendices: Appendices should be placed at the end of the main manuscript document, following the Reference List and preceding the Figure Legends (if any) and Tables (if any). As a guideline, appendices should be no longer than three manuscript pages. Appendices longer than that should be submitted as *Supporting Information*. In this case, please upload the Supporting Information document as a separate file.

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Each figure should be submitted individually - one figure per file.

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All manuscript revisions should be submitted online as soon as possible, but by the date stipulated in the decision letter. If more time is required to make revisions, please contact the Editorial Office for an extension. If you do not request an extension and your revisions are not submitted within the allotted time, your manuscript will be withdrawn by the Editorial Office. At that point, any further work by you will be considered as a new submission.

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Appendix B – Joanna Briggs Institute quality appraisal tools

Checklist for Prevalence Studies:

1) Was the sample frame appropriate to address the target population?
2) Were study participants sampled in an appropriate way?
3) Was the sample size adequate?
4) Were the study subjects and the setting described in detail?
5) Was the data analysis conducted with sufficient coverage of the identified sample?
6) Were valid methods used for the identification of the condition?
7) Was the condition measured in a standard, reliable way for all participants?
8) Was there appropriate statistical analysis?
9) Was the response rate adequate, and if not, was the low response rate managed appropriately?

Checklist for Case Series Studies:

1) Were there clear criteria for inclusion in the case series?
2) Was the condition measured in a standard, reliable way for all participants included in the case series?
3) Were valid methods used for identification of the condition for all participants included in the case series?
4) Did the case series have consecutive inclusion of participants?
5) Did the case series have complete inclusion of participants?
6) Was there clear reporting of the demographics of the participants in the study?
7) Was there clear reporting of clinical information of the participants?
8) Were the outcomes or follow up results of cases clearly reported?
9) Was there clear reporting of the presenting site(s)/clinic(s) demographic information?
10) Was statistical analysis appropriate?

Checklist for Analytical Cross Sectional Studies:

1) Were the criteria for inclusion in the sample clearly defined?
2) Were the study subjects and the setting described in detail?
3) Was the exposure measured in a valid and reliable way?
4) Were objective, standard criteria used for measurement of the condition?
5) Were confounding factors identified?
6) Were strategies to deal with confounding factors stated?
7) Were the outcomes measured in a valid and reliable way?
8) Was appropriate statistical analysis used?

Checklist for Cohort Studies:

1) Were the two groups similar and recruited from the same population?
2) Were the exposures measured similarly to assign people to both exposed and unexposed groups?
3) Was the exposure measured in a valid and reliable way?
4) Were confounding factors identified?
5) Were strategies to deal with confounding factors stated?
6) Were the groups/participants free of the outcome at the start of the study (or at the moment of exposure)?
7) Were the outcomes measured in a valid and reliable way?
8) Was the follow up time reported and sufficient to be long enough for outcomes to occur?
9) Was follow up complete, and if not, were the reasons to loss to follow up described and explored?
10) Were strategies to address incomplete follow up utilized?
11) Was appropriate statistical analysis used?

Appendix C - Included studies and extracted data included within meta-analysis

Study	Sample Size	Age (years)		% Female	Location	Method of FASD assessment	Psychological need			Data collection	Measure	Risk of bis
		Range	M(SD)				Type	N	%			
Depression												
Flannigan et al. (2019)	38	12.4-18.5	15.7 (NR)	69.21	Canada	4-Digit	Depression	0	0.00	Passive	File Review (NR)	Low
O'Conner et al. (2019)	54	13-18	15.69 (1.74)	55.60	USA	4-Digit, Hoyme et al. (2016)	Depressive disorder	20	37.04	Active	ChIPS	Low
Green et al. (2009)	89	8-15	10.7 (0.2)	56.81	Canada	Canadian guidelines (2015)	Depression	10	11.24	Passive	NR	Low
Connor et al. (2020)	199	2-31	10.5 (4.9)	33.20	Australia	4-Digit	Depression	11	5.53	Passive	NR	Low
Chasnoff et al. (2015)	156	NR	9.36* (NR)	36.20	USA	4-Digit	Depression NOS	12	7.69	Mixture	ND battery + clinical interview with psychologist	Low
Malisza et al. (2012)	23	6-13	9.2 (NR)	73.91	Canada	4-Digit	Depression	1	4.35	Passive	File review (NR)	Moderate
Greenbaum et al. (2009)	33	10-14	12.21 (1.63)	27.27	Canada	4-Digit	Depression	1	3.03	Passive	Parental report	Low
Rasmussen et al. (2013)	32	6-16	12.08 (NR)	56.20	Canada	4-Digit	Depression	2	6.25	Passive	File review (NR)	Low
Hayes et al. (2020)	163	5-17	11.24 (3.38)	39.26	Various	NR	Depression	20	12.27	Passive	File review: SDQ, demographic survey	Moderate

Study	Sample Size	Age (years)		% Female	Location	Method of FASD assessment	Psychological need			Data collection	Measure	Risk of bis
		Range	M(SD)				Type	N	%			
Kambeitz et al. (2019)	98	2-20*	8.62* (4.51)	NR	USA	FAS Diagnostic Checklist/Alcohol related neurodevelopmental disorder checklist	Depression	22	22.45	Passive	NR	Low
Paolozza et al. (2013)	27	8-16	12 (NR)	NR	Canada	Canadian guidelines (2015)	Depression	3	11.11	Passive	NR	Moderate
Paolozza et al. (2014)	72	NR	11.5 (3)	NR	Canada	Canadian guidelines (2015)	Depression	4	5.56	Passive	NR	Low
Rai et al. (2017)	52	9-16	13.2 (2.7)	44.20	Canada	4-Digit	Mood disorder	1	1.92	Mixture	Clinical evaluation and file review	Moderate
Palmeter et al. (2021)	9382	1-17	NR	NR	Canada	NR	Mood Disorder	3087	32.90	Passive	Parent report	High
Ipsiroglu et al. (2019)	40	1.8-17.5	9.4 (NR)	42.50	Canada	Canadian guidelines (2015)	Mood disorder	17	42.50	Passive	File review (NR)	High
Burns et al. (2021)‡	665	<18	10.6 (3.9)	29.70	Canada	Canadian guidelines (2015)	Mood disorder	86	12.93	Passive	ND battery (NR)	Moderate
Fagerlund et al. (2011)	73	8-21	13.5 (3.9)	60.27	Finland	IOM	Serious Depression	8	10.96	Active	CBCL	Low
Tsang et al. (2017)	18	NR	8.6 (0.6)	66.67	Australia	Canadian guidelines (2015)	Withdrawn /depressed	7	38.89	Active	CBCL	Low

Study	Sample Size	Age (years)		% Female	Location	Method of FASD assessment	Psychological need			Data collection	Measure	Risk of bis
		Range	M(SD)				Type	N	%			
Wells et al. (2012)	78	6-11	8.68 (NR)	32.05	USA	4-Digit	Mood disorder	14	17.95	Active	File review + medical, neurological, dysmorphology assessment	Moderate
Franklin et al. (2008)	44	5-10	NR	31.82	USA	4-Digit	Withdrawn / depressed	15	34.09	Passive	CBCL (syndrome scale)	Low
Agnihotri et al. (2019)	41	9-17	13.73 (2.04)	43.90	Canada	4-Digit	Mood disorder	1	2.44	Passive	Parent report/file review	Low
Burd et al. (2003)	303	.08-56*	8.2* (NR)	14	USA	FAS Diagnostic Checklist	Mood disorder	29	9.57	Passive	File review (NR)	Moderate
Anxiety												
Flannigan et al. (2019)	38	12.4-18.5	15.7 (NR)	26.3	Canada	4-Digit	Anxiety	0	0.00	Passive	file review (NR)	Low
Palmeter et al. (2021)	9382	1-17	NR	NR	Canada	NR	Anxiety	4156	44.30	Passive	Parent report (NR)	High
Oesterheld et al. (1998)	4	5 -11	8.25 (NR)	50	USA	Sokol and Clarren (1989)	Anxiety	1	25.00	Active	Conners (CPRS and CTRS), psychiatric interviews	Low
Green et al. (2009)	89	8 -15	10.7 (0.2)	45	Canada	Canadian guidelines (2015)	Anxiety	15	16.85	Passive	NR	Low

Study	Sample Size	Age (years)		% Female	Location	Method of FASD assessment	Psychological need			Data collection	Measure	Risk of bis
		Range	M(SD)				Type	N	%			
Chasnoff et al. (2015)	156	NR	9.36* (NR)	36.20	USA	4-Digit	Anxiety	15	9.62	Mixture	ND battery + clinical interview with psychologist	Low
Rai et al. (2017)	52	9-16	13.2 (2.7)	44.2	Canada	4-Digit	Anxiety disorder	1	1.92	Active	Clinical evaluation and file review	Moderate
Greenbaum et al. (2009)	33	6 -13	9.2	17	Canada	4-Digit	Anxiety Disorder	2	6.06	Passive	Parental report (NR)	Low
Paolozza et al. (2013)	27	8 -16	12 (NR)	14	Canada	Canadian guidelines (2015)	Anxiety	3	11.11	Passive	NR	Moderate
Rasmussen et al. (2013)	32	6 -16	12.08 (NR)	56.2	Canada	4-Digit	Anxiety	3	9.38	Passive	File review (NR)	Low
Kambeitz et al. (2019)	98	2-20*	8.62* (4.51)	NR	USA	FAS checklist/Alcohol related neurodevelopmental disorder checklist	Anxiety disorder	40	40.82	Passive	NR	Low
Connor et al. (2020)	199	2-31	10.5 (4.9)	33.20	Australia	4-Digit	Anxiety	50	25.13	Passive	NR	Low
Wells et al. (2012)	78	6 -11	8.68 (NR)	25	USA	4-Digit	Anxiety	5	6.41	Active	File review + medical, neurological, dysmorphology assessment	Moderate

Study	Sample Size	Age (years)		% Female	Location	Method of FASD assessment	Psychological need			Data collection	Measure	Risk of bis
		Range	M(SD)				Type	N	%			
Hayes et al. (2020)	163	5 -17	11.24 (3.38)	64	Various+	NR	Anxiety	71	43.56	Passive	SDQ, demographic survey	Moderate
Burns et al. (2021)‡	665	<18	10.6 (3.9)	29.70	Canada	Canadian guidelines (2015)/4-Digit	Anxiety disorder	87	13.08	Passive	ND battery (NR)	Moderate
Paolozza et al. (2014)	72	NR	11.5 (3)	NR	Canada	Canadian guidelines (2015)	Anxiety disorders	9	12.50	Passive	NR	Low
Mughal et al. (2020)	91	6 -15	9.69 (2.86)	37	UK	NR	Anxiety	67	73.63	Active	Spence Childrens Anxiety Scale	Low
Ipsiroglu et al. (2019)	40	1.8 - 17.5	9.4 (NR)	17	Canada	Canadian guidelines (2015)	Anxiety	23	57.50	Passive	File review (NR)	High
Tsang et al. (2017)	18	NR	8.6 (0.6)	12	Australia	Canadian guidelines (2015)	Anxiety problems	4	22.22	Active	CBCL	Low
Agnihotri et al. (2019)	41	9-17	13.73 (2.04)	18	Canada	4-Digit	GAD	3	7.32	Passive	Parent report/file review (NR)	Low
Fagerlund et al. (2011)	73	8-21	13.5 (3.9)	44	Finland	ION	Panic attacks	2	2.74	Active	CBCL	Low
Behavioural												
Flannigan et al. (2019)	38	12.4-18.5	15.7 (NR)	26.3	Canada	4-Digit	Conduct Disorder	21	55.26	Passive	File review (NR)	Low
Greenbaum et al. (2009)	33	6 -13	9.2 (NR)	17	Canada	4-Digit	Conduct Disorder	1	3.03	Passive	Parent report	Low

Study	Sample Size	Age (years)		% Female	Location	Method of FASD assessment	Psychological need			Data collection	Measure	Risk of bis
		Range	M(SD)				Type	N	%			
Malisza et al. (2012)	23	10 -14	12.21 (1.63)	9	Canada	Conservative diagnostic criteria	Conduct disorder/ODD	2	8.70	Passive	Parent report/file review (NR)	Moderate
Rai et al. (2017)	52	9 -16	13.2 (2.7)	44.2	Canada	4-Digit	Conduct Disorder	2	3.85	Active	Clinical evaluation and file review	Moderate
Burns et al. (2021)‡	665	<18	10.6 (3.9)	29.7	Canada	Canadian guidelines (2015)/4-Digit	Conduct Disorder	53	7.97	Passive	ND and clinical assessment (NR)	Moderate
Tsang et al. (2017)	18	NR	8.6 (0.6)	12	Australia	Canadian guidelines (2015)	Conduct	9	50.00	Active	CBCL	Low
Connor et al. (2020)	199	2-31	10.5 (4.9)	33.20	Australia	4-Digit	Conduct Disorder	13	6.53	Passive	NR	Low
Wells et al. (2012)	78	6-11	8.68 (NR)	25	USA	4-Digit	ODD	6	7.69	Active	File review + medical, neurological, dysmorphology assessment	Moderate
Landgren et al. (2010)	37	4.8-10.5*	7.5* (NR)	16	Sweden	IOM	Conduct disorder/ODD	15	40.54	Mixture	Interview, Asperger syndrome screening Questionnaire, WISC-3, Leiter revised rating scales (attention and activity), file review	Low

Study	Sample Size	Age (years)		% Female	Location	Method of FASD assessment	Psychological need			Data collection	Measure	Risk of bis
		Range	M(SD)				Type	N	%			
Oesterheld et al. (1998)	4	5-11	8.25 (NR)	2	USA	Sokol and Clarren (1989)	ODD	3	75.00	Active	Conners (CPRS and CTRS), psychiatric interviews	Low
Paolozza et al. (2013)	27	8 -16	12 (NR)	14	Canada	Canadian guidelines (2015)	ODD	4	14.81	Passive	File review (NR)	Moderate
Hayes et al. (2020)	163	5 -17	11.24 (3.38)	64	Various[+]	NR	ODD	51	31.29	Passive	Demographic survey	Moderate
Ipsiroglu et al. (2019)	40	1.8-17.5	9.4 (NR)	17	Canada	Canadian guidelines (2015)	ODD	8	20.00	Passive	File review (NR)	High
Rasmussen et al. (2013)	32	6 -16	12.08 (NR)	56.2	Canada	4-Digit	ODD	2	6.25	Passive	File review (NR)	Low
Paolozza et al. (2014)	72	NR	11.5 (3)	not stated	Canada	Canadian guidelines (2015)	ODD	10	13.89	Passive	File review (NR)	Low
Stromland et al. (2015)	16	2.5-12.8	6.19 (2.47)	9	Brazil	Hoyme et al. (2016)	Behaviour problems	1	6.25	Active	Wisconsin card sort, Rey complex figure, Raven coloured progressive matrices, interviews, Swanson, Nola and Pelham rating scale	High
Green et al. (2014)	52	4-12	NR	24 (46.2)	Canada	NR	ODD	19	36.54	Mixture	File review (NR)	High

Study	Sample Size	Age (years)		% Female	Location	Method of FASD assessment	Psychological need			Data collection	Measure	Risk of bis
		Range	M(SD)				Type	N	%			
Banerji et al. (2017)	49	0 -18	9 (NR)	14	Canada	Canadian guidelines (2015)	conduct disorder	39	79.59	Passive	File review (NR)	Low
Franklin et al. (2008)	44	5 -10	NR	14	USA	4-Digit	Conduct disorder/ODD	8	18.18	Passive	File review (NR)	Low
Breiner et al. (2013)	17	4-6	NR	NR	Canada	Canadian guidelines (2015)	ODD	15	88.24	Active	CBCL	High
Chasnoff et al. (2015)	156	NR	9.36* (NR)	36.20	USA	4-Digit	ODD	4	2.56	Passive	ND battery and clinical interview	Low
Agnihotri et al. (2019)	41	9-17	13.73 (2.04)	18	Canada	4-Digit	ODD	3	7.32	Passive	Parent report/ File review (NR)	Low
Burd et al. (2003)	303	.08-56*	8.2* (NR)	41*	USA	FAS diagnostic checklist	ODD	53	17.49	Passive	File review (NR)	Moderate
Green et al. (2009)	89	8-15	10.7 (0.2)	45	Canada	Canadian guidelines (2015)	ODD	19	21.35	Passive	NR	Low
ADHD												
Flannigan et al. (2019)	38	12.4 - 18.5	15.7 (NR)	26.3	Canada	4-Digit	ADHD	8	21.05	Passive	File review (NR)	Low
Herman et al. (2008)	36	6.3-16.5	10.7 (3)	17	USA	IOM	ADHD	18	50.00	Passive	File review (NR)	Low
Green et al. (2014)	52	4-12	NR	46.2	Canada	NR	ADHD	31	59.62	Mixture	File review (NR)	High

Study	Sample Size	Age (years)		% Female	Location	Method of FASD assessment	Psychological need			Data collection	Measure	Risk of bis
		Range	M(SD)				Type	N	%			
Agnihotri et al. (2019)	41	9-17	13.73 (2.04)	18	Canada	4-Digit	ADHD	26	63.41	Passive	Parent report/ File review (NR)	Low
Palmeter et al. (2021)	9382	1-17	NR	NR	Canada	NR	ADHD	6530	69.60	Passive	Parent report	High
Mukherjee et al. (2019)	97	6-26	78% under 14	40	UK	Canadian guidelines (2015)	ADHD	72	74.23	Passive	File review: DISCO, ND battery interviews	Low
Chen et al. (2012)	33	4.1-12.1	7.5 (2.2)	NR	USA	4-Digit	ADHD	25	75.76	Passive	Care giver report	Low
Williams et al. (2014)	31	5-18	11.5 (3.3)	39	Canada	Canadian guidelines (2015)	ADHD	27	87.10	Passive	NR	Moderate
Oesterheld et al. (1998)	4	5-11	8.25 (NR)	2	USA	Sokol and Clarren (1989)	ADHD	4	100.00	Active	Conners (CPRS and CTRS), psychiatric interviews	Low
Zhou et al. (2011)	20	6-30*	12.3* (6)	NR	Canada	4-Digit	ADHD	10	50.00	Passive	File review (NR)	Low
Yu et al. (2022)	26	NR	16.8 (0.7)	10	SA	Hoyme et al. (2016)	ADHD	11	42.31	Active	Clinical interview, DBDC	Low
Montag et al. (2022)	15	5-7	7.17 (8.6)	29	USA	Hoyme et al. (2016)	ADHD	1	6.67	Active	Neuro battery, including CBCL and TRF, maternal interview	Low

Study	Sample Size	Age (years)		% Female	Location	Method of FASD assessment	Psychological need			Data collection	Measure	Risk of bis
		Range	M(SD)				Type	N	%			
Hayes et al. (2020)	163	5-17	11.24 (3.38)	64	Various[+]	NR	ADHD	107	65.64	Passive	SDQ, demographic survey	Moderate
Paolozza et al. (2013)	27	8-16	12 (NR)	14	Canada	Canadian guidelines (2015)	ADHD	11	40.74	Passive	NR	Moderate
Malisza et al. (2012)	23	10-14	12.21 (1.63)	9	Canada	Conservative diagnostic criteria	ADHD	11	47.83	Passive	Parent report (NR)	Moderate
Rai et al. (2017)	52	9-16	13.2 (2.7)	44.2	Canada	4-Digit	ADHD	12	23.08	Active	Clinical evaluation and file review	Moderate
Lidstone et al. (2020)	17	7.4-17.6	12 (3)	11	USA	NR	ADHD	15	88.24	Active	Vanderbilt parent informant and health history questionnaire	Moderate
Geier & Geier. (2022)	321	0-12	DOB average 1997	47.98	USA	NR	ADD/ADHD	166	51.71	Passive	NR	Low
Rasmussen et al. (2013)	32	6-16	12.08 (NR)	56.2	Canada	4-Digit	ADHD	18	56.25	Passive	File review (NR)	Low
Mattson et al. (2013)	79	8-17	12.37 (2.83)	48.1	Various[•]	CIFASD criteria	ADHD	19	24.05	Active	Parent interviews and questionnaires, C-DISC-4	Low
Reid et al. (2017)	31	6-13	8.5 (1.71)	NR	Australia	4-Digit	ADHD	19	61.29	Passive	File review (NR)	High

Study	Sample Size	Age (years)		% Female	Location	Method of FASD assessment	Psychological need			Data collection	Measure	Risk of bis
		Range	M(SD)				Type	N	%			
Greenbaum et al. (2009)	33	6-13	9.2 (NR)	17	Canada	4-Digit	ADHD	20	60.61	Passive	Parent report	Low
Banerji et al. (2017)	49	0-18	9(NR)	14	Canada	Canadian guidelines (2015)	ADHD	21	42.86	Mixture	Psychology assessment (NR)	Low
Ipsiroglu et al. (2019)	40	1.8-17.5	9.4 (NR)	17	Canada	Canadian guidelines (2015)	ADHD/ADD	27	67.50	Passive	File review (NR)	High
Kooistra et al. (2011)	28	7-10	8.81 (1.25)	42.9	Canada	4-Digit	ADHD	27	96.43	Active	Diagnostic interview for Children and Adolescents -IV, Summary ADHD checklist, Conners	Low
Kooistra et al. (2010)	30	7-10	7.17 (1.2)	13	Canada	4-Digit	ADHD	29	96.67	Active	Diagnostic interview for Children and Adolescents -IV, Summary ADHD checklist, Conners	Low
Uecker et al. (1996)	15	NR	10.03 (2.33)	NR	USA	NR	ADHD	2	13.33	Passive	File review	Moderate
Webster et al. (2020)	15	3-13	6.4 (3.04)	20	Australia	4-Digit/Australian guide	ADHD	11	73.33	Passive	AUDIT-C, facial photographic analysis software,	Moderate

Study	Sample Size	Age (years)		% Female	Location	Method of FASD assessment	Psychological need			Data collection	Measure	Risk of bis
		Range	M(SD)				Type	N	%			
											psychometric battery	
Rasmussen et al. (2010)	52	4-17	8.8 (NR)		Canada	4-Digit	ADHD	33	63.46	Passive	Conners and continuous performance test	Low
Elgen et al. (2007)	47	NR	7.67¥ (NR)	15	Norway	CDC	ADHD	42	89.36	Active	NR	Moderate
Paolozza et al. (2014)	72	NR	11.5 (3)	NR	Canada	Canadian guidelines (2015)	ADHD	45	62.50	Passive	NR	Low
Lange et al. (2019)	21	7.9-11	9.7 (1.0)	10	Canada	Canadian guidelines (2015)	ADHD	5	23.81	Passive	File review: ND assessment, CBCL	Low
Green et al. (2009)	89	8-15	10.7 (0.2)	45	Canada	Canadian guidelines (2015)	ADHD/ADD	53	59.55	Passive	NR	High
Wells et al. (2012)	78	6-11	8.68 (NR)	25	USA	4-Digit	ADHD	55	70.51	Active	File review + medical, neurological, dysmorphology assessment	Moderate
Stromland et al. (2015)	16	2.5-12.8	6.19 (2.47)	9	Brazil	Hoyme et al. (2016)	ADHD	6	37.50	Active	Wisconsin card sort, Rey complex figure, Raven coloured progressive matrices,	High

Study	Sample Size	Age (years)		% Female	Location	Method of FASD assessment	Psychological need			Data collection	Measure	Risk of bis
		Range	M(SD)				Type	N	%			
											structured questionnaires, Swanson, Nola and Pelham rating scale	
Connor et al. (2020)	199	2-31	10.5 (4.9)	33.20%	Australia	4-Digit	ADHD	83	41.71	Passive	File review (NR)	Low
Kambeitz et al. (2019)	98	2-20*	8.62* (4.51)	NR	USA	FAS checklist/Alcohol related neurodevelopmental disorder checklist	ADHD	84	85.71	Passive	NR	Low
Chasnoff et al. (2015)	156	NR	9.36* (NR)	36.20	USA	4-Digit	Attention deficit problems	88	56.41	Mixture	ND battery (NR) and clinical interview	Low
Greenbaum et al. (2002)	28	4-18	8.81(NR)	42.90	Canada	Non-validated ARND behaviour checklist for ARND	Attention deficits	9	32.14	Mixture	Caregiver questionnaires for ADHD	Low
Lane et al. (2014)	14	7-12	11.73 (1.36)	9	Canada	Canadian guidelines (2015)	ADHD	10	71.43	Active	Conners	Low
Lane et al. (2014)	14	7-12	11.73 (1.36)	9	Canada	Canadian guidelines (2015)	ADHD	9	64.29	Passive	File review (NR)	Low
Fagerlund et al. (2011)	73	8-21	13.5 (3.9)	44	Finland	ION	ADHD	44	60.27	Active	CBCL	Low

Study	Sample Size	Age (years)		% Female	Location	Method of FASD assessment	Psychological need			Data collection	Measure	Risk of bis
		Range	M(SD)				Type	N	%			
Okulicz-kozaryn et al. (2017)	50	7-9	NR	20	Poland	Canadian guidelines (2015)	ADHD	19	38.00	Active	NST plus new non-validated measure for teachers	Low
Tsang et al. (2017)	18	NR	8.6 (0.6)	12	Australia	Canadian guidelines (2015)	ADH problems	4	22.22	Active	CBCL	Low
Burd et al. (2003)	303	0.8-56*	8.2* (NR)	41*	USA	FAS diagnostic checklist	ADHD	119	39.27	Passive	File review (NR)	Moderate
Landgren et al. (2010)	37	4.8-10.5*	7.5* (NR)	16	Sweden	IOM	ADHD	21	56.76	Active	Structured interviews, Asperger Syndrome Screening questionnaire, WISC-3, Leiter revised rating scales.	Low
Franklin et al. (2008)	44	5-10	NR	14	USA	4-Digit	ADHD	23	52.27	Passive	File review (NR)	Low
ASD												
Montag et al. (2022)	15	5-7	7.17 (8.6)	29	USA	Hoyme et al. (2016)	ASD	10	66.67	Unclear	ND battery: CBCL and TRF, maternal interview	Low
Ipsiroglu et al. (2019)	40	1.8-17.5	9.4 (NR)	17	Canada	Canadian guidelines (2015)	ASD	2	5.00	Passive	File review (NR)	High

Study	Sample Size	Age (years)		% Female	Location	Method of FASD assessment	Psychological need			Data collection	Measure	Risk of bis
		Range	M(SD)				Type	N	%			
Geier & Geier (2022)	321	0-12	DOB average 1997	47.98	USA	NR	ASD	22	6.85	Passive	File review (NR)	Low
Burns et al. (2021)‡	665	<18	10.6 (3.9)	29.70	Canada	Canadian guidelines (2015)/4-Digit	ASD	27	4.06	Passive	Fil review: ND assessment (NR), clinical assessment	Moderate
Lange et al. (2019)	21	7.9-11	9.7 (1.0)	10	Canada	Canadian guidelines (2015)	ASD	3	14.29	Passive	File review: ND assessment, CBCL	Low
Hayes et al. (2020)	163	5-17	11.24 (3.38)	64	Various+	NR	ASD	37	22.70	Passive	File review: SDQ, demographic survey	Moderate
Reid et al. (2017)	31	6-13	8.5 (1.71)	NR	Australia	4-Digit	ASD	4	12.90	Passive	File review, CBCL	High
Kambeitz et al. (2019)	98	2-20*	8.62* (4.51)	NR	USA	FAS checklist/Alcohol related neurodevelopmental disorder checklist	Autism	4	4.08	Passive	NR	Low
Webster et al. (2020)	15	3-13	6.4 (3.04)	20	Australia	4-Digit/Australian guide	ASD	1	6.67	Passive	AUDIT-C, facial photographic analysis software, psychometric battery	Moderate

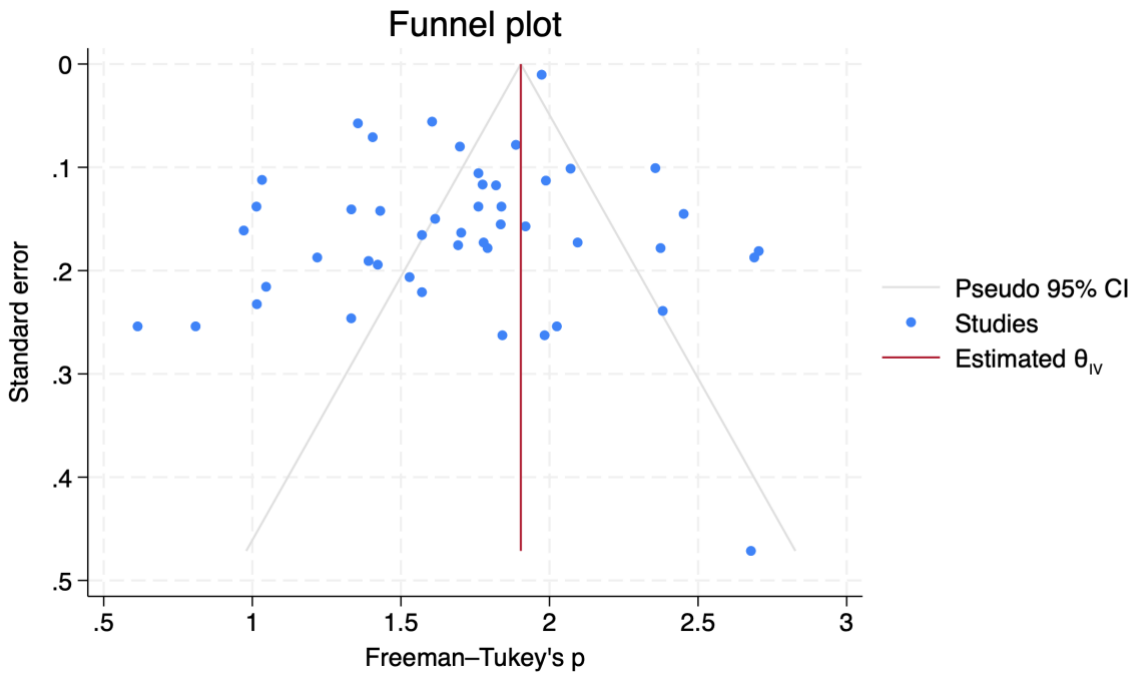
Study	Sample Size	Age (years)		% Female	Location	Method of FASD assessment	Psychological need			Data collection	Measure	Risk of bis
		Range	M(SD)				Type	N	%			
Chasnoff et al. (2015)	156	NR	9.36* (NR)	36.20	USA	4-Digit	ASD	8	5.13	Passive	File review: ND battery (NR) and clinical interview	Low
Mukherjee et al. (2019)	91	6-26	78% under 14)	40	UK	Canadian guidelines (2015)	ASD	62	68.13	Passive	File review: DISCO, ND battery (NR) interviews	Low
Landgren et al. (2010)	37	4.8-10.5*	7.5* (NR)	16	Sweden	1996 IOM criteria	Autism	2	5.41	Active	Structured interviews, Asperger Syndrome Screening questionnaire, WISC-3, Leiter revised rating scales.	Low

NR= Not reported. ChiPS = Children's Interview for Psychiatric Syndromes. ND = Neurodevelopmental. SDQ = Strengths and Difficulties questionnaire. CBCL = Child Behaviour Checklist. CPRS = Conners' Parent Rating Scales. CTRS = Conners' Teacher Rating Scale. WISC-3 = Wechsler Intelligence Scale for Children-III. DISCO = Diagnostic Interview for Social and Communication Disorders. DBDC = Disruptive Behaviours Disorders Checklist. TRF = Teacher Rated Form. C-DISC = Computerised Diagnostic Interview Schedule for Children-IV. AUDIT-C = Alcohol Use Disorders Identification Test for Consumption. 4-Digit = 4-Digit Diagnostic Code. IOM = Institute of Medicine. CIFASD = Collaborative Initiative on Fetal Alcohol Spectrum Disorder. Australian guide =. CDC = Centers for Disease Control and Prevention. FAS = Fetal Alcohol Syndrome. ADHD = Attention Deficit Hyperactivity Disorder. ADD = Attention Deficit Disorder. ASD = Autism Spectrum Disorder. USA = United States of America. UK = United Kingdom. SA = South Africa

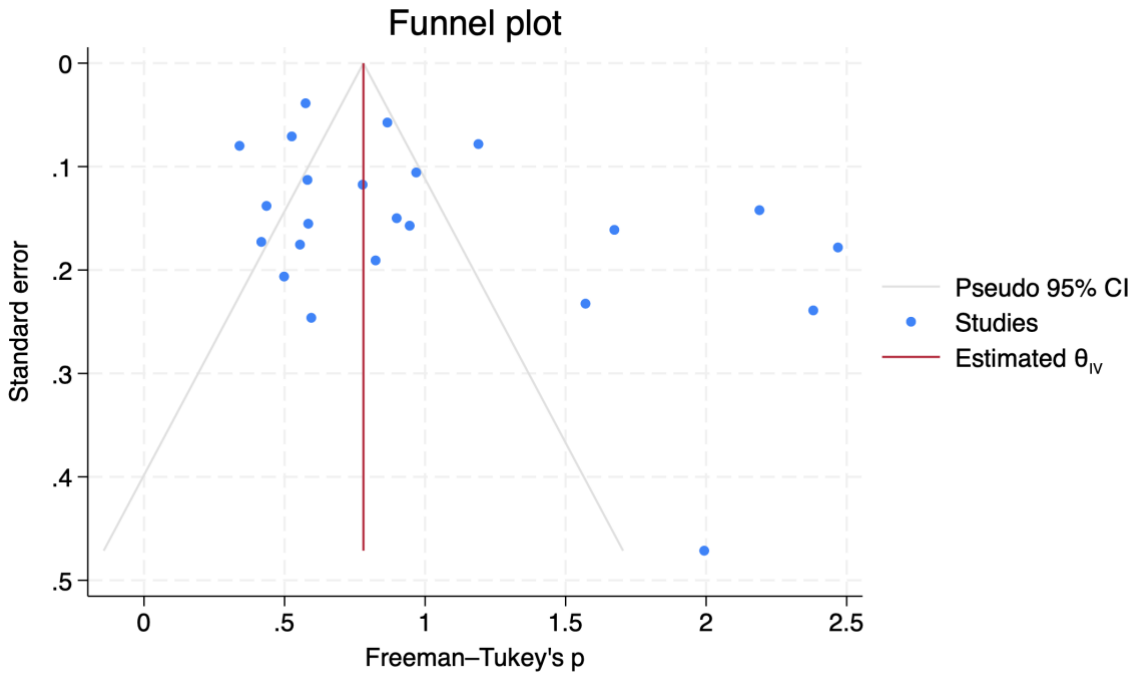
***** Number/percentage reported for whole sample including non-FASD participants. ‡National database study incorporating data from 26 clinics. ⁺Online survey with responses from Australia, USA, New Zealand, Canada, UK and South Africa. •Children recruited from various centres (USA, India, South Africa, Russia, Finland. ¥Number reported for whole sample including those not assessed for co-occurring needs.

Appendix D – Funnel plots (meta-analysis)

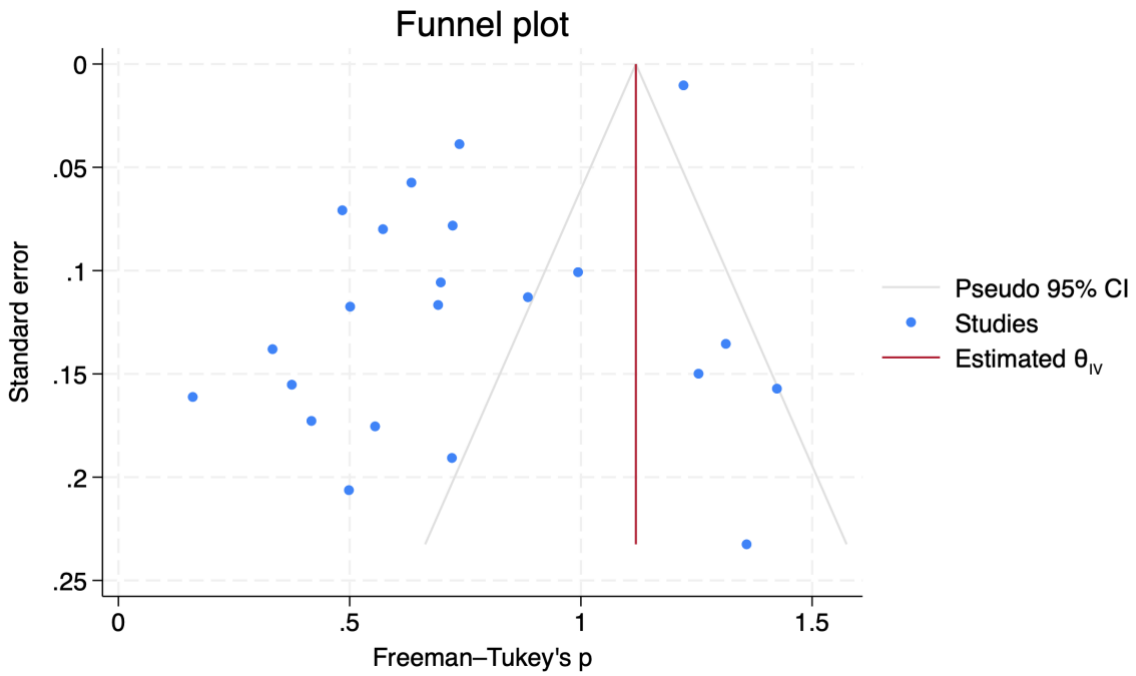
Assessing for publication bias for Attention Deficit Hyperactivity Disorder prevalence data:



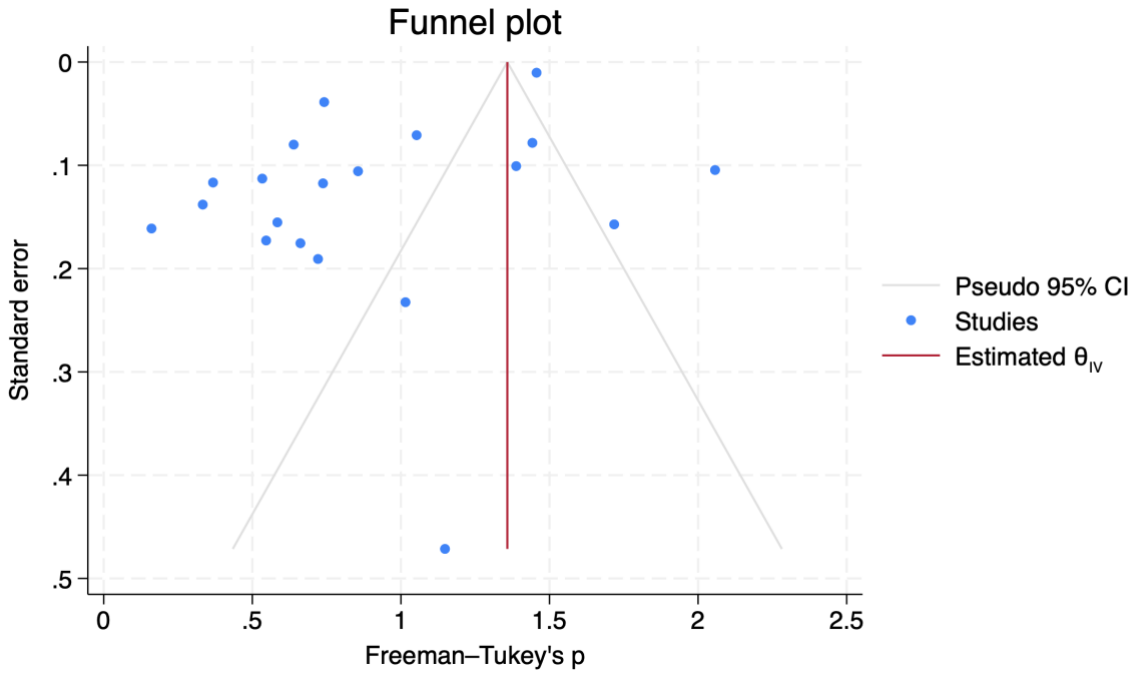
Assessing for publication bias for Behavioural prevalence data:



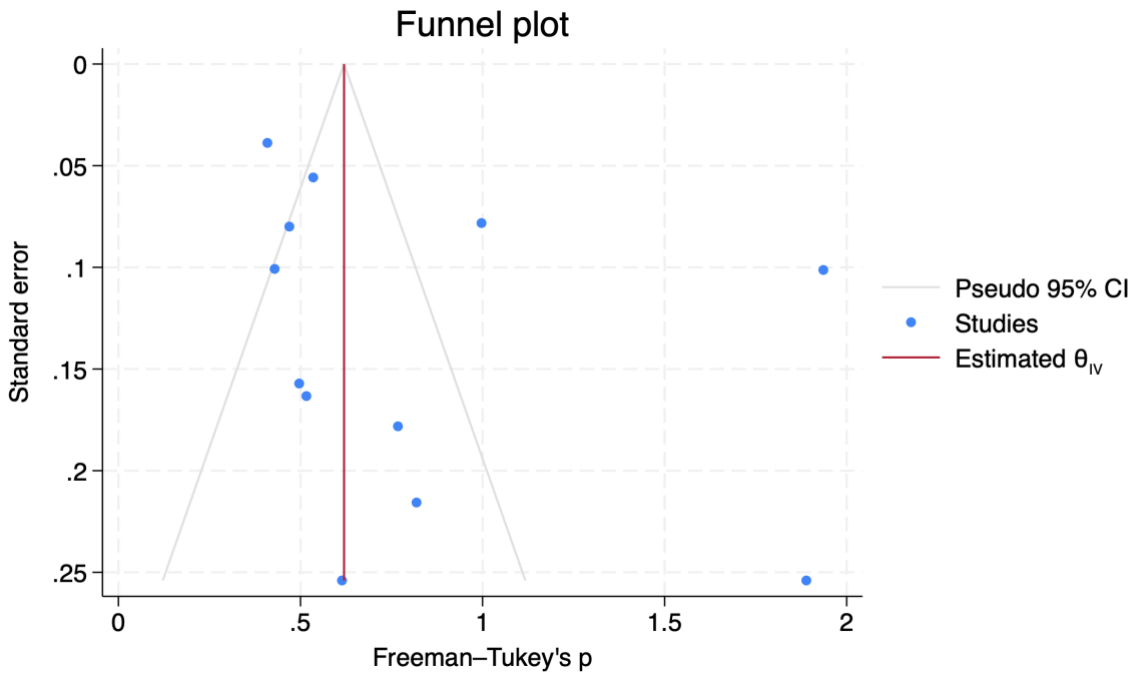
Assessing for publication bias for Depression prevalence data:



Assessing for publication bias for Anxiety prevalence data:



Assessing for publication bias for Autism Spectrum Disorder prevalence data:



Appendix E - Demographic and Employment Information survey

Please select your age from the following categories:

- 21-30
- 31-40
- 41-50
- 51-60
- 61-70
- 70+
-

What gender do you identify as?

_____

What is the highest level of education you have completed?

o GCSE/BTEC Levels 1-2/NVQ Level 1-2 or equivalent

o A Levels/BTEC Level 3/NVQ Level 3 or equivalent

o Certificate of Higher Education/BTEC Professional Diplomas/NVQ Level 4

o Foundation Degree/Diploma of Higher Education/HND

o Bachelor's Degree/PGCE

o Master's Degree/Postgraduate certificate or diploma

o Doctoral Degree

What is your current profession?

- Clinical Psychologist
- Psychiatrist
- Speech and Language Therapist
- Occupational Therapist
- Social worker
- Nurse/Mental Health Practitioner
- Other (please specify) _____

How many years of experience do you have working in child neurodevelopmental services?

- less than 1
- 1-5
- 6-10
- 11-15
- 16+

Appendix F - Case vignettes

Vignette a – mention of prenatal alcohol exposure

Dear x

Thank you for seeing this patient. Patient x is a 12-year-old female who has difficulties managing her emotions. X can become very upset when she is asked to do something which can lead to outbursts. There are also concerns about her safety, with reports that X doesn't appear to think before she acts. It is reported that she struggles with the concept of time and the reporting of information. X can also misread social situations and can often appear younger than she is which can place her in vulnerable situations.

X is also struggling at school, with her teachers stating that she struggles to follow instructions and often acts impulsively. She particularly dislikes maths. As a result, she is receiving detention often, but this doesn't appear to deter her behaviour. She benefits from support to complete the work and

can struggle to keep on task. School also note a difficulty with moving on from one task to the next, which again can lead to X becoming upset very quickly.

X was reported to be delivered naturally with no complications during the birth, although birth weight was in the low range. Some alcohol was reported to be consumed during pregnancy. X's family appear supportive.
Your guidance on further assessment and possible diagnosis to support this young person would be greatly appreciated.

Vignette b – no mention of prenatal alcohol exposure

Dear x
Thank you for seeing this patient. Patient x is a 12-year-old female who has difficulties managing her emotions. X can become very upset when she is asked to do something which can lead to outbursts. There are also concerns about her safety, with reports that X doesn't appear to think before she acts. It is reported that she struggles with the concept of time and the reporting of information. X can also misread social situations and can often appear younger than she is which can place her in vulnerable situations.
X is also struggling at school, with her teachers stating that she struggles to follow instructions and often acts impulsively. She particularly dislikes maths. As a result, she is receiving detention often, but this doesn't appear to deter her behaviour. She benefits from support to complete the work and can struggle to keep on task. School also note a difficulty with moving on from one task to the next, which again can lead to X becoming upset very quickly.
X was reported to be delivered naturally with no complications during the birth, although birth weight was in the low range. X's family appear supportive.

Your guidance on further assessment and possible diagnosis to support this young person would be greatly appreciated.

Appendix G - Clinical decision survey

From reading the case vignette, what potential diagnosis/diagnoses come to mind that you might consider and wish to further assess for?

_____

What would your full assessment include?

Appendix H - Confidence, practice and views survey

The National Institute of Clinical Excellence (NICE) have recently introduced new quality standards about the assessment, diagnosis and treatment for Fetal Alcohol Spectrum Disorder (FASD). FASD is a neurodevelopmental disorder caused by prenatal exposure to alcohol. Those affected can present with physical, behavioural, cognitive and emotional difficulties. The standards set by NICE recommend that any child with significant difficulties in at least three neurodevelopmental domains, plus probable prenatal alcohol exposure is referred for assessment. Children with confirmed alcohol exposure or all three facial features associated with prenatal alcohol exposure should also be referred for an assessment.

This study is specifically interested in clinician's views and understanding of FASD. We invite you to complete the following questions about factors which may act as facilitators or barriers to clinicians considering FASD as a potential diagnosis. If you would like to exit the study, your data up to this point will not be stored.

Please read the statements below and indicate how much you agree/disagree with them by using the scale.

- *I am confident in my ability to recognise FASD*

1-Strongly disagree, 2-Disagree, 3-neutral, 4-Agree, 5-Strongly agree

- *I am confident in my ability to select appropriate assessments when querying FASD*

1-Strongly disagree, 2-Disagree, 3-neutral, 4-Agree, 5-Strongly agree

- *I received adequate teaching about FASD during my training*

1-Strongly disagree, 2-Disagree, 3-neutral, 4-Agree, 5-Strongly agree

- *I have received further training about FASD since qualifying*

1-Strongly disagree, 2-Disagree, 3-neutral, 4-Agree, 5-Strongly agree

- *I am confident in asking about alcohol intake during pregnancy*

1-Strongly disagree, 2-Disagree, 3-neutral, 4-Agree, 5-Strongly agree

- *I regularly ask about alcohol intake during assessments*

1-Strongly disagree, 2-Disagree, 3-neutral, 4-Agree, 5-Strongly agree

- *I would only ask about alcohol intake if a child presents with the associated facial features*

1-Strongly disagree, 2-Disagree, 3-neutral, 4-Agree, 5-Strongly agree

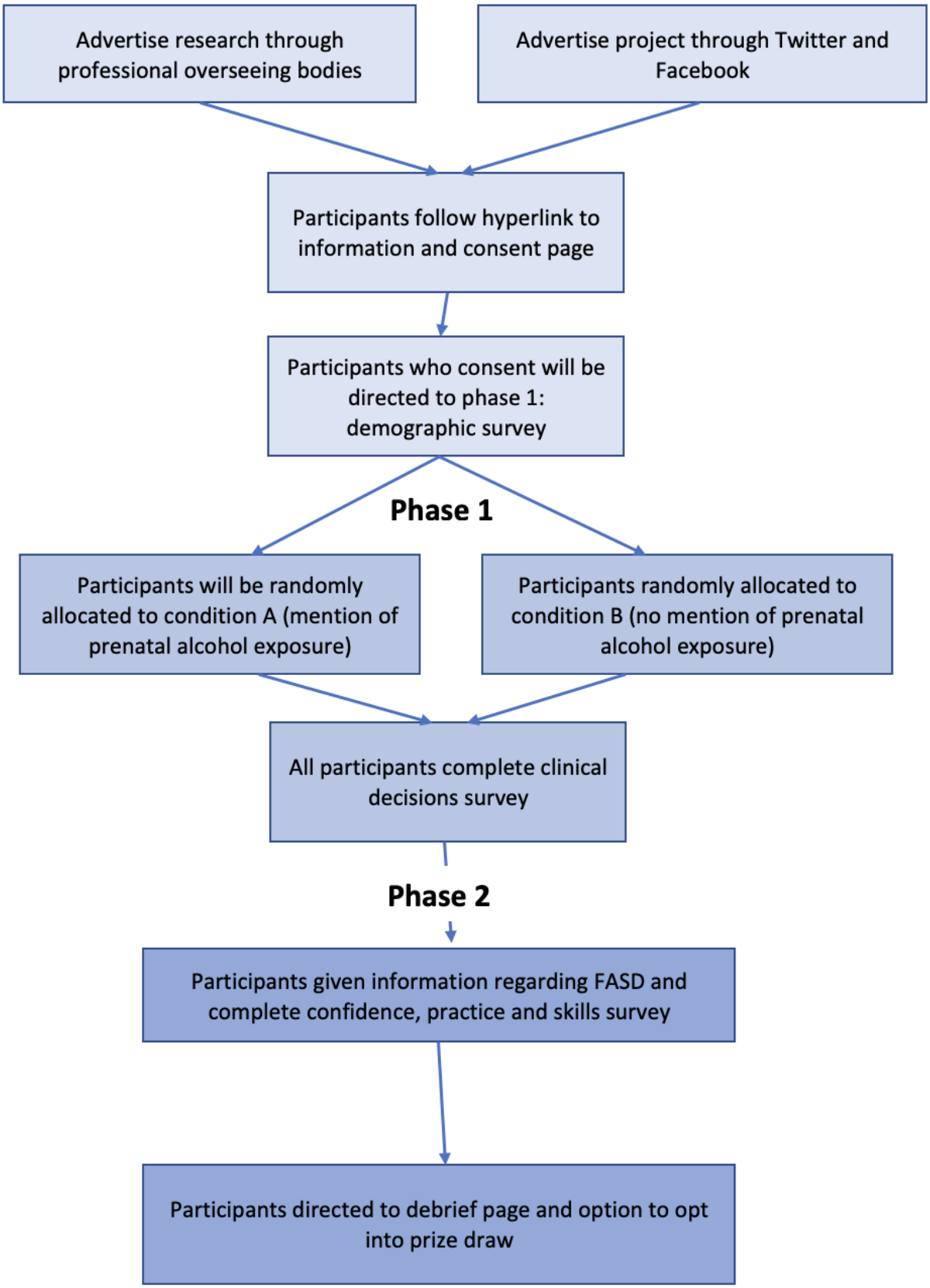
- *I would only consider FASD as a potential diagnosis if prenatal alcohol exposure is mentioned in the referral*

1-Strongly disagree, 2-Disagree, 3-neutral, 4-Agree, 5-Strongly agree

- *I believe there is still merit in diagnosing a condition/disorder for which there is no cure*

1-Strongly disagree, 2-Disagree, 3-neutral, 4-Agree, 5-Strongly agree

Appendix I - Flowchart of procedure



Appendix J - Participant Information Sheet

Participant Information Sheet

Norwich Medical School
Postgraduate Research Office 2.30
Elizabeth Fry Building
University of East Anglia
Norwich Research Park
Norwich
NR4 7TJ
Email: clinpsyd@uea.ac.uk
Tel: +44 (0) 1603 593076
Fax: +44 (0) 1603 591132

Title of Project: Exploring the decision making of clinicians working in neurodevelopmental child services, when considering potential assessments and diagnoses based on referral information, using a hybrid case vignette and survey methodology

Name of Researcher: Emma Heathcote
Primary Supervisor: Dr Aaron Burgess
Secondary Supervisor: Dr Kenny Chiu

We are researchers at the University of East Anglia and we are inviting you to particate in a study exploring what helps and what hinders clinicians in neurodevelopmental services being able to further assess and potentially diagnose adolescents they are referred. This research study is being conducted as part of a Doctoral Programme in Clinical Psychology, at the University of East Anglia. Before continuing, please read this page carefully to help you decide whether you would like to take part. If there is anything you would like to discuss, please contact Emma Heathcote via Emma.Heathcote@uea.ac.uk.

What is the study about?

Children in neurodevelopmental services can present with a wide array of different conditions. Referrals that are sent to clinicians within these services can vary in the amount and quality of

information they contain, on which the clinician has to decide what, if any, further assessment is needed to support a hypothesised diagnosis.
To determine the training needs within the whole neurodevelopmental pathway, it is important to establish what helps a clinician within this decision-making process, and what makes it more difficult.

Why have I been invited?
You have been invited to take part because you are a clinician working in a neurodevelopmental service.

Do I have to take part?
No, you do not have to take part in this study.

What will the study involve?
If you would like to take part in the study, you will be invited to complete a survey asking some demographic style questions first, for example the discipline of work. This is so we can see if particular factors influence how confident clinicians feel in making their decisions. You will then be invited to read a case vignette of a referral letter, and then to complete two surveys following this, based on what your initial ideas around potential and diagnoses are.

What will happen if I decide not to carry on with the study?
That is fine, no data will be stored, and you will not be affected.

What are the potential disadvantages of taking part?
The whole study will take approximately 20 minutes of your time to complete. There are no known risks to taking part in this study.

What are the potential benefits of taking part?
Whilst there are no guaranteed benefits to taking part, it is hoped that your participation will support us to identify the current training needs within the neurodevelopmental pathway to support clinicians in the assessment and diagnosis process.

Will the data provided by myself be kept confidential?

All privacy laws and procedures will be followed during every stage of this study. All of the information you enter will be kept confidential, anonymised and safe. You will not be required to enter your name to take part in the study.

Electronic data will be stored on a UEA approved password protected, and encrypted memory stick and an encrypted UEA server.

At the end of the study, there will be an opportunity for you to take part in a raffle for an Amazon voucher. Should you wish to do so, we will require your email address in order to send the voucher to you. However, your email address will not be linked to the information you have entered in any of the surveys.

Who has reviewed the study?

This study has been reviewed and approved by the University of East Anglia Faculty of Medicine and Health Sciences Research Ethics Committee. Research Ethics Reference:

What will happen to the results of this study?

The results from the study may be published into an academic journal. You will not be identifiable.

What if there is a problem?

If you have any concerns about any part of the study, you can contact the research supervisor Dr Aaron Burgess via Aaron.Burgess@uea.ac.uk. If you have any further problems or complaints then you are welcome to contact Professor Sian Coker, Director of the Doctorate of Clinical Psychology Programme via S.Coker@uea.ac.uk

What happens next?

If you decide to take part in the study, you will need to read the statement below. If you are happy to proceed, then you can provide your consent to participate by clicking the hyperlink which will direct you to the study.

How do I find out more?

You are welcome to contact Emma Heathcote, Trainee Clinical Psychologist via email at Emma.Heathcote@uea.ac.uk, if you would like further information.

Please note: This project has been ethically approved by the University of East Anglia. Please do not mail onto your colleagues in your service.

Thank you for reading this information.

Appendix K – Recruitment poster

Appendix L – Recruitment email to universities

Hello,

I am currently a trainee completing my doctorate in Clinical Psychology at the University of East Anglia. As part of this, I am researching how clinicians make decisions about assessment and diagnosis, when they receive referrals into neurodevelopmental services. From this research, we hope to advocate for the training professionals require in order to support them with this process.

For this, I am looking to recruit clinical professional from a range of disciplines, including **clinical psychology, psychiatry, occupational therapy, speech and language therapy, nursing and social work**. I am currently advertising my research project through social media, but I was wondering and hoping whether the University of ____ could support me by forwarding this email on

to the relevant departments to aid with recruitment? Meeting my minimum number of participants would help with publishing my research and ultimately supporting these clinicians and service users in the future, so any help would be much appreciated!

Thank you for taking time to read this email and I look forward to hearing from you

Many thanks
Emma
Trainee Clinical Psychologist
University of East Anglia

Appendix M – Recruitment email to regulation bodies

Dear (professional body)

My name is Emma Heathcote and I am a Trainee Clinical Psychologist completing my Doctorate in Clinical Psychology, at the University of East Anglia.

As part of my course, I am completing a project regarding the decisions that clinicians make in relation to assessment and diagnosis, when they receive referrals within neurodevelopmental services. For this, I am hoping to recruit clinicians currently working within neurodevelopmental services who are registered professionals within (Clinical Psychology, Psychiatry, Speech and Language Therapy, Occupational Therapy, Nursing and Social Care). This study has been ethically approved via the UEA's ethics committee (ETH2223-0111).

I have attached a participant information sheet and a project advert for further information. I was wondering whether the (body) would be so kind to advertise this project through its streams of communication with its members? This would support with recruitment and potential contributions to this field of research. Any support would be greatly appreciated.

Thank you for taking the time to read this email, please do not hesitate to contact myself for further information.

Yours sincerely,
Emma Heathcote

Appendix N – Ethical Approval from University of East Anglia's Ethics Committee

University of East Anglia
Norwich Research Park
Norwich. NR4 7TJ
Email:
ethicsmonitor@uea.ac.uk
Web: www.uea.ac.uk

Study title: Exploring decisions made by clinicians, when assessing and diagnosing in neurodevelopmental child services.

Application ID: ETH2223-0111

Dear Emma,

Your application was considered on 30th January 2023 by the FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee).

The decision is: approved.

You are therefore able to start your project subject to any other necessary approvals being given.

If your study involves NHS staff and facilities, you will require Health Research Authority (HRA) governance approval before you can start this project (even though you did not require NHS-REC ethics approval). Please consult the HRA webpage about the application required, which is submitted through the IRAS system.

This approval will expire on 1st December 2023.

Please note that your project is granted ethics approval only for the length of time identified above. Any extension to a project must obtain ethics approval by the FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee)

before continuing.

It is a requirement of this ethics approval that you should report any adverse events which occur during your project to the FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee) as soon as possible. An adverse event is one
which was not anticipated in the research design, and which could potentially cause risk or harm to the participants or the researcher, or which reveals potential risks in the treatment under evaluation. For research involving animals, it may be the unintended death of an animal after trapping or carrying out a procedure.

Any amendments to your submitted project in terms of design, sample, data collection, focus etc. should be notified to the FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee) in advance to ensure ethical compliance. If the amendments are substantial a new application may be required.

Approval by the FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee) should not be taken as evidence that your study is compliant with the UK General Data Protection Regulation (UK GDPR) and the Data Protection Act
2018. If you need guidance on how to make your study UK GDPR compliant, please contact the UEA Data Protection Officer (dataprotection@uea.ac.uk).

Please can you send your report once your project is completed to the FMH S-REC (fmh.ethics@uea.ac.uk).

I would like to wish you every success with your project.

On behalf of the FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee)

Yours sincerely,

Dr Paul Linsley

Appendix O - Participant Consent Statement

Statement of Consent

Norwich Medical School
Postgraduate Research Office 2.30
Elizabeth Fry Building
University of East Anglia
Norwich Research Park
Norwich
NR4 7TJ
Email: clinpsyd@uea.ac.uk
Tel: +44 (0) 1603 593076
Fax: +44 (0) 1603 591132

Title of Project: Exploring the decision making of clinicians working in neurodevelopmental child services, when considering potential assessments and diagnoses based on referral information, using a hybrid case vignette and survey methodology

Statement of consent:

• I confirm that I have read the participant information above for the research study. I have had the opportunity to consider the information and ask any questions that I have. The researchers have answered any questions I have had satisfactorily.

• I understand that my participation in the study is voluntary and that I am free to withdraw at any time without giving any reason, without my employment or legal rights being affected.

• I consent to the storage and processing of my personal information for the purposes of this and future research studies. I understand that such information will be treated as strictly confidential and handled in accordance with the Data Protection Act 1998.

• I agree to take part in the above study.

If you would like to participate in the study, and agree with the statements above, please click the link below to be directed to the survey.

Appendix P - Participant Debrief Information

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University of East Anglia
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NR4 7TJ
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Title of Project: Exploring the decision making of clinicians working in neurodevelopmental child services, when considering potential assessments and diagnoses based on referral information, using a hybrid case vignette and survey methodology

Thank you for taking part in this research study. Your information will support us to identify what facilitates and what act as barriers to clinicians receiving referrals within neurodevelopmental services, allowing us to identify what the training needs are for clinicians working within these services. This study aimed to identify these needs specifically in relation to Fetal Alcohol Spectrum Disorder (FASD), due to the introduction to the recently released NICE quality standards.

For more information about NICE quality standards, please visit https://www.nice.org.uk/guidance/qs204. The National Organisation for FASD also has a wealth of information available (https://nationalfasd.org.uk/).

If you have any questions or concerns, would like a summary of the findings, or you have decided to withdraw from the study, please contact Emma Heathcote via Emma.Heathcote@uea.ac.uk.

The results of the study may be shared in a range of formats including:

-Publication in academic journals

-Presentation at research conferences

If the study has caused you any distress by taking part, we advise that you contact a member of the research team directly, or speak to your GP. There is also the Samaritans number you can call for from on 116 123.

For any further problems or complaints, please contact the research supervisor, Dr Aaron Burgess via Aaron.Burgess@uea.ac.uk.

Thank you once again for your valued participation. If you would like to be entered into a draw for a £20 Amazon voucher, please enter your email address below. Please note that by doing so, the researchers will have access to your email address, but this will not be linked to your survey information.

Appendix Q – Report of survey bot activity and ethics committee response

04 Aug 2023 – Report to ethics committee:

The survey which is used to collect my data has been infiltrated with fake responses, possibly by bots. The survey has been closed to stop any more responses being collected, and I am currently having meetings with my supervisors on how best to move forward.

We have discussed deleting all of the data and collecting again, however this does not feel a viable option. This is because my study has an element of deception in order to explore its aims - participants do not know the full aims (i.e the topic in question) until the end of the survey. This is an essential element of the study. If I was to try and collect my data again, I would have to ensure that it would be a complete new sample, so that no participants are aware of the aim. This would be very difficult to control for, and it also seems unlikely that I would be able to collect the necessary numbers to allow for analysis. This option also has ethical dilemmas, as I would be deleting data collected from true participants too.

Another option we have discussed is creating a criteria, which would allow us to identify what responses are considered 'real' and which are considered to be 'fake'. This would include criteria such as length of time taken to complete survey for example. Another way to help us identify 'real' data, would be to link survey responses with those who have used an NHS account to complete the form for entry into the raffle. Whilst this wouldn't identify all 'real' participants, It would give us an idea of the style of answers given by 'real' participants, in order to shape our criteria. Of course, this would mean that some of the data may become identifiable by email address, which would need to be considered appropriate by the ethics committee.

On reflection, although not foolproof and there still is a risk that cleaning the data in this way may remove real responses or keep fake responses, I feel like the second option is more viable. Once the criteria has been set, I believe it will be clearer whether this option will work.

Provide details of the parties affected.

I have been affected due to the time this will take to implement a plan and move forward. Should data have to be deleted, then those who are 'true' participants will also be affected, as their meaningful data will not be used for research.

06 Aug 2023 – Ethics response

Study title: Exploring decisions made by clinicians, when assessing and diagnosing in neurodevelopmental child services.

Application ID: ETH2223-0111

Date of approval: 30th January 2023

Dear Emma

The FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee) has received your report of an adverse event and has decided that no action is required.

- All reasonable and practicable steps have been taken to address the matter and safeguard data integrity.

- It is advised that you adopt option 2 in regards to the data collected and its analyses.

On behalf of the FMH S-REC (Faculty of Medicine and Health Sciences Research Ethics Subcommittee)

Yours sincerely,

Dr Paul Linsley

Appendix R - Assessment tools/methods considered by clinicians

Assessment	PAE group		Non PAE group		Total	
	n	%	*n*	%	*n*	%
Full assessment	0	0	1	<0.1	1	<0.1

Partial assessment	51	78.5	55	74.3	106	76.3
ND assessment sections						
Motor/sensory	5	8%	4	5%	9	6%
Neuroanatomy/ Neurophysiology	3	5%	2	3%	5	4%
Cognition/learning	23	35%	23	31%	46	33%
Language	16	25%	8	11%	24	17%
Academic Achievement	1	2%	1	1%	2	1%
Memory	3	5%	1	1%	4	3%
Attention	8	12%	9	12%	17	12%
Executive Functioning	4	6%	0	0%	4	3%
Affect regulation/mental state	10	15%	12	16%	22	16%
Adaptive behaviour/social skills/ social communication	6	9%	6	8%	12	9%
ND assessment	25	38%	26	35%	51	37%
Medical/physical	9	14%	14	19%	23	23%
Sentinel facial features	0	0%	1	1%	1	1%
Genetic Testing	1	2%	0	0%	1	1%
Alcohol use	4	6%	3	4%	7	5%
Developmental history	29	45%	35	47%	64	46%
Observations	31	48%	16	22%	47	34%
Interviews/reports	25	38%	25	34%	50	36%
Screening tools	22	34%	18	24%	40	29%
Other/vague	21	32%	34	46%	55	40%

ND = Neurodevelopmental assessment. ND assessment sections refers to the frequencies and percentages of clinicians stating specific sections covered by an ND assessment, whereas ND assessment refers to clinicians stating ND assessment without reference to specific sections.

Appendix S: Logistic regression between predictor variables and FASD diagnosis

	B	S.E.	Wald	df	*P*-value	OR	95% C.I.	
							Lower	Upper
Gender								
Male*								

Female	1.761	1.203	2.145	1	0.143	5.82	0.551	61.475
Qualification								
Cert of higher education*			2.323	4	0.677			
Foundation degree	-19.718	17243.6	0	1	0.999	0	0	.
Bachelors	-3.604	3.267	1.217	1	0.27	0.027	0	16.419
Masters	-3.393	3.152	1.159	1	0.282	0.034	0	16.201
Doctoral	-2.529	3.101	0.666	1	0.415	0.08	0	34.732
Age								
21-30*			2.946	4	0.567			
31-40	-1.465	2.167	0.457	1	0.499	0.231	0.003	16.147
41-50	-0.323	1.832	0.031	1	0.86	0.724	0.02	26.24
51-60	-0.432	1.762	0.06	1	0.806	0.649	0.021	20.492
61-70	1.31	1.811	0.523	1	0.47	3.707	0.106	129.06
Profession								
Clinical Psychologist*			0.778	5	0.978			
Psychiatrist	-2.102	3.185	0.436	1	0.509	0.122	0	62.847
SLT	-18.638	11663.99	0	1	0.999	0	0	.
OT	0.043	1.016	0.002	1	0.966	1.044	0.142	7.65
Social worker	0.364	0.974	0.14	1	0.709	1.439	0.213	9.716
Nurse	-19.641	15393.1	0	1	0.999	0	0	.
Experience								
<1*			0.478	4	0.976			
1-5	-17.464	17280.22	0	1	0.999	0	0	.
6-10	0.094	0.991	0.009	1	0.924	1.099	0.157	7.665
11-15	-0.11	0.895	0.015	1	0.902	0.896	0.155	5.176
16+	0.558	1.135	0.242	1	0.623	1.747	0.189	16.162
Condition								
Non-PAE condition*								
PAE condition	3.537	0.886	15.926	1	<.001	34.379	6.05	195.345
Constant	-4.128	3.881	1.131	1	0.287	0.016		

NB: Logistic regression explores the difference between levels of a variable and a baseline.* = baseline variable

Appendix T: Logistic regression between predictor variables and appropriate assessment methods

	B	S.E.	Wald	df	Sig.	OR	95% C.I.	
							Lower	Upper
Gender								

Male*								
Female	0.533	0.647	0.678	1	0.41	1.703	0.48	6.048
Qualification								
Cert of higher education*			9.302	4	0.054			
Foundation degree	0.814	1.599	0.259	1	0.611	2.256	0.098	51.85
Bachelors	0.431	1.414	0.093	1	0.761	1.538	0.096	24.599
Masters	1.554	1.341	1.343	1	0.247	4.731	0.341	65.544
Doctoral	2.96	1.334	4.92	1	0.027	19.294	1.411	263.755
Age								
21-30*			3.52	4	0.475			
31-40	1.283	1.885	0.463	1	0.496	3.609	0.09	145.212
41-50	1.874	1.789	1.097	1	0.295	6.514	0.196	217.005
51-60	1.914	1.796	1.136	1	0.287	6.782	0.201	229.22
61-70	0.022	1.739	0	1	0.99	0.978	0.032	29.574
Profession								
Clinical Psychologist*			8.398	5	0.136			
Psychiatrist	1.694	1.473	1.323	1	0.25	5.443	0.303	97.655
SLT	-1.27	1.308	0.942	1	0.332	0.281	0.022	3.649
OT	0.112	0.833	0.018	1	0.893	0.894	0.175	4.571
Social worker	0.898	0.934	0.925	1	0.336	2.455	0.394	15.306
Nurse	-2.19	1.285	2.903	1	0.088	0.112	0.009	1.39
Experience								
<1*			1.54	4	0.82			
1-5	1.608	1.668	0.929	1	0.335	0.2	0.008	5.268
6-10	1.306	1.239	1.111	1	0.292	0.271	0.024	3.074
11-15	-1.24	1.136	1.192	1	0.275	0.29	0.031	2.681
16+	1.451	1.25	1.348	1	0.246	0.234	0.02	2.715
Condition								
Non-PAE condition*								
PAE condition	0.388	0.502	0.597	1	0.44	1.474	0.551	3.944
Constant	2.424	2.496	0.943	1	0.332	0.089		

NB: Logistic regression explores the difference between levels of a variable and a baseline.* = baseline variable

List of Abbreviations

FASD: Fetal Alcohol Spectrum Disorder

PFAS: Partial Fetal Alcohol Spectrum

FAS: Fetal Alcohol Syndrome

FAE: Fetal Alcohol Effects

ARND: Alcohol-Related Neurodevelopmental Disorder

ND-PAE: Neurodevelopmental Disorder Associated with Prenatal Alcohol Exposure

ARBD: Alcohol Related Birth Defects

PAE: Prenatal alcohol exposure

ND: Neurodevelopmental

SDQ: Strengths and Difficulties Questionnaire

CBCL: Child Behaviour checklist

ADHD: Attention deficit Hyperactivity Disorder

ASD: Autism Spectrum Disorder

OT: Occupational Therapy/Therapist

SLT: Speech and Language Therapy/Therapist

CAMHS: Child and Adolescent Mental Health Services

NICE: National Institute for Health and Care Excellence

SIGN 156: Scottish Intercollegiate Guidelines Network 156

SES: Socioeconomic status