

Portfolio Volume 1: Major Research Project

Anxiety and Extreme Demand Avoidance in Children and Adolescents: the roles of Sensory Sensitivities and Intolerance of Uncertainty

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Glossary of Terminology

Definitions and explanations of terminology used within this thesis are provided within the text; however, for those unfamiliar with ‘demand avoidance’ and the various terms often used interchangeably to describe similar constructs, definitions and descriptions are provided. We hope these provide the reader with a foundation for clarity whilst engaging with this thesis.

Demand Avoidance – this generic term describes a natural human trait of not being able to do certain things at certain times, either for yourself or for others, and refers to the things we do to avoid demands.

Pathological Demand Avoidance (PDA) – although there is no conclusive and agreed upon definition of PDA, it is often termed ‘a profile of autism’, meaning it is conceptualised as within the neurodevelopmental diagnosis of autism. PDA is characterised by the individual being driven to avoid everyday demands and expectations to an extreme extent; however, this behavioural aspect is considered just one of a cluster of characteristics that make up the profile.

Extreme Demand Avoidance (EDA) behaviours – this term emerged as an alternative to describe the same construct as PDA but with terminology that was considered more appropriate. However, with the development of a questionnaire (EDA-Q) to specifically measure observable behavioural avoidance, EDA may be considered a description of the behavioural outcomes related to PDA, and not of the other characteristics that are said to make up the cluster of the ‘PDA profile of autism’.

Rational Demand Avoidance (RDA) – this term is used to describe a similar behavioural construct to EDA but attempts to redefine the concept from ‘extreme’ to an understandable

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and rational response to demands which cause distress, anxiety, discomfort, or are overwhelming to the individual.

Demand Avoidance Phenomena (DAP) - this term is an evolution of RDA and is used to describe a similar construct but conceptualised without the assumption of being situated within or outside of autism. In DAP, there is an acknowledgement of the presence of demand avoidance alongside distress; however, this term highlights the need for further research to make sense of the phenomena before being described as part of a neurodevelopmental diagnosis as PDA currently implies.

Autism / Autistic – Much debate continues as to appropriate language to use when describing autistic people. There are various uses of terminology throughout the literature with some referring for example to autistic spectrum disorder (ASD) to describe autism as spectrum, where others replace ‘disorder’ with ‘condition’ (ASC). Furthermore, some do not align with their autism diagnosis being described as on a spectrum. We decided to be guided by our Experts by Experience from the PDA Society who recommended the use of autism to describe the neurodevelopmental diagnosis and refer to those with autism as ‘autistic individuals/people/children/adults’.

Neuro-divergence/diverse – Neurodiversity describes the diversity or variation of cognitive functioning in people, recognising that everyone has a unique brain and therefore different skills, abilities, and needs. Neurodivergence describes those who process, learn, and/or behave differently from what is considered "typical.". Distinguishing itself from ‘ableist’ language, there is a recognition of a difference, without a medical or deficit model required to understand this difference. Although typically associated with autism, ‘Neurodiverse’ was used in this study as an option for parents to identify their child as different to ‘typically developing’ children where those parents also did not associate their child with either autism or pathological demand avoidance.

Intolerance of Uncertainty (IU) – Intolerance of uncertainty describes an unwillingness to tolerate the possibility that negative events may occur in the future, no matter how low the probability. IU is a construct referred to throughout this thesis due to its role in our understanding of anxiety.

Sensory Sensitivities (SS) – Sensory sensitivities can be understood as sensory processing difficulties and can refer to over or under responsiveness to external stimuli. SS is well described in autism research due to its role in making sense of anxiety in autistic individuals.

ABSTRACT

Pathological Demand Avoidance (PDA) is a diagnosis that has been described in clinical practice but not recognised by official diagnostic and statistical manuals. The National Autistic Society (UK) describes PDA as ‘a profile of autism’; however, there is some evidence that strategies for supporting those with autism may be less effective for those identifying with the PDA profile. PDA is described as ‘driven by anxiety’; therefore, it is suggested that a better understanding of the underlying factors contributing to anxiety in this under researched group may support more effective treatment strategies, including shaping school strategies to work with autistic children. No research to date has empirically investigated the role of sensory sensitivities in PDA.

Aim: This study investigates the relationship between autistic traits and extreme demand avoidance (EDA), exploring the contributing roles of both intolerance of uncertainty (IU) and sensory sensitivities (SS) to levels of anxiety; and aims to further develop an understanding of those who identify with the diagnosis of pathological demand avoidance (PDA).

Method: A quantitative, between and within groups design was used. Scores from parent-report measures were analysed using descriptive and inferential statistics, including correlational analysis of the variables, mediation and structural equation modelling (SEM), and repeated measures ANOVA with paired sample t-tests of subscales.

Results: Those identifying with PDA scored higher on the EDA-Q than those with Autism; however, there was no difference between ASSQ scores between those identifying with PDA and those identifying with Autism. SEM found a model with good fit of autism and EDA traits being highly correlated and predicting anxiety. IU and SS had moderating roles in the relationship between autism and EDA traits with anxiety.

Discussion: PDA shows discriminant properties to autism but should be seen within the context of autism. Understanding of the impact of autistic traits in those who identify with

PDA, whether they identify with autism or not, could help make sense of their diagnostic and treatment needs, and supports the concept of PDA as ‘a profile of autism’. It is suggested that there is a need to provide clarity and distinctions with the often-overlapping language used to describe different constructs within the PDA literature. Importantly, understanding sensory needs as a key underlying mechanism behind anxiety in those identifying with PDA , and moving towards a strengths based approach can support tailored, comprehensive assessment schedules and more individualised behaviour management strategies for this population.

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CHAPTER 1: INTRODUCTION

1.1 Foreword

This thesis has been completed as part of a doctoral programme in Clinical Psychology at the University of Hertfordshire. It explores underlying causes of anxiety in children aged 4-17 years old with and without a diagnosis of autism, including children whose parents identify them with the diagnosis of pathological demand avoidance (PDA). This work contains five chapters. In the first chapter, an introduction to the history of pathological demand avoidance (PDA) and how it is situated in relation to autism is presented. An overview of the debates surrounding the topic are offered, and alternate terms and conceptualisations considered with reference to cultural and medical understandings and utility. The second chapter presents a systematic review summarising the literature on what is understood of the underlying causes of anxiety in PDA across the lifespan. Chapters three and four outline the methodology and results of the current study, and chapter five contextualises these results and suggests ideas for future research.

1.1.1 Epistemological position

Prior to undertaking this research project, the primary author experienced working in a special educational needs (SEN) school for children with a diagnosis of autism. Whilst there, the primary author encountered children who identified with the diagnosis of pathological demand avoidance (PDA). Anecdotally, the primary author encountered and observed confusion about the meaning of the diagnosis, from the terminology used to describe the children and their presentations, to the relationships to autism including what were described by teachers as fundamental differences between PDA and what was understood of a more 'typical' autistic presentation. This lack of clarity meant there were reported potential unmet needs in these children identifying with PDA that would not be met by understanding their presentations in relationship to autism alone.

To investigate this phenomenon, we adopted a pragmatic and curious, less expert, position. We wanted to explore what was understood of PDA from the position of those who identify their children with the diagnosis, hence our consultation with Experts by Experience as described below. An investigation into the autism literature revealed contradictory relationships of underlying causes of anxiety (see below) and investigation in the PDA literature (see Systematic Literature Review section) revealed measures of PDA being inconsistently applied. We therefore undertook a more neutral approach to analysis in this study to understand what the response from parents reveals about the profile of PDA and the relationships of underlying causes of anxiety in this population.

1.2 Pathological Demand Avoidance

1.2.1 What is PDA?

Pathological Demand Avoidance (PDA) is conceptualised as a neurodevelopmental diagnostic construct. PDA was first coined in the 1980's by Professor Elizabeth Newson, a Consultant Child Psychologist running an autism assessment clinic in children's services in the UK, to conceptualise behavioural phenomena characterised by "an obsessional avoidance of the demands of everyday life" (Newson et al., 2003). Despite originating from observations in children's services, the term PDA describes a presentation that is also characterised by distress and florid challenging and socially inappropriate behaviour in children, adolescents and adults (Egan et al., 2020). Describing the profile, Newson et al. (2003) identified eight main characteristics associated with PDA: 1) a passive early history over the first year of development; 2) avoidance of demands with use of 'social manipulation' and extreme outbursts if the demands are escalated; 3) surface sociability but apparent lack of sense of social identity, pride or shame; 4) lability of mood and impulsivity that is led by a need to be in control; 5) comfort in role play and pretending with some appearing to "lose

touch with reality”; 6) language delay possibly attributable to passivity, and with strange content in speech; 7) obsessive behaviour; and 8) soft neurological signs (clumsiness, dyspraxia and other signs of physical awkwardness).

1.2.2 PDA, EDA, RDA, DAP...

Whilst *demand avoidance* is a generic term that describes a natural human trait of not being able to do certain things at certain times either for yourself or for others, and refers to the things we do to avoid demands (PDA Society website, 2023), PDA was termed “a necessary distinction” by Newson et al. (2003) because the demand avoidance in this population is often out of proportion, sometimes resulting in violent meltdowns to “seemingly tiny everyday requests” (Sinson, 2013). PDA is often termed ‘a profile of autism’, meaning it is conceptualised as within the Neurodevelopmental diagnosis of autism. There has been ongoing controversy around “ableist language” in autism research and the risk that this language “defines, excludes and marginalises people” (Bottema-Beutel et al., 2021). The term ‘pathological’ being used to describe this group of people from a socio-political perspective has been discussed, with one consideration in the PDA literature being to replace the term ‘pathological’ with ‘extreme’ (O’Nions & Eaton, 2020) to reduce stigma associated with the label of ‘pathological’ (Gillberg, 2014).

Despite the behavioural marker of the individual being driven to avoid everyday demands and expectations to an extreme extent being just one of the cluster of characteristics that make up the ‘PDA profile of autism’ as described above; this could prove a useful marker in the identification of PDA for research purposes. Hence, O’Nions et al. (2014) developed a behavioural screening tool (the Extreme Demand Avoidance Questionnaire; EDA-Q) to identify features of the PDA profile to promote research in this population. It is of note that, as the EDA-Q measures observable behavioural avoidance which has no specificity to autism (Woods, 2022), high scores on the EDA-Q could represent high demand avoidance

behaviours in a non-autistic person and may not necessarily be related to someone who presents with ‘a PDA profile of autism’. EDA itself therefore can be considered a behavioural indicator of the presence of an underlying profile of PDA rather than a replacement term to describe the same diagnostic construct as proposed by Gillberg (2014). Woods (2022) suggested that the term ‘rational demand avoidance’ would more appropriately describe the demand avoidance behaviours that may initially appear extreme to those external to the individual but emphasised the understandable and rational response to demands which cause distress, anxiety, discomfort, or are overwhelming to the individual. Furthermore, Woods (2019) proposed a change in terminology to demand avoidance phenomena (DAP) to describe a similar construct to EDA but conceptualised without the assumption of being situated within or outside of autism. In DAP, there is an acknowledgement of the presence of demand avoidance alongside distress; however, this term highlights the need for further research to make sense of the phenomena before being described as part of a neurodevelopmental diagnosis such as autism, as PDA diagnosis currently implies. Woods (2019) suggests this change in terminology until research can provide validity to the profile when formulating this presentation, in deliberate contrast to what is construed as the dominant ontology of PDA being a profile within autism.

The varying terminology used in the PDA literature, including PDA, EDA, RDA, and DAP are often used interchangeably. This shows the need for clearly defined criteria for the terms used as often they could be said to be referring to slightly different concepts as described above.

However, it is important to note that despite lack of agreement and consistency in the concepts associated with PDA, there is recognition by experienced clinicians throughout child psychiatry, child neurology and paediatrics of the existence of unique distress in the PDA cohort of children and the very major problems encountered when it comes to

intervention and treatment (Gillberg, 2014). Consistent with NICE (2012) guidelines listing demand avoidance as a ‘sign and symptom’ of autism, there is a need to build on the evidence base of these phenomena (Green, 2020) to make sense of this presentation in context and whether a requirement for a separate diagnostic category is indicated.

1.3 Prevalence of Autism & PDA

Since the term PDA was coined in the 1980’s, the criteria for autism within the DSM have also changed, incorporating distinctions to severity level and specifiers (Hare, 2016). New guidelines for identification of autism were introduced in the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-V) by the American Psychiatric Association (2013) due to the increased incidence of and concerns about overdiagnosis (Lobar, 2016). Previously used terms such as ‘autistic disorder’, ‘Asperger disorder’, ‘childhood disintegrative disorder’ and ‘pervasive developmental disorder-not otherwise specified (PDD-NOS)’ were replaced by the collective term ‘autism spectrum disorder’ in the DSM-V. This may have contributed to an observed heterogeneity in presentations of autism (Matson & Kozlowski, 2011) and means prevalence rates across time are difficult to compare due to the changes in diagnostic criteria over time. It has been suggested that these changes facilitate an understanding of PDA as being within the wider spectrum of autism (Green et al., 2018).

The global prevalence of autism is also unclear due to the varying degrees of awareness and diagnostic practices found across different countries. The World Health Organisation (2022) suggest that autism affects around one in 100 people around the world. Elsabbagh et al. (2012) undertook a systematic worldwide review of epidemiological studies and found autism prevalence estimates of one in 161 people. And similarly, Baxter et al. (2014) estimated that in 2010 there was a prevalence of one in 132 people. The UK National Autistic Society (2018) suggest that autism affects around one in 100 people in the UK,

consistent with empirical research in the UK (e.g. Baird et al., 2006), and similar consensus on these prevalence rates has been found across Western Europe (e.g. Idring et al., 2012).

The UK and Western Europe therefore seem to have lower prevalence rates than other countries such as South Korea which has a claimed prevalence rate of 2.64% (Elsabbagh et al., 2012).

In summary, it has been suggested that autism has a prevalence rate of around 1 in 100 people and has increased steadily over the past four decades (Baron-Cohen et al., 2009). The Autism and Developmental Disabilities Monitoring Network reviewed epidemiological surveys concentrating on children in the US and found four times as many boys being diagnosed with autism as girls, but that there were no overall differences between racial, ethnic, and socioeconomic groups (Maenner et al., 2021), and autism is considered a pervasive developmental disorder meaning it is lifelong.

As PDA is construed as a profile of autism, prevalence rates of PDA are considered in the literature in relation to autism. Gillberg et al. (2015) undertook a study to determine the prevalence of PDA within a group of individuals identified as having met the diagnostic criteria for autism. The entire population of 15- to 24-year-olds living in the Faroe Islands were screened for autism, of which sixty-seven met the diagnostic criteria. Nine of these individuals also met criteria for “a possible clinical diagnosis of PDA” meaning one in five of those with autism had indications of having PDA in childhood. It was noted that the study was retrospective and only one of the nine who had previously met criteria for PDA presented as such at the time of assessment, which is contrary to what is understood of autism where the effects of this neurodevelopmental difference are considered pervasive across the lifetime of the autistic individual (American Psychiatric Association, 2013). It remains unclear whether this suggests that the presence of PDA declined in this population study, or whether those living with PDA are better able to cope with or ‘mask’ symptoms with age.

1.3.1 Diagnostic framing

The diagnostic terminology of PDA conceptualises the phenomenon within a dominant individualist and problem-saturated narrative, which may neglect to recognise the strengths of the individual, the environmental factors contributing to anxiety, as well as dominant socio-political narratives that may influence the experience of people given these labels (Watson, 2019). The social impact of an available descriptive label may have contributed to the reported surge of interest in the phenomena of PDA since 2010 (O’Nions et al., 2018a) which may result in people being on the ‘look out’ for such behaviours and using the label of PDA as explanatory. This may explain the increased requests for diagnosis of PDA by parents or caregivers over recent years (Green et al., 2018), which could be said to locate the cause of the presentation within the characteristics of the individual displaying the behaviours when this in fact may not be the most appropriate or useful explanation as to the occurrence of these behaviours. It has been suggested that an alternative nondiagnostic conceptual system such as The Power Threat Meaning Framework (PTMF; Johnstone & Boyle, 2018) could help children and their families make sense of the problems outside of medicalized narratives. However, where autistic traits are conceptualised within the PTMF as ‘meaningful threat responses’, it acknowledges that a neurodevelopmental component is likely to play a role meaning in some circumstances the PTMF would be considered complimentary rather than present itself as an alternative framework to all uses of the diagnosis (Johnstone and Boyle, 2018, p.70). This highlights the importance of developing a clearer conceptualisation of PDA in relation to autism, while at the same time being aware of the social and cultural influences which contribute to that meaning making.

1.4 The PDA profile of Autism

Unlike PDA, autism is recognised by both the International Classification of Diseases (ICD-11) and Diagnostic and Statistical Manual of Mental Disorders (DSM-V) as autism spectrum

disorder (ASD) and characterised by significant difficulties with social communication / interaction, and restricted, repetitive behaviours (American Psychiatric Association, 2013b). The National Autistic Society (2023) additionally name four more difficulties autistic people may share: over or under sensitivity to light, sound, taste, or touch; highly focussed interests or hobbies; extreme anxiety; and meltdowns and shutdowns. Autism is considered a lifelong developmental disability, meaning difficulties have been present from birth and affecting multiple domains of the individual's life. PDA was proposed as requiring its own diagnostic category due to the presence of "social manipulation" as seen in all children meeting the criteria for PDA and having a greater degree of difficulty with emotion regulation compared to those with autism (O'Nions, et al., 2014). O'Nions & Eaton (2020) discriminate between the two suggesting that in autistic children there is often a clearly identifiable reason for their resistance to certain tasks, unlike in PDA where the demand avoidance is described as 'pathological' because it does not appear to make logical sense to the outside observer and leads to severe disruption to the child's everyday functioning.

1.4.1 PDA Society survey

The PDA Society (2018) conducted an online survey for two weeks in March 2018, recruiting through their website and social media. One thousand one hundred and ninety-four parents of children who either identified with or were suspected to have PDA, participated in the study. Consistent with what has been found to be the case for autistic children, 96% of parents reported their child had a need to be in control; 80% reported sensory issues; and 80% reported their child had severe anxiety. They concluded that for outcomes to improve, professionals need to know that they can speak openly about this group using unambiguous terminology and, most importantly, promote appropriate strategies. Moreover, parents reported that the usual Autism strategies were ineffective with helping their children, namely: High level of structure; Usual boundaries; Rewards and consequences; and Praise. These

strategies implemented by Special Educational Needs Schools are effective in supporting autistic children with attendance; however, are ineffective with PDA children. Instead, parents have reported the collaborative strategies which work in PDA to include: Flexibility around structure; Limited ground rules; Negotiation; and Indirect affirmation. PDA is therefore described as having some features consistent with autism and some that are not, and PDA is described in summary as being ‘driven by anxiety’ (PDA Society, 2018).

1.4.2 Anxiety, PDA and Autism

There is an ongoing debate around the terminology of PDA, undermining the self-advocacy of people living with a diagnosis of autism as well as neglecting the potential role of anxiety as a possible underlying or contributing factor (Kildahl et al., 2021). Anxiety disorders are a cluster of psychiatric disorders recognised by both the International Classification of Diseases (ICD-11) and Diagnostic and Statistical Manual of Mental Disorders (DSM-V). The American Psychological Association (2023) defines anxiety as ‘an emotion characterized by feelings of tension, worried thoughts and physical changes like increased blood pressure.’ Anxiety has long been considered one of the most common mental health problems amongst youth (Costello et al., 2005). Assessing for anxiety among adolescents is imperative as previous research suggests that adolescents with anxiety have poorer family relationships, lower life satisfaction, substance use difficulties, alcohol abuse/dependence difficulties, are diagnosed with other comorbid disorders, and have significant educational underachievement (Essau et al., 2014). Additionally, anxiety that begins early and is left untreated tends to become chronic, persisting later in life (Letcher et al., 2012).

It is estimated that just under half of all autistic people have experienced a comorbid anxiety disorder at some point. For example, a meta-analysis focusing on prevalence of co-occurring anxiety disorder in children identified that 40% of children with autism had at least one comorbid anxiety disorder (Van Steensel & Heeman, 2017) and that admissions to mental

health inpatient services for young people affected by autism, although seen by many practitioners as a last resort, have in fact increased over time (Narendorf et al., 2011).

Another meta-analysis reported a lifetime prevalence of an anxiety disorder in 42% of autistic adults (Hollocks et al., 2019).

Research into anxiety in the autistic population has described underlying contributing factors to anxiety. For example, Neil et al. (2016) found that although autism and anxiety were highly correlated, there was no direct effect of autism on anxiety when *Intolerance of Uncertainty* and *Sensory Sensitivities* were accounted for. This means that there are underlying constructs that contribute towards anxiety in autistic individuals, and it is not simply that being autistic means the individual will experience anxiety.

1.4.2.1 *Intolerance of Uncertainty*

Intolerance of Uncertainty (IU) can be viewed as a dispositional characteristic that results from a set of negative beliefs about uncertainty and its implications, and the tendency to react negatively to uncertain situations and events (Dugas & Robichaud, 2007). Individuals who are intolerant of uncertainty believe that not knowing what is going to happen is negative and should be avoided (Jenkinson et al., 2020), and they over-value predictability and tend to become overwhelmed by the unexpected or the unknown (Carleton, 2016).

The construct of IU has been identified as a critical construct and risk factor for the development and maintenance of a range of anxiety disorders (Carleton, 2012) in general population samples (e.g. Dugas et al., 2001) including generalised anxiety (Carleton et al., 2012) and social anxiety (Buhr & Dugas, 2006; Whiting et al., 2014) and obsessive compulsive disorder (Calleo et al., 2010). IU has also been found to be a shared mechanism in anxiety and autism (Chamberlain et al., 2013), with a causal pathway of increased IU in this population related to increased anxiety (Boulter et al., 2014; Jenkinson et al., 2020). The effect of IU was found to be higher in autistic than neuro-typical individuals (Chamberlain et

al., 2013) as “even slight uncertainty” is reported to lead to anxiety in autistic people (Ashburner et al., 2013). Related to specific aspects of anxiety, IU was found to be related to generalised anxiety (Holaway et al., 2006) and social anxiety (Boelen & Reijntjes, 2009), moderately related to obsessions/compulsions, and weakly related to panic sensations (Dugas et al., 2001).

It is suggested that understanding variance within the construct of anxiety may help differentiate those with PDA from autism. Heightened levels of IU make autistic children more anxious and one strategy to manage anxiety is Repetitive, Restrictive Behaviours (RRBs; Joyce et al., 2017), which are a well-known behavioural trait in autism. RRBs are described as the individual’s attempt to exert some control over the environment and make the world more predictable (Lidstone et al., 2014), and it is suggested that Extreme Demand Avoidance behaviours could serve a similar purpose in those with PDA. As Carleton et al. (2012) describe IU as a cross-diagnostic construct and not simply an aspect of anxiety, and there is variance in the impact of IU within autistic individuals as a population, IU may also be a significant factor when considering the underlying factors contributing to anxiety in those with PDA.

1.4.2.2 Sensory Sensitivities

The research into underlying factors that have contributed towards anxiety in Autistic individuals is reflected in the DSM-V describing changes from the DSM-IV (American Psychiatric Association, 2000) to include sensory behaviours into the diagnostic criteria of autism. Sensory processing involves receiving sensory stimuli from the environment and the neurological processes that generate the conscious experience of sensation (Miller et al., 2007), and it also encompasses responses to stimuli including behavioural responses (Tseng et al., 2011). Some people have difficulties regulating and organising their behavioural responses to sensory input in line with environmental demand (Miller et al., 2007), including

both autistic children and adults, who have been reported to respond to sensory experiences differently to their peers (Tomchek & Dunn, 2007). Importantly, anxiety in autism has often been linked to an individuals' sensory sensitivities (Green et al., 2012).

Much of the existing research into sensory sensitivities in autistic children has been conducted indirectly through the mothers' accounts; however, there is evidence directly from autistic people themselves that they have impairments with modulating incoming sensory input (Grandin, 1995). Furthermore, SS has often been explored in the context of IU and anxiety in the autistic population. For example, Wigham et al. (2015) examined the relationships between sensory processing abnormalities and repetitive behaviours, with IU and anxiety as mediators in the relationship. They reported a moderate positive correlation between intolerance of uncertainty and sensory over-responsiveness, meaning a relationship between IU and SS is suggested in those with autism.

It is of note that the model adopted by Wigham et al. suggested a relationship of SS predicting RRBs, and IU and anxiety were considered moderators in this relationship. In their findings, it is suggested that sensory over-responsiveness gives rise to anxiety. In contrast, where Neil et al. (2016) explored these constructs in autistic and non-autistic children, their model described IU predicting SS, with anxiety as mediating the relationship. Neil et al. followed the theory of Pellicano & Burr (2012) who hypothesise that a desire to reduce uncertainty may increase anxiety symptoms such as becoming hyper-vigilant to signs of threat in the environment, and it may be in these times of hyper-vigilance that those individuals may be more likely to notice or react to aversive sensory stimuli. By making sense of these constructs and how they related to each other, including differences across diagnostic groups, may help differentiate those with PDA from those with autism.

1.5 Clinical Implications

Anxiety is a difficulty for many autistic people which can affect them psychologically and physically and impact their quality of life. Therefore, it is therefore important that autistic people learn to recognise triggers and find coping mechanisms to help reduce anxiety and help them to thrive (National Autistic Society, 2023). However, many autistic people have difficulty recognising their emotions and regulating emotions such as anxiety, and therefore, those supporting them, including services, should recognise the external, environmental factors which can help alleviate this distress as well as those internal to the autistic individual. Currently, the treatment with the most evidence supporting its efficacy in ameliorating childhood anxiety disorders is cognitive behaviour therapy (CBT; Velting et al., 2004). Cognitive behaviour therapy utilizes both cognitive restructuring and exposure techniques to reduce anxiety and enable anxious individuals to cope more effectively with their anxiety. Understanding the different underlying factors contributing to anxiety can shape CBT treatment, such as a focus on cognitive distortions related to uncertainty for those with autism. Alternatively, where SS is conceptualised as a predictor of anxiety in those with autism, it is suggested that CBT can support the development of sensory modulation through techniques such as graded exposure (Rodgers & Ofield, 2018), or attention can be paid to the external stimuli whereby a suitable environment with reduced excess sensory stimuli (such as reduced noise) could support the emotional wellbeing of autistic people. It is of note however, that CBT has often been found unhelpful in meeting the particular needs of autistic children (PDA Society, 2018), with it needing flexibility to account for sensory processing and the unique attributes of neurodivergent individuals. For example, application of CBT that relies on recognising emotions may be incongruent for those experience alexithymia or having poor interoception which are understood to be associated with autism. Furthermore, graded exposure techniques often target ‘irrational fears’, such as a phobia of spiders in

typically developing individuals; however, CBT needs to consider the context of autism, whereby the fear may indeed be a 'rational' fear, such as the sensory overload of a school environment for an autistic person with additional sensory needs. This highlights the need for mainstream therapeutic methods to be flexible and recognise the significant adaptation and development to meet the needs of the neurodivergent population, including those who identify with autism and PDA, to consider not only the internal world of the individual's thoughts, feelings and behaviours, but the environment, and the experience of that environment which may be the focus of intervention. Understanding the relationships of the underlying constructs contributing to anxiety in those who identify with PDA can therefore help shape management plans and clinical interventions that may be more suited to this population.

CHAPTER 2: SYSTEMATIC REVIEW OF RELEVANT LITERATURE

2.1 Overview

This chapter presents a systematic literature (SLR) of what is understood of the underlying factors contributing to anxiety in children and adults with pathological demand avoidance (PDA). Analysis and critical appraisal are provided in relation to identified papers.

Thereafter, gaps in the literature are highlighted to inform the rationale for the present study.

2.2 Introduction to the Literature Review

The aim of this SLR was to bring together a body of evidence in relation to the topic, and is “characterised by being methodical, comprehensive, transparent, and replicable” (Siddaway et al., 2019). This SLR offers explanations as to how research studies fit together, allowing for robust conclusions to be drawn, and provides implications for future research. This systematic literature review aimed to explore the existing scientific evidence across the PDA lifespan and ultimately work towards the goal of understanding how underlying factors contributing to anxiety support or even challenge PDA as ‘a profile of autism’ and/or extreme anxiety.

2.3 Rationale for current SLR

An initial scoping literature search revealed that a previous systematic review on PDA in children and adolescents has been conducted by (Kildahl et al. (dated 2020; published 2021)). However, a more recent review was deemed necessary to account for both the current surge of interest from researchers and clinicians in PDA (O’Nions et al., 2018) in a very under-researched population group, but also there have been empirical studies completed since the Kidahl et al. review, including studies that have incorporated adults. In addition, the current review focusses specifically on what is understood of the underlying factors of anxiety in the population group across the lifespan, and what we know of the relationship between these,

which have not been explored previously. To the author's knowledge, there is no published or ongoing review that examines and compares underlying factors contributing to anxiety in pathological demand avoidance across the lifespan, whether conceptualised as a separate diagnostic construct or a 'PDA profile of autism'. This systematic review of the literature on PDA aims to investigate the following questions:

- What is the relationship between anxiety and demand avoidance?
- Can intolerance of uncertainty (IU) and/or sensory sensitivities (SS) relate to anxiety in pathological demand avoidance?
- How does the relationship compare to other neurodivergent conditions, and those who have co-occurring symptoms?
- Is the relationship affected by age, gender, IQ or even informant type (self-report vs proxy-report)?

This systematic review of peer-reviewed empirical literature is entitled:

Investigating the factors underlying anxiety in individuals with pathological demand avoidance across the lifespan: a systematic review.

2.4 Methods

2.4.1 Protocol and Registration

A systematic review was undertaken according to the principles outlined in Preferred Reporting Guidelines for Systematic Reviews and Meta-Analyses (PRISMA; Page et al., 2021). A systematic review protocol was registered on 9th February 2023 (see Appendix A) prior to data extraction (https://www.crd.york.ac.uk/prospero/export_details_pdf.php).

2.4.2 Search strategy

In keeping with the methodology described by (Siddaway et al. (2019), the search was divided into individual concepts to create search terms and consideration was given to

different terminology that may be used to describe the same constructs. Previous reviews on PDA were also consulted to review search terms and ensure no overlapping content.

Consultation from an independent expert on carrying out systematic reviews supported the framing of the search strategy. Search concepts were constructed with reference to ‘the identified phenomenon’ and ‘the underlying factors’ as outlined in Table 1 below.

Table 1: Search concepts

The identified phenomenon:	pathological demand avoidance
	AND
The underlying factors:	anxiety OR intolerance of uncertainty OR sensory sensitivities

From existing reviews in PDA, it was noted that some authors had replaced the word ‘pathological’ with alternatives, such as ‘rational’ or ‘extreme’, and others included the term ‘syndrome’. Therefore, to capture the various terminology, the search included the term ‘demand avoidance’ to capture all associated terms. Some authors use the term ‘anxious’ whilst others use ‘anxiety’, and therefore, using truncation, the term ‘anxi*’ was used to capture all variations. Due to the research literature on autism highlighting ‘intolerance of uncertainty’ and ‘sensory disturbances’ to often underlie their anxiety (Neil et al., 2016) the terms ‘intolerance of uncertainty’ and ‘sensory’ were also used respectively as additional search terms to include these known constructs. Search terms were identified by extensive search of the literature related to PDA but also related to our key factors of interest (intolerance on uncertainty and sensory sensitivity). These terms are exhaustive of what is currently outlined related to all factors of interest for the search terms. Boolean operators ‘AND’ and ‘OR’ were used to combine the search terms into the following finalised search combination:

demand avoidance AND (anxi OR 'intolerance of uncertainty' OR sensory)*

The search was performed in four databases accessed via the University of Hertfordshire: Scopus, PsycArticles, PubMed, and Medline. Reference lists for identified papers were scrutinised. The systematic literature search concluded on 30th May 2023.

2.4.3 Eligibility Criteria

Any paper describing empirical data involving participants with pathological/extreme demand avoidance, children and adults (both with and without co-occurring disorders) or caregivers of a child/children with PDA published in a peer-reviewed journal was included in the review. There was no limitation on the date range of the published articles, in view that the literature on PDA is fairly sparse. Equally, there were no restrictions to the location of the study or type of methodology that has attempted to address this relationship albeit quantitative, qualitative, mixed methods and/or randomised control trials. Studies not in English, or in book chapters, book reviews, commentaries or position papers were excluded.

Table 2: Inclusion and Exclusion Criteria

<i>Inclusion criteria</i>	<i>Exclusion criteria</i>
Published in the English language	Not published in the English language
Published in a peer-reviewed journal	Books
Qualitative and quantitative studies	Book reviews
Intervention based studies	Commentaries

2.4.4 Study Selection

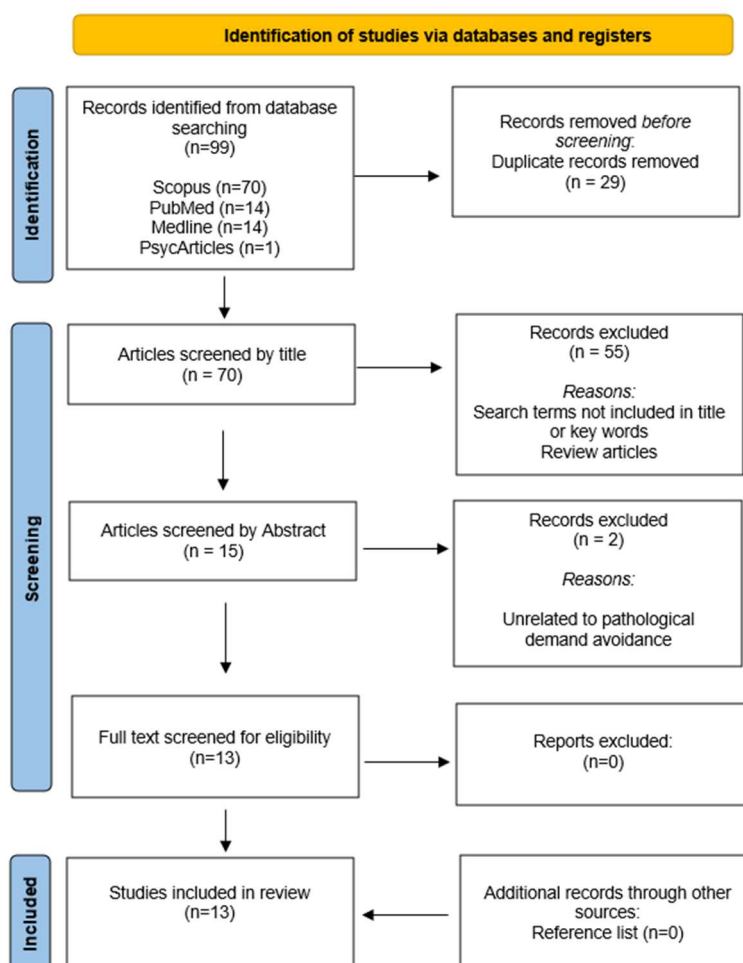
Search results were exported from bibliographic databases to reference management software. Duplicates were then removed. Authors A.J.R. and A.K.L. independently screened all titles and abstracts according to the inclusion / exclusion criteria in Table 2 (see above) for

relevance, and a full-text review was performed on all papers referring to PDA or demand avoidant behaviours. Final study selection was completed independently by A.J.R. and A.K.L., and no discrepancies occurred in the final list of included studies. Conflicts were resolved through collaborative discussion and critical review of the individual study aims. Data including sample population, sample size, mean age range, comparison groups, study aims, measures used, and main outcomes were tabulated (see Table 3 below).

2.4.5 Search Process

The PRISMA flowchart (Page et al., 2021) shown in Figure 1 below outlines the process of identifying, screening and selecting papers meeting eligibility criteria.

Figure 1: PRISMA flow chart for study selection



2.4.6 Data Extraction

The initial search of databases identified 99 papers. 29 were duplicates and removed. Titles and then abstracts were screened using the inclusion/exclusion criteria leaving 13 for full text review. No studies from outside of this search were included maintaining the systematic nature of this review. All 13 met criteria for inclusion. Reference sections for each of these papers was checked, and no further papers were identified for inclusion in the final studies selected for inclusion in the review. Data from individual studies were extracted by A.J.R (see Table 3 below) and checked by A.K.L.

Table 3: Summary of final papers identified for inclusion in the SLR

No.	Study authors	Clinical (PDA) Group details & Diagnostic / screening measures used for PDA?	Control or comparison group used? Y/N?	Aim of study (paragraph)	Methodology used: Quant? Qual? Type of measures used?	Results: main outcome (concluding paragraph)
DATABASE SEARCH: Research addressing anxiety in those presenting with demand avoidant behaviours						
1	Schneider et al., 2022	<p>CYP: EDA-Q with care team members. 19% (n=6) of autistic children fulfilled the criteria of a PDA syndrome as assessed by the EDA-Q</p> <p>32 children and adolescents (29 boys and 3 girls) with ASD (ages 6 to 16 years). Inclusion criteria were children admitted to the “therapeutic classes” (which require a diagnosis of ASD to enrol).</p>	Y - comparison	To investigate the impact of severe ASD, psychiatric comorbidities, adaptive level and a PDA syndrome on the evolutions of behaviour, school adjustment and QoL in autistic children	<p>QUANT: Observational study design. Exploratory only and not experimental or confirmatory.</p> <p>ASD assessed with the Social Communication Questionnaire (SCQ) – with care team members; and the Autism Diagnostic Interview – Revised (ADI-R).</p> <p>ADHD assessed with the Revised Conners’ Parent and Teacher Rating Scales (CRS)</p> <p>Intellectual efficiency was assessed with the WISC-IV, WPPSI-III or WNV depending on the age and the profile of the child.</p> <p>Anxiety assessed with the parent version of the Screen for Child Anxiety Related Disorders (SCARED)</p> <p>QoL was assessed with the Kidscreen-27</p>	<p>PDA diagnosis was not associated with any specific evolution of behaviour, QoL or adjustment at school.</p> <p>Severe ASD was associated with a better evolution of school adjustment. Psychiatric comorbidities (anxiety and ADHD) were associated with a worse time course of school adjustment.</p>

No.	Study authors	Clinical (PDA) Group details & Diagnostic / screening measures used for PDA?	Control or comparison group used? Y/N?	Aim of study (paragraph)	Methodology used: Quant? Qual? Type of measures used?	Results: main outcome (concluding paragraph)
					<p>Adaptive behaviour was assessed with the Vineland Adaptive Behaviour Scale, Second edition (VABS-II)</p> <p>Parent report Kiddie-Schedule for Affective Disorders and Schizophrenia for school-age children (K-SADS) (limited to anxiety disorders and ADHD)</p>	
2	O’Nions et al., 2018	<p>CYP: 29 parents of children reported to have an autism spectrum diagnosis reporting features of PDA (aged 7 years 10 months to 16 years 11 months)</p> <p>A short version of the EDA-Q (consisting of 11 items (items 1, 3, 9, 10, 11, 14 (Reversed), 15, 18, 21, 22, and 25 of the original version) to screen for PDA</p>	N	To provide an in-depth exploration of parent perspectives on maladaptive behaviours in children reported to have an autism spectrum diagnosis and features of PDA	<p>QUAL: Semi-structured interview (N = 12 by telephone, N = 17 using a self-administered electronic or written format).</p> <p>Interview has not been validated and is not presumed to assay behaviours that are necessarily specific to this presentation. 22 questions, with 55 subquestions, drawing on content from the Diagnostic Interview for Social and Communication Disorders (DISCO; Wing, Leekam, Libby, Gould, & Larcombe, 2002) and from an interview developed by Newson et al. (2003)</p>	Descriptions consistently emphasised the child’s attempts to control situations and others’ activities as major areas of difficulty. Strategic behaviour was reportedly employed both to avoid demands and to insist that things were done on their terms.
3	Brede et al., 2017	CYP: 9 students (eight male, one female; M age = 13.65 years), with independently received clinical diagnosis of autism and the majority with	N	<p>1) to understand the factors that precipitated exclusion from school</p> <p>2) to understand which factors needed to be</p>	<p>MIXED: Quant for screening tools; Qual for interviews re: schooling.</p> <p>Quant:</p>	Parents reported elevated levels of demand avoidant behaviour on the EDA-Q, presumably observed at home, whereas teachers did not.

No.	Study authors	Clinical (PDA) Group details & Diagnostic / screening measures used for PDA?	Control or comparison group used? Y/N?	Aim of study (paragraph)	Methodology used: Quant? Qual? Type of measures used?	Results: main outcome (concluding paragraph)
		<p>a history of demand avoidant behaviour.</p> <p>PDA screened with EDA-Q. All parents reported their children to present with EDA; however, none of the teachers rated children as having clinically significant EDA.</p> <p>All had received an independent clinical diagnosis of Asperger's syndrome (n=6), autism (n=2) or atypical autism (n=1).</p> <p>All being educated in an SEN 'Inclusive Learning Hub', to re-integrate excluded, autistic students back into school. Had Statement of Purpose or EHCP that cited Autism as primary need.</p>		<p>in place for these young people to be successfully reintegrated in school</p>	<p>Self-report; parent and teaching staff report.</p> <p>SCQ cut off score of 15 to validate Autism diagnosis.</p> <p>Raven's Standard Progressive Matrices (Raven, Raven, & Court, 1992) for non-verbal reasoning scores.</p> <p>Social Responsiveness Scale-2 (SRS-2) (Constantino & Gruber, 2012) completed by teachers which assesses social and behavioural difficulties associated with autism.</p> <p>Spence Child Anxiety Scale for Parents (SCAS-P) (Spence, 1999) to measure anxiety.</p> <p>Eight questions were asked about each child's previous educational placements (derived from Gore-Langton & Frederickson's, 2015, Educational Experience Questionnaire [EE-Q])</p> <p>Qual: Semi-structured interviews were conducted with children, parents and teachers. The majority of questions were open-ended, but the style was individually</p>	<p>Several parents described how their child's difficulty with accessing school was due to a lack of understanding of their children's EDA-related behaviours, explaining that the demands placed on the child 'caused distress and massive anxiety'.</p> <p>Interview themes: 1) gradual decline in engagement with school; 2) perpetual state of crisis.</p>

No.	Study authors	Clinical (PDA) Group details & Diagnostic / screening measures used for PDA?	Control or comparison group used? Y/N?	Aim of study (paragraph)	Methodology used: Quant? Qual? Type of measures used?	Results: main outcome (concluding paragraph)
					adapted to meet the needs of individual participants.	
4	O’Nions et al., 2021	<p>CYP: Parents/caregivers of 334 children with reported ASD aged 5–17 years.</p> <p>EDA-Q to measure EDA traits (n=103; 30.8%)</p> <p>Social Communication Questionnaire (SCQ) – Lifetime Version to measure ASD severity.</p>	<p>Y – comparison.</p> <p>Autism</p>	<p>To conduct psychometric analysis to refine the EDA-Q using data from parents/ caregivers of children reported to have an ASD diagnosis. To identify whether one or more dimensions best described EDA-Q items in an ASD sample, and to drop items that were not sufficiently discriminating, or which behaved differently with respect to quantifying EDA dependent on the child’s age, gender, ability level, or independence in daily living activities.</p>	<p>QUANT: Parent-report Strengths and Difficulties Questionnaire (SDQ)</p> <p>Emotion Dysregulation Inventory (EDI) was used to quantify observable signs of emotional dysregulation in children with ASD</p> <p>Non-compliance measured with Home Situations Questionnaire – PDD</p>	<p>PCA and IRT analyses identified eight items that are discriminating indices of EDA traits, and behave similarly with respect to quantifying EDA irrespective of child age, gender, reported academic level, or reported independence in daily living activities. The “EDA-8” showed good internal consistency (Cronbach’s alpha = .90) and convergent and divergent validity with other measures.</p> <p>23 of the 26 EDA-Q items loaded significantly onto a single underlying component</p> <p>Those with more severe ASD are not more likely to show PDA characteristics</p>
5	O’Nions et al., 2016	<p>CYP & ADULTS: N=27 with PDA. 11 participants with “substantial PDA features”. 8 male, 3 female (mean age = 15.7 years, range = 6–27 years). 16 participants with “some” PDA features. 10 male, 6 female (mean age 15.7 years (range = 6–41 years)</p>	<p>Y – comparison.</p> <p>Autism.</p> <p>153 individuals were assessed for ASD using the DISCO</p>	<p>To identify interview questions from the DISCO items that are characteristic of and relatively specific to PDA, being uncommon in the autism spectrum in general</p>	<p>QUAL: Analysis from scoring of DISCO semi-structured interviews.</p>	<p>The imperative behind this work is the very significant behavioural challenge this sub-group present compared to most individuals with ASD</p> <p>‘Apparently manipulative behaviour, difficulties with other people, harassment of others, fantasising, lying, cheating, stealing and socially shocking</p>

No.	Study authors	Clinical (PDA) Group details & Diagnostic / screening measures used for PDA?	Control or comparison group used? Y/N?	Aim of study (paragraph)	Methodology used: Quant? Qual? Type of measures used?	Results: main outcome (concluding paragraph)
		<p>Clinician rated diagnostic criteria with use of DISCO. Screened with 11 of the most PDA-specific subset of relevant DISCO items.</p> <p>26 of those with PDA also met criteria for ASD.</p>				<p>behaviour' are features that differentiate PDA from Autism</p>
6	Egan et al., 2020	<p>ADULTS: 126 adults (32 male, 87 female and 6 other), mean age 34.6 years (range 18 to 68).</p> <p>PDA (n=12) screened with EDA-Q.</p> <p>No corroboration of diagnoses. Measured against screening tools. RAADS for Autism</p>	<p>Y – comparison. Autism.</p> <p>ASD (n=75) screened with RAADS; depression n=59; dyspraxia n=7; dyslexia n=8.</p>	<p>To explore the correlation between impulsivity, ADHD, and general personality traits such as low agreeableness, low conscientious and low emotional stability and measures of PDA and ASD</p>	<p>QUANT: Online survey: demographics, EDA-QA, IPIP, ASRS, RAADS, SRED, and PID-BF</p>	<p>Individual differences underlie self-reported PDA but not Autism. PDA had three significant independent predictors: ADHD, low emotional stability, and antagonism.</p> <p>A previously observed predictive association between PDA and delinquency was not seen in the current study</p>
7	Stuart et al., 2020	<p>CYP: Study 1: N = 214; 141 boys and 73 girls. Parents report diagnoses. 69 with PDA (44 male; 25 female)</p> <p>Study 2: N=11; mean age of 9.45 years And mostly male (63.6%). = 5 had comorbid PDA and ASD, n = 4 had probable PDA only (i.e., no formal diagnoses), n = 1 had PDA</p>	<p>Study 1: Y – comparison. Autism</p> <p>Study 2: Y – comparison. Autism</p>	<p>To explore the idea that, when faced with external demands, particularly those involving uncertainty, children demonstrating EDA behaviours experience significant anxiety that leads to avoidant, noncooperative and at times extreme, defiant behaviour.</p>	<p>MIXED: Study 1: online survey; Extreme Demand Avoidance Questionnaire (EDAQ) The Revised Children's Anxiety and Depression Scale: parent report version (R-CADS-P) Social Responsiveness Scale-Version 2: Parent report Intolerance of Uncertainty Scale: parent report (IUSP)</p> <p>Study 2: semi-structured interviews and content analysis.</p>	<p>Both anxiety and IU significantly predict EDA behaviour in those with PDA. However, the combined contribution of IU and anxiety was 15% leaving much variance still unexplained. IU emerged as a stronger predictor of EDA behaviour than anxiety. These results were observed when co-occurring ASD diagnosis was controlled for; ruling out the possibility that this association was attributable to the overlap of PDA and ASD symptomology.</p>

No.	Study authors	Clinical (PDA) Group details & Diagnostic / screening measures used for PDA?	Control or comparison group used? Y/N?	Aim of study (paragraph)	Methodology used: Quant? Qual? Type of measures used?	Results: main outcome (concluding paragraph)
		only, and n = 1 had ASD only. All but one scored above the clinical cut-off on the EDAQ				There is variation in the individual contributions of IU and anxiety according to the type of EDA behaviour: ‘control’ behaviour seems to be directly influenced by IU, ‘fantasy’ behaviour appears to be the result of a combined effect of IU and anxiety, and ‘meltdown’ behaviour is influenced by IU with a mediation effect via anxiety.
8	O’Nions et al., 2014	<p>CYP: PDA (n=25). Parents of 25 children clinically identified as having PDA (mean age = 11.69 years, standard deviation (SD) = 2.00 years, 68% males)</p> <p>Parents confirmed that their child had been identified as having PDA by a healthcare professional (paediatrician, educational psychologist, clinical psychologist, child psychologist, psychiatrist).</p> <p>No screening measure used.</p>	<p>Y – comparison</p> <p>Autism (N=39; mean age= 11.28 years, SD= 0.74 years, 85% males)</p> <p>Children with conduct problems and callous-unemotional traits (CP/CU) (in DSM-V) (n=28)</p>	To compare parent-reported behavioural difficulties in children receiving the PDA label and children with autism or conduct problems and callous unemotional traits.	<p>QUANT:</p> <p>Screening measures:</p> <p>Autism group: the Childhood Autism Spectrum Test (CAST; Scott et al., 2002)</p> <p>CP/CU group: a score of 25 or above on the Anti-social Process Screening Device (APSD) measure of anti-social traits</p> <p>Scales: the Strengths and Difficulties Questionnaire (SDQ; Goodman, 1997)</p>	The PDA group had significantly higher levels of SDQ-rated emotional symptoms (anxiety/internalizing problems) than either the ASD or CP/CU group.
9	Reilly et al., 2014	CYP: Children identified by the study research psychologist with PDA (n=4).	<p>N.</p> <p>Comorbid diagnoses screened for.</p>	To develop an understanding of the range of neurobehavioural comorbidities associated with childhood epilepsy	<p>QUANT: Strengths and Difficulties Questionnaire</p> <p>Autism Spectrum Screening Questionnaire</p> <p>ADHD Rating Scales-IV</p> <p>DCD-Questionnaire</p> <p>Spence Children’s Anxiety Scale</p>	<p>All four children with significant PDA features showed significant neurobehavioural comorbidity.</p> <p>All four children exhibiting PDA features met criteria for ADHD including inattention and</p>

No.	Study authors	Clinical (PDA) Group details & Diagnostic / screening measures used for PDA?	Control or comparison group used? Y/N?	Aim of study (paragraph)	Methodology used: Quant? Qual? Type of measures used?	Results: main outcome (concluding paragraph)
		Assessed using the Extreme Demand Avoidance Questionnaire (EDA-Q)				hyperactivity/ Impulsivity, three for ASD and two for ODD
10	Doyle & Kenny, 2023	<p>ADULTS: Survey: 335 participants. N = 264 parents, N = 9 life partners, N = 6 individuals experiencing PDA and N = 54 practitioners.</p> <p>Self-report; no screening tool.</p> <p>Semi-structured interviews: individuals living with PDA (N = 2), life partners of individuals living with PDA, practitioners diagnosing and supporting children and adults with PDA (N = 2); one education professional (secondary teacher) and one healthcare practitioner (psychologist) and parents, carers or guardians of children and adults with PDA (individual interviews N = 5);</p> <p>Focus group participants (N = 4).</p>	N	<p>To gather the viewpoints of individuals who experience PDA, families, and practitioners, with the intention of:</p> <ol style="list-style-type: none"> 1) determining levels of knowledge, recognition and understanding of PDA; 2) exploring current pathways to assessment and diagnosis; 3) capturing experiences of access to education and healthcare services; 4) identifying effective intervention and support strategies; and 5) illuminating education and health outcomes for individuals and families. 	<p>MIXED method.</p> <p>Quant: Online survey adapted from the 'Being Misunderstood' (PDA Society, 2018) complemented by semi-quantified thematic analysis of open-ended survey questions.</p> <p>Qual: In-depth, semi-structured individual interviews and a focus group.</p>	<p>61 in PDA group reported extreme anxiety and intolerance of uncertainty</p> <p>37% of professionals believed that they had a good understanding of the presentation of PDA, whereas Parent/carer respondents believed that professionals were 'rarely' or 'never' aware of PDA.</p> <p>While sensory and anxiety issues are more prevalent among younger children in the study, where these are experienced by older children, they have a greater impact on parental well-being.</p> <p>Reported interplay between sensory issues (81.42%), need to feel in control (81.82%) and severe anxiety (78.26%) which may be impacting on difficulties with daily routine (67.59%), school absence (52.57%) and problems sleeping (56.52%).</p>
11	Truman et al., 2021	<p>ADULTS: AUT-PDA group: 57 children with Autism and PDA diagnosis.</p> <p>Total: 211 PDA children.</p>	<p>Y – comparison.</p> <p>AUT-EDA group (n=91): Autism, no</p>	<p>To extend Gore Langton and Frederickson's (2016) work by directly comparing the educational experiences</p>	<p>MIXED methods. Online survey. Assessing parental views on their children's current and previous educational experiences (taken</p>	<p>EDA-Q scores of the AUT group were significantly lower than those of the AUT-EDA and AUT-PDA groups (ps <.001).</p>

No.	Study authors	Clinical (PDA) Group details & Diagnostic / screening measures used for PDA?	Control or comparison group used? Y/N?	Aim of study (paragraph)	Methodology used: Quant? Qual? Type of measures used?	Results: main outcome (concluding paragraph)
		<p>Respondents were: 200 biological parents, 7 adoptive parents and 4 grandparents.</p> <p>Parent report diagnoses.</p> <p>EDA-Q screen for PDA.</p>	<p>PDA diagnosis, but above EDA-Q cut off score.</p> <p>AUT group (n=63): Autism (no PDA and below cut off on EDA-Q).</p>	<p>of autistic children with and without EDA behaviours, and with and without a PDA diagnosis.</p>	<p>directly from the Educational Experiences Questionnaire, EE-Q.</p> <p>Extreme Demand Avoidance Questionnaire (EDA-Q) Strengths and Difficulties Questionnaire (SDQ) Autism traits screen: Social Responsiveness Scale-2 (SRS-2)</p>	<p>There were no significant differences in the latter two groups' EDA-Q scores ($p = 1.00$).</p> <p>AUT group had lower SDQ scores, reflective of fewer emotional and behavioural difficulties, than children in the other two groups</p> <p>Parents of children with EDA behaviours (with and without a diagnosis of PDA) felt that the schools' misunderstandings were attributable to their children's unusual presentation</p>
12	Trundle et al., 2022	<p>ADULTS: 28 with PDA. 24 self-reported; 4 diagnosed by a doctor.</p> <p>All participants: 220 participants (n = 167 women, n = 45 men, n = 5 other, n = 3 gender not reported; mean age = 32.14 years, SD = 11.28, range = 18–75).</p> <p>Self-reported diagnosis.</p> <p>Screened with EDA-QA</p>	<p>Y – comparison and control.</p> <p>Autism group. 35 self-reported; 21 diagnosed by a doctor.</p> <p>Control group: no diagnosis (n=112)</p>	<p>To explore relationships between autism traits, PDA traits, camouflaging, offending and victimisation.</p>	<p>QUANT: A cross-sectional online quantitative predictor–outcome study: outcome variables (self-reported victimisation and offending behaviour) and predictor variables (self-reported social camouflaging, autism and PDA traits and symptoms of depression and anxiety).</p> <p>Autism - RAADS-14 Camouflaging - CAT-Q Depression - The Patient Health Questionnaire-9 Anxiety - Generalised Anxiety Disorder Screener (GAD-7) JVQ (Hamby et al., 2005) – Adult Retrospective</p>	<p>Camouflaging could represent a component of the PDA profile.</p> <p>Autism traits do not increase the risk of victimisation; PDA traits significantly predicted victimisation.</p> <p>More mental health difficulties (Symptoms of depression and anxiety) predicted more autism and PDA traits, and which in turn predicted offending behaviour.</p>

No.	Study authors	Clinical (PDA) Group details & Diagnostic / screening measures used for PDA?	Control or comparison group used? Y/N?	Aim of study (paragraph)	Methodology used: Quant? Qual? Type of measures used?	Results: main outcome (concluding paragraph)
					<p>Questionnaire (JVQ-AR; Weiss and Fardella, 2018): A self-report questionnaire assessing the frequency of childhood victimisation.</p> <p>NVOBS (Thornton et al., 2013): A self-report questionnaire of violent and nonviolent offending occurring in the past year.</p>	
13	White et al., 2022	<p>ADULTS: Not taken from clinical or PDA population.</p> <p>Study 1: N=267; student sample. 64% female; Mage = 32.7 years, SD age = 13.4 years)</p> <p>Study 2: N=549; recruited through Prolific. 49% female, aged from 18 to 67 years (Mean age=35.5 years, SD age=11.8 years).</p>	N – within group design with traits of each measure	To examine the relative importance of autistic traits and anxiety in relation to demand avoidance traits in the adult general population	<p>QUANT: Online questionnaire.</p> <p>Study 1: Autism Spectrum Quotient (AQ-10, Allison et al., 2012).</p> <p>Study 2: Autism Spectrum Quotient (AQ-50; Baron-Cohen et al., 2001)</p> <p>Study 1&2: Depression, Anxiety and Stress Scale – 21 Items (DASS-21; Lovibond & Lovibond, 1995). Extreme Demand Avoidance Questionnaire – Adult version (EDA-QA; Egan et al., 2019)</p>	<p>Autistic traits were a weaker unique predictor of demand avoidance traits than anxiety.</p> <p>Autism makes a unique contribution to predicting EDA, consistent with the prevailing theory that EDA is a part of the autism spectrum.</p> <p>By accounting for depression and stress, the current study demonstrates there is a link between EDA and anxiety rather than with general emotional symptoms,</p>

2.4.7 *Synthesis method*

Because the included studies were few and methodologically heterogeneous, it was decided to synthesise data using a narrative and descriptive approach (Popay et al., 2006). For the current review, this constituted an iterative process, including developing a preliminary synthesis of findings from the included studies, exploring relationships in the data and assessing the robustness of the synthesis (Popay et al., 2006).

2.4.8 *Methodological rigour*

Methodological quality assessment for 13 studies included in the final data synthesis were independently evaluated by AJR and AKL using the Quality Assessment with Diverse Studies tool (QuADS; Harrison et al., 2021), a 13-item development of the Quality Assessment Tool for Studies with Diverse Designs (QATSDD; Sirriyeh et al., 2012) to evaluate the quality, strengths, limitations and unassessed areas within quantitative, qualitative, or mixed-method studies (for scores see Appendix B). The QuADS was chosen because it shows good reliability and validity, allows for analysis of different study types, factors in collaborative research with the population group, and promotes transparency in quality assessments (Harrison et al., 2021). Krippendorff's alpha test (Hayes & Krippendorff, 2007) was used to estimate the inter-rater reliability, which for the current raters was high ($\alpha = .95$). Consultation comparing results derived a consensus on scores.

Overall, studies provided clear theoretical frameworks with six identified as 'complete'. Clear aims were identified in the studies with eight scored as 'complete'. Only one study (O'Nions et al., 2018) failed to explicitly state their aims due to the wording describing what the study provides rather than being explicit in the study's actual aims. The studies utilized appropriate designs and appropriate sampling to address the stated aims with eight studies scored as 'complete'. Overall, studies utilized appropriate data collection tools, with seven

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studies using tools that are described as 'complete' for allowing detailed data to be gathered around all relevant issues required. A mixed methods paper (Brede et al., 2017) described adaptations made to the data collection tool to support children's attentiveness during administration. Trundle et al. (2022) describe their interview schedule as taken directly from the Educational Experiences Questionnaire (EE-Q). (Stuart et al., 2020) provide a description of their interview schedule and rationale for analysis undertaken. One qualitative study (O'Nions et al., 2018) reported some credibility processes explaining that the interview schedule had not been validated and is not presumed to assay behaviours that are necessarily specific to the PDA presentation. Clear recruitment data were provided including details around attrition rates with eight studies scored as 'complete' for this information. Overall, clear strengths and weaknesses were identified in the papers; however, none of the papers evidenced consideration of research stakeholders in the design or conduct of their research.

2.4.9 Community Involvement Statement

Both authors (AJR and AKL) have content-specific expertise in neurodevelopmental disorders, with the lead author's clinical work and clinical doctorate specialising in this. AKL has 20 years of experience working in neurodevelopmental research (with 50+ publications) and has published in scoping literature and meta-analyses. There was no further community involvement.

2.5 Results of Systematic Literature Review

2.5.1 Selection of sources of evidence

Of the thirteen papers, seven focused on the relationship between demand avoidance and anxiety; two considered how intolerance of uncertainty (IU) and/or sensory sensitivities (SS) relate to anxiety in pathological demand avoidance (PDA); seven considered how the relationship to anxiety may differ from PDA to other neurodiverse conditions; and seven

considered whether the relationship was moderated by age, gender, IQ or informant type (self-report vs proxy-report)?

2.5.2 Study / Participant(s) Characteristics

Study sizes were between 4 and 335 for questionnaire-based studies and 27 and 29 for each of the qualitative studies. Mixed methods study sizes ranged from 3 to 211. Of the thirteen studies, three focussed on the adult population (Trundle et al., 2022; White et al., 2022; Egan et al., 2020), nine centred on parent, carer or clinician report of child and adolescent samples, and one study (O’Nions et al. 2016) captured both with an aged range of 6-27 years. The studies focussed on the adult population had a female majority sample from 64-87%, whereas studies focussed on children had predominantly males. Only one study (O’Nions et al., 2018) had an equal gender split.

Six studies recruited people with PDA from an autistic population group, with one study (Truman et al., 2021) excluding participants (n=56; 21%) from the study due to not having an autism diagnosis. Four of these studies used the EDA-Q to confirm and validate the diagnosis of PDA, where two studies (O’Nions et al., 2021 & Truman et al., 2021) used the EDA-Q as a measure of ‘Extreme Demand Avoidance traits’ within the PDA sample. Four studies recruited from the population and allowed for participants to self-describe their diagnoses, of which one study (Egan et al., 2020) used the EDA-QA as a screening tool to confirm and validate the diagnosis of PDA, one study (Doyle & Kenny, 2023) did not use any screening tool to confirm or validate the diagnosis, and two studies used the EDA-QA as measures of ‘EDA traits’. The remaining three studies sampled directly from those identifying with PDA, with Stuart et al. (2020) using the EDA-Q as a measure of ‘Extreme Demand Avoidance behaviours’ within the PDA sample, Reilly et al. (2014) using the EDA-Q to confirm and validate the diagnosis of PDA, and Doyle & Kenny (2023) not employing a measure for either PDA or EDA traits. Ten of the studies screened for co-morbid

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neurodevelopmental diagnoses, all of which screened for autism. Six studies screened specifically for anxiety, another five screened for emotional and behaviour difficulties, and one study (Egan et al., 2020) screened for personality traits.

2.6 Results of studies

2.6.1 Measures

Of the eleven studies using quantitative or mixed methods designs, two studies did not use a screen for PDA (O’Nions et al., 2014 & Doyle & Kenny, 2023). All other studies provided justification for the measure used and reported on the measure’s reliability and validity. Stuart et al. (2020) used Confirmatory Factor Analysis on the measure and excluded two items from analysis after this; however, the justification for the modification of the scale in this study was unclear. Of the eleven studies, four described the use of the screening tool to confirm and validate the diagnosis of PDA, whilst the other seven studies used the measure to capture ‘PDA traits’ within a self-described PDA or non-PDA sample. As part of the sampling procedure for one of the qualitative studies (O’Nions et al., 2018) a shortened, 11-item version of the EDA-Q screen was used; however, it is unclear why this tool was shortened, and the justification as to the items chosen and those removed.

Of the eleven studies using quantitative or mixed methods designs, four studies (Brede et al., 2017; O’Nions et al., 2021; Egan et al., 2020 & Truman et al., 2021) did not screen for anxiety. Doyle & Kenny (2023) captured anxiety; however, this was not with a standardised screening tool. All other studies provided justification for the measure used and reported on the measure’s reliability and validity.

Other constructs that may help understand underlying factors in PDA were captured by four studies: Stuart et al. (2020) screened for intolerance of uncertainty; Egan et al. (2020) screened for personality traits and impulsivity; O’Nions et al. (2014) screened for conduct

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problems and callous unemotional traits; and O’Nions et al. (2021) captured emotional dysregulation.

All studies captured other neurodevelopmental diagnostic information in addition to PDA, with ten using a screening tool to confirm and validate the diagnosis of Autism, and three providing self/parent/carer-report alone. Within the ten studies there were eight different screening tools used showing a lack of consistency in the measure used in the PDA literature to capture autism traits. Three studies (Schneider et al., 2022; O’Nions et al., 2016; & Reilly et al., 2014) used a screening measure to collect information pertaining to ADHD, whilst one study (Doyle & Kenny, 2023) captured this with self-report and enquired as to whether this was self-diagnosed or diagnosed “by a doctor”. All studies using diagnostic screening tools provided justification for the measures used, reporting on reliability and validity.

Demographic characteristics of age and gender were gathered in all studies as was the informant type (e.g. self vs proxy report). IQ was captured in only one of the studies (Schneider et al., 2022) using a standardised measure with justification reporting on reliability and validity.

2.6.2 Aim 1: the relationship between anxiety and demand avoidance

Seven studies captured both anxiety and demand avoidance, with only one (Doyle & Kenny, 2023) not using standardised screening tools for this. Doyle & Kenny (2023) reported that of 119 parents reporting that the PDA profile described their child, 61 (51%) reported a presentation of extreme anxiety. All six studies measuring demand avoidance used the EDA-Q. Two of these used the EDA-Q to capture or validate PDA as a diagnosis, and four used the EDA-Q to capture ‘behavioural traits’ of demand avoidance.

Of the two studies using the EDA-Q as a screen for PDA, Schneider et al. (2022) captured PDA and anxiety both as predictors of school behaviour and quality of life (QoL), and Reilly et al. (2014) captured PDA and anxiety and to assess “neurobehavioural co-morbidities”

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associated with epilepsy. Neither study considered the relationship between PDA and anxiety; however, Schneider et al. (2022) differentiate the two findings, anxiety was associated with “worse time course of school adjustment” than PDA.

Of the four studies using the EDA-Q as a measure of ‘demand avoidance traits’, Brede et al. (2017) found “many children had clinically significant levels of anxiety and behaviours reminiscent of EDA”; however, did not consider the relationship between demand avoidance and anxiety. Stuart et al. (2020) found that anxiety predicts demand avoidance behaviours in children with PDA. Trundle et al. (2022) found that anxiety predicted “more autism and PDA traits” in children, and White et al. (2022) found anxiety predicts demand avoidance traits in children and adults.

2.6.3 Aim 2: IU and/or SS related to anxiety in PDA

Two papers captured IU and/or SS related to anxiety in PDA. Stuart et al. (2020) found anxiety to be a predictor of demand avoidance behaviours in children. They also found IU to moderate this effect and be a bigger predictor of demand avoidance behaviours than anxiety alone. Further analysis found variation in the individual contributions of IU and anxiety according to the type of EDA behaviour: ‘control’ behaviour seemingly directly influenced by IU; ‘fantasy’ behaviour appearing to be the result of a combined effect of IU and anxiety; and ‘meltdown’ behaviour being influenced by IU with a mediation effect via anxiety. Doyle & Kenny (2023) reported an interplay between sensory issues, of which 81% reported problems and severe anxiety, of which 78% of a PDA sample reported problems; however, this relationship was not described or explained beyond reported prevalence rates.

2.6.4 Aim 3: Comparing anxiety in PDA to other neurodiverse conditions

Schneider et al. (2022) reported that 19% of autistic children in their study fulfilled the criteria of a PDA syndrome as assessed by the EDA-Q. Reporting on “school adjustment”,

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they reported that severe autism was associated with “a better evolution of school adjustment”, anxiety was associated with “a worse time course of school adjustment”, and PDA diagnosis was “not associated with any adjustment at school”. Trundle et al. (2022) differentiated PDA and autism, finding that PDA traits significantly predicted victimisation whereas autism traits did not, and that anxiety predicted both autism and PDA traits. White et al. (2023) found that autism makes a unique contribution to predicting EDA, which they report is consistent with the prevailing theory that PDA is a “part of the autism spectrum”. Truman et al. (2021) found that autistic people who do not identify with the diagnosis of PDA score lower on the EDA-Q than those who do identify with the PDA diagnosis. Reilly et al. (2014) found that the four children meeting the criteria for PDA in their study also met the criteria for ADHD. O’Nions et al. (2014) found that those with PDA had significantly higher levels of emotional symptoms including anxiety than either the autism group or those with behavioural conduct problems. Egan et al. (2020), reporting on individual differences in personality, found that individual differences underlie self-reported PDA but not autism, with PDA having three significant independent predictors of ADHD, low emotional stability, and antagonism. O’Nions et al. (2016) report that “apparently manipulative behaviour, difficulties with other people, harassment of others, fantasising, lying, cheating, stealing and socially shocking behaviour” are features that differentiate PDA from autism.

2.6.5 Aim 4: the effects of age, gender, IQ or informant type

Brede et al. (2017) found discrepancies in informant type with parents reported elevated levels of demand avoidant behaviour on the EDA-Q, presumably observed at home, whereas teachers did not. Doyle & Kenny (2023) reported that sensory and anxiety issues were more prevalent among younger children with PDA in their study suggesting an effect of age. Moreover, O’Nions et al., 2021 reported in the development of a short version of the EDA-Q

that eight items were found to be discriminating indices of EDA traits and behave similarly with respect to quantifying EDA, irrespective of child's age or gender.

2.7 Discussion

2.7.1 *Pathological / Extreme demand avoidance and anxiety*

Current understanding of underlying factors contributing to anxiety in those with PDA are scarce. Consistent with PDA being an unrecognised diagnosis within manuals, most of the research focusses on those who *identify* with PDA either for themselves or by proxy, which is most often a parent of a child whom they describe as having PDA. Most research in the PDA literature uses the EDA-Q to capture the construct; however, use of this tool is varied. Some research uses it to validate the diagnosis of PDA, some use it to screen for cut-off scores to group those with PDA and those who do not meet the criteria, and some use it to refer only to the behavioural outcomes being observed. Some researchers investigating PDA conceptualise the diagnosis as part of autism and therefore also exclude those who do not meet the criteria for autism from their research. Furthermore, with varied use of the EDA-Q, some research has amended the scale within their research based on preliminary data analyses, which further causes inconsistencies in the literature. This has meant that, of the research investigating anxiety in the PDA population, some describe those identifying with PDA as having high levels of anxiety, whereas some research describes high levels of anxiety as causing an increase in demand avoidance behaviours. This highlights the need for clarity on language and conceptual frameworks regarding the relationship between anxiety, PDA and demand avoidance behaviours, differentiating between the underlying neurodevelopmental diagnosis of PDA being researched, and the observable demand avoidance behaviours / traits that are often referred to as EDA.

2.7.2 *Intolerance of Uncertainty (IU) and Sensory Sensitivities (SS)*

Intolerance of uncertainty (IU) was found to be a significant construct related to anxiety in PDA. Two papers captured this relationship; however, only one used a standardised measurement tool. Both papers investigating IU referred to this construct being investigated as it is known to be relevant in the autism literature. The same research using non-standardised measurement tools found sensory sensitivities (SS) to be a relevant construct to those identifying with PDA; however, this is yet to be investigated with a structured research tool. Additionally, the nature of the relationship of SS to anxiety is yet to be explored beyond prevalence figures.

2.7.3 *PDA and other diagnostic constructs*

Much of the research in PDA also screens for autism and has found discriminations from what is understood about anxiety in autism. Where autism research suggests only an indirect link between autism and anxiety mediated through IU, Stuart et al. (2020) suggest PDA has a direct relationship with anxiety as well as being mediated through IU. Another diagnosis often captured in PDA research is attention deficit hyperactivity disorder (ADHD), with some research finding all their sample that met the criteria for PDA also met the criteria for ADHD. It is unclear whether this represents diagnostic overshadowing or whether heightened anxiety may have been presenting as 'hyperactivity' within these research papers. Furthermore, some research has helped to develop an understanding of anxiety in PDA as both distinct from autism and beyond that of behavioural traits in their findings that anxiety was higher in those with PDA than either those with autism or those with conduct problems.

2.8 Clinical Implications

It is recommended that to support consistency in future PDA research, clear definitions are needed to separate out 'demand avoidance' in the general population, 'extreme demand

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avoidance (EDA)' in the general population, and 'pathological demand avoidance (PDA)' as an underlying neurodevelopmental diagnosis. Further research into PDA could help make sense of its relationship to autism, and how other constructs known to be relevant in autism literature, such as IU and SS, could moderate the relationship between PDA and anxiety. This would help develop our understanding of the PDA presentation, the overlapping presentations and/or symptoms with other diagnoses such as autism, and guide what interventions in autism are more and/or less relevant when treating someone with PDA.

2.9 Conclusion

The inconsistency in terminology within PDA research means that it is difficult to provide clarity of overlapping or disorder specific effects of PDA related to other constructs of interest such as autism, anxiety, intolerance of uncertainty, or sensory sensitivities. There has been some evidence of underlying relationships contributing to anxiety such as IU, which is consistent with the autism literature; however, no studies have investigated the role of sensory sensitivities, which is known to be a significant factor in understanding anxiety in autistic people. Future research should consider the role of sensory sensitivities to those who identify with PDA.

2.10 Rationale for Present Study

Stuart et al. (2020) found that anxiety and intolerance of uncertainty (IU) significantly predicted EDA behaviours in children with diagnosed or probable PDA. It is suggested that affected children use EDA behaviours to increase certainty and to alleviate anxiety. Wigham et al. (2015) reported a moderate positive correlation between scores of IU and sensory over-responsiveness in children living with autism. Furthermore, IU has been shown relevant to the construct of sensory sensitivities in children with and without autism, even when controlling for anxiety (Neil et al., 2016). The relationship between IU and sensory

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sensitivity has yet to be addressed with PDA and may differentiate those with PDA from those with autism.

The clinical implications of the proposed research study concern three main areas: The label of PDA; core symptoms; and potential treatment - clinical and educational. Pathological demand avoidance is mainly characterised by a tendency to avoid everyday demands and expectations to an extreme extent. While avoiding everyday demands are seen as a key marker, hence the diagnostic label, evidence suggests that, in autism, uncertainty to change relates to having higher sensory needs. It is quite possible this group of children presenting with demand avoidance behaviours show more extreme sensory sensitivity that have yet to be identified compared to those with autism, and that this should be the key focus of diagnosis and treatment. This could shape schools' strategies working with this population and help make sense of the terminology around demand avoidance; to consider whether PDA requires a new label that pertains to a more sensory oriented sub-type of autism.

2.11 Aims and Research Questions / Hypotheses

The current study proposes to extend the work of Stuart et al. (2020); of examining the relationship between intolerance of uncertainty (IU) and anxiety in children with PDA. We propose to expand this study to investigate the relationship between autistic traits and extreme demand avoidance behaviours (EDA), in addition to exploring the contributing roles of both IU and SS to levels of anxiety.

Specifically, our study has five aims: 1) to describe differences between overall scores on each variable (autistic traits, EDA behaviours, IU, SS and anxiety) between children with different diagnoses including PDA and autism; 2) to examine the relationships between each of the variables (autistic traits, EDA behaviours, IU, SS and anxiety) for the whole sample; 3) to test the mediating roles of IU and SS in the relationship of EDA traits and anxiety; 4) to test a Structural Equation Model incorporating relationships between all five variables; and 5)

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to explore any potential areas of relative strength in those identifying with the diagnosis of PDA.

CHAPTER 3: METHOD

3.1 Research Design

Using a quantitative, between groups design, an online survey was used to explore relationships between, demand avoidance, anxiety, sensory sensitivities, and intolerance of uncertainty in children with and without a diagnosis of autism. The online survey consists primarily of quantitative standardised questionnaires (see ‘measures’ section below) and supplemented by open ended questions at the end providing opportunities for qualitative data to be captured.

3.2 Ethics

Ethical approval for this research project was granted by the University of Hertfordshire’s Health, Science, Engineering and Technology Ethics Committee with Delegated Authority. The protocol number is: aLMS/PGR/UH/05062(2) (see Appendix C). This project did not require NHS ethics as recruitment was conducted via the UK school system and third-sector organisations. Participants volunteered to take part and informed consent was obtained from all participants before any data was collected. Data was collected via Qualtrics; a secure, online survey hosting site approved by the University’s ethics committee. In compliance with General Data Protection Regulation (GDPR) under the Data Protection Act 2018 (DPA, 2018) data collected for analysis was stored on an encrypted, password protected computer, and was anonymised with participants requested to provide a unique anonymity code in the case of requests to remove data from the study. Participants were informed they could remove themselves from the survey at any time and were provided with the researcher and primary supervisor’s email addresses to contact in case of requests to remove their data from the study. The participant information sheet and consent form containing these details can be found in Appendix D and E respectively. Additionally, to remain inclusive wherever possible, with consideration as to the potential experience of selecting ‘Other’ in

demographics questions, we provided open text boxes for participants to identify with diagnoses and ethnicity when describing their child. Furthermore, we chose to leave all questions in the survey as optional, with no forced responses. Original ethical approval was granted, and updated ethical approval was sought and obtained as required before any changes were made to the survey, such as following consultation and advice from Experts by Experience (see section below).

3.3 Participants

Participants in the study were parents or carers of children aged four to seventeen who completed the parent rated measures about their children on whom the data was based. There were no other inclusion / exclusion criteria.

Power calculations were conducted to infer an appropriate sample size for this study. A priori power analysis was conducted using G*power version 3.1.9.7 (Faul et al., 2007) to determine minimum sample size required to test the main study hypothesis addressing the relationship between demand avoidance behaviours and overall anxiety in children with PDA. Results indicated a sample size to achieve 80% power for detection of a medium effect at a significance criterion of $\alpha = .05$ was $N = 55$ ($F = 4.02$) (see Appendix Q).

The study recruited 899 participants, with 96 removed from analysis for not having completed at least one full scale, and a further 8 removed for not meeting inclusion/exclusion criteria such as their child being older than 18 or younger than 4 years old. As such, the final count of participants included in the analysis were parents of 795 children between the ages of 4-17 years old ($M = 11$ years, 0 months; $SD = 3$ years, 5 months). The number of participants exceeded the requirements from power analysis and the expectations of the researchers after the PDA Society published the research online via their social media (see Procedure section below). The primary researcher and principal supervisor reflected on data requirements and the ethics of withdrawing a study from an under-represented group who

could be having the potential for their voices to be heard in research and concluded that the ethical approach would be to allow those who wish to complete the study to do so, and keep the study open for the intended period of time. Parents assigned their child to one of five diagnostic groups: 55 identified their child with the diagnosis of PDA (without autism), 171 identified their child as autistic (without PDA), 475 identified their child as having dual diagnosis (a PDA profile of autism), 51 identified their child as neurodiverse (without PDA and/or autism), and 43 allocated their child to ‘none of the above’. Using a cut-off score of 17 (Posserud et al., 2009), 699 scored their child above the threshold for autism on the ASSQ, and using a cut-of scores of 45 for those below 12-years-old and 50 for those above 12-years-old (O’Nions et al., 2014), 511 scored their child above the threshold for PDA on the EDA-Q. Of these, 489 scored their child above the threshold for both autism and PDA (see Appendix Q).

3.4 Procedure

3.4.1 Recruitment

The study utilized a purposive non-probability sampling method whereby participants were recruited via opportunity sampling and snowballing recruitment techniques. Recruitment was undertaken through the UK school system, a national third-sector organisation supporting children with PDA, social media, and word of mouth. The study was promoted online using a variety of non-NHS platforms. The (anonymised) advertisement poster for the study can be found in Appendix F. The poster included information about the option for participants to be entered into an anonymous raffle from which a ‘winner’ would be selected to win a £50 voucher. Details of the procedure for randomly selecting the prize winner on 16th April 2023 at random are provided in Appendix G.

A Special Educational Needs (SEN) for children aged 8-18 years old situated in the Midlands, UK in which the author had previously worked was contacted and provided

information about the study to share with students' parents (see Appendix H for information shared). In addition, the Behaviour Co-ordinator at the school shared the information with his colleague who posted on a Facebook group she was a member of named 'Small Talk Speech and Language Therapy'. National third sector organisations (Autism Bedfordshire; Autism Anglia; The PDA Society; and The National Autistic Society) were contacted via email (see Appendix I for an example email), and members of The PDA Society began a dialogue with us, which supported the development of our research project, after which they supported with recruitment through their website and social media presence. The primary supervisor of the project also advertised on Twitter.

3.4.2 Consultation with Experts by Experience

As it is recognised that this is an under-research population group, this project sought opportunities for a research 'stream' to be implemented around this study. Therefore, we included at the end of the survey an option for participants to opt-in for a potential follow-up study (outside of the remit of this thesis) which would involve an interview about PDA. We worked with Experts By Experience (EBE) to help develop the study at initial stages to support our ethic of continual openness to feedback and reflection to implement changes where appropriate.

3.4.2.1 Special Educational Needs School

EBE consultation began with support we received from the Principal and Behaviour Co-ordinator and Deputy Head Teacher. We contacted the school via email who agreed to support the study by offering participation to the parents of their students. Prior to 'publishing' the survey on Qualtrics, the team at the school checked the survey for usability across multiple devices such as phones and desktop computers. They checked the appropriateness of length of survey, the readability of the content, the appropriateness of

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terminology, and the usability of the functions (such as the buttons to move to the next question etc.).

3.4.2.2 The PDA Society

As part of our strategy of contacting external organisations to request support in advertising the study, we contacted the PDA Society. Our communications with the PDA society included email exchanges and an online video conference meeting between them, myself, and my primary supervisor. We were also directed to resources to help provide clarity over definitions of some of the terminology in the literature related to ‘demand avoidance’ that is often used interchangeably. This led to us re-wording the title of the survey, re-wording various aspects of the information sheet including the addition of signposting to suggested support networks, including a note before the ASSQ (see Appendix J) related to the terminology used in this standardised questionnaire, and updating the debrief, again to include the additional signposting. The PDA Society published our poster (see Appendix F for the poster) on their social media sites and remained in close contact throughout to provide us with the opportunity to respond to feedback from the community.

3.4.2.3 The PDA Community

The PDA Society monitored feedback in the comments sections of its social media posts and fed what they felt important back to us via email. One of the comments we construed as a request for further research with adults who identify with the diagnosis of PDA. Therefore, we developed, in collaboration with the PDA Society, another survey for adults based on the template of this study. The adults’ survey is not within the remit of this thesis; however, is part of the ‘research stream’ that was intended to be promoted to explore the needs within this underrepresented population group.

3.5 Measures

Participants were asked to provide demographic information including their child's age, gender, ethnicity, type of schooling, and which diagnoses, if any, best described their child. Open questions related to PDA were provided at the end of the survey. The following standardised questionnaires were used to capture the constructs of autistic traits, demand avoidance, anxiety, sensory sensitivities, and intolerance of uncertainty:

3.5.1 *The Autism Spectrum Screening Questionnaire (ASSQ)*

Developed from the Asperger's Syndrome and High-Functioning Autism Screening Questionnaire (Ehlers & Gillberg, 1993), the ASSQ (Ehlers et al., 1999) consists of 27 items / statements scored using a 3-point Likert scale, with potential responses being: 'not true' (0 points); 'somewhat true' (1 point); and 'certainly true' (2 points). Responses from each item are summed to produce a total score ranging from 0 to 54, with higher scores indicating higher degree of Autistic symptomology. The scale has reported test-retest reliability of 0.90-0.94. The ASSQ is one of the most widely used autism screening tools in the world with cut off scores varying across populations. For example, suggest Ehlers et al. (1999) suggest that parent rated scores of 18 or above indicate high likelihood of their child meeting the threshold for an autism or Asperger's syndrome diagnosis, with a specificity of 0.90, and sensitivity for this cut off was 0.62. However, in Norway, the scale has reported sensitivity of 0.91 and specificity of 0.86 when a cut-off score of 17 was used in a general population sample (Posserud et al., 2009). Mattila et al. (2009) suggest that a valid cut-off score for parents' single score cannot be estimated; however, can be useful as a screening tool for research rather than a clinical diagnostic tool. The ASSQ showed a good degree of internal consistency ($\alpha=.869$).

3.5.2 *The Extreme Demand Avoidance Questionnaire (EDA-Q)*

The EDA-Q (O’Nions et al., 2014) is a 26-item parent report measure of demand avoidance traits in their child. The EDA-Q was developed as a checklist to quantify behaviours reported in the clinical accounts of pathological demand avoidance (PDA) as described by Newson et al. (2003). The EDA-Q aims to provide an opportunity to identify the PDA profile in a consistent way for research purposes (rather than a diagnostic tool). Each item is rated on a four-point Likert scale of how true a behavioural statement represents their child over the past six months: 0 (not true); 1 (somewhat true); 2 (mostly true); and 3 (very true). Item scores (items 14 and 20 are reverse scored) are summed to give a total ranging from 0 to 78.

(O’Nions et al., 2014) suggest a cut-off score of 50 in children aged 5-11 years old, and a score of 45 in children aged 12-17 years old would indicate elevated risk of parents reporting that the child had been clinically identified as having a profile resembling PDA. However, as it is suggested that as there is no diagnostic algorithm for PDA, it is not possible to objectively assess the validity of the measure or the suggested cut-offs. However, it could be useful to consider the EDA-Q scores dimensionally. The suggested eight dimensions in the scale are ‘avoiding demands and social manipulation’ for the purposes of: avoidance / controlling interactions (6 items); insensitivity to hierarchy praise/reputation with peers (6 items); emotional lability in response to demands or perceived pressure (4 items); need for control (2 items); lack of responsibility/blaming (2 items); mimicry and role play (4 items) distractedness (1 item); and passivity (1 item), with high scores indicating ‘high EDA traits’. The EDA-Q showed a good degree of internal consistency ($\alpha=.880$).

3.5.3 *Spence Children’s Anxiety Scale – Parent Version (SCAS-P)*

The SCAS-P (Nauta et al., 2004) is a 38-item parent report measure of their child’s anxiety, adapted from the original child version of the scale (Spence, 1997). Compared to the child version, parent–child agreement ranged from 0.41 to 0.66 in an anxiety-disordered

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group, and from 0.23 to 0.60 in a control group. The SCAS-P scores represent observable child behaviours based on their frequency. Respondents rate each item on a four-point Likert scale: 0 (Never), 1 (Sometimes), 2 (Often) and 3 (Always). Items are summed to create a total score ranging from 0 to 114, with higher scores reflecting greater levels of symptoms. Confirmatory factor analysis provided support for six inter-correlated factors that corresponded with the child self-report measure as well as the classification of anxiety disorders by DSM-IV (American Psychiatric Association, 2000) at the time. These six inter-correlated factors are: 1) Panic attack and agoraphobia; 2) Separation anxiety; 3) Physical injury fears; 4) Social phobia; 5) Obsessive compulsive; 6) Generalized anxiety disorder / overanxious disorder. The SCAS-P showed a good degree of internal consistency ($\alpha=.933$), which also applied to the sub-scales ($\alpha=.866$; $\alpha=.785$; $\alpha=.567$; $\alpha=.802$; $\alpha=.834$; $\alpha=.790$ respectively).

3.5.4 Short Sensory Profile (SSP)

The SSP (McIntosh et al., 1999) is a shortened, 38-item adaptation of the Sensory Profile (SP; Dunn, 1999) representing caregiver report of children's behavioural sensory processing that is scored on a five-point Likert scale ranging from 1 ('Always') to 5 ('Never') based on frequency. Scores are summed giving a range of 38 to 190, with lower scores indicating greater levels of sensory sensitivities. The SSP is made up of seven subscales: 1) Tactile Sensitivity (seven items); 2) Taste/Smell Sensitivity (four items); 3) Movement Sensitivity (three items); 4) Underresponsive/Seeks Sensation (seven items); 5) Auditory Filtering (six items); 6) Low Energy/Weak (six items); and 7) Visual/Auditory Sensitivity (five items). Part of the development of the SSP included the removal of items in the SP that were related to social-communication and motor items. Thus, the SSP isolates sensory sensitivities that are less confounded by items overlapping with the diagnostic features of autism as described in the DSM-V, making the SSP ideal as a research tool within this population. The scale has

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shown good convergent validity with physiological measures and a discriminant validity of >95% in distinguishing children with and without sensory modulation difficulties. The SSP showed a good degree of internal consistency ($\alpha=.887$), as did the sub-sections within the scale ranging from 0.697 to 0.943.

3.5.5 Intolerance of Uncertainty Scale – Parent (IUS-P)

The IUS-P (Comer et al., 2009) is a 27-item parent report measure of their child's tendency to react negatively on an emotional, cognitive, and behavioural level to uncertain situations and events. Respondents rate the extent to which the statement in each item is like their child on a five-point Likert scale from: 1 (Not at all); to 3 (Somewhat); to 5 (Very Much). The scale has been shown previously to provide good internal consistency ($\alpha = 0.97$; Cornacchio et al. (2017)). The IUS-P, developed in conjunction with the Intolerance of Uncertainty Scale–Child (IUS-C), was adapted from the adult version of the measure, the IUS (Freston et al., 1994) which was found to have good psychometric properties in general population cohorts (Buhr & Dugas, 2002). All items in the IUS-P are directly parallel to the items on the IUS. Of note, where the IUS-C incorporated language to give the scale “child compatibility”, such as the replacement of the phrase “I can't stand...” to “I don't like...”, the IUS-P retains the language of the original (e.g. “My child can't stand...”) meaning the IUS-P retains closer resemblance to the original IUS and it's good psychometric properties. The IUS-P showed a good degree of internal consistency ($\alpha=.954$).

3.6 Data Cleaning

Prior to data analysis, the data was downloaded from Qualtrics and stored securely on an encrypted hard drive which was only accessible by the primary researcher. Data was initially ‘cleaned’ to ensure it was prepared to be analysed using quantitative methods. Cleaning was undertaken with use of Microsoft Excel and predominantly included converting text

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responses to uniform data, including sections on ‘Ethnicity’, ‘Age of Child’, ‘Relationship to Child’ and ‘Diagnosis’.

3.6.1 Ethnicity

As mentioned, for ethical reasons, we decided to leave the ‘Ethnicity’ question as an open text box rather than options to avoid the situation of a participant potentially having to tick an ‘Other’ option to describe this part of their identity. Concerns have already been raised in the literature regarding the socio-political implications of using single-item measures to capture ethnic groups (Aspinall, 2002), and the methodological difficulties of using measures to capture the nature of ethnicity as an individual subjective experience vs measures which derive from supposedly valid and reliable categories such as those captured via Office for National Statistics (ONS, 2022) census data (Williams & Husk, 2013).

3.6.2 Age

Child’s Age was provided in various formats (e.g. ‘11 years and 4 months’ or ‘11y 4m’ or ‘11.4’ etc). This involved the use of the ‘Ctrl E’ function to autofill responses, and then manually checking through each data point for accuracy, which was particularly required for responses that were provided in a unique format such as ‘June 2009’. Participants not meeting the inclusion criteria of between 4 and 17 were then able to be identified calculating the range of ages, and these were removed from the data prior to analysis.

3.6.3 Relationship to child

Relationship to child required use of the ‘unique’ function in Excel to identify all the unique responses and find those that could be clustered together. For example, where some had written ‘Mother’ and others ‘Mum’, these were combined as one group.

3.6.4 Diagnosis

There were two demographic questions related to diagnoses. One was a selection between the five groups: PDA, autism, dual diagnosis (PDA and autism), neurodiverse, and none of the above. In addition, there was an open text box for participants to write their child's diagnoses. Firstly, the different written forms of diagnostic constructs (autism spectrum, ASD, ASC etc.) were compiled into one unifying term. Participants were then grouped based on their combined responses to the two questions. For example, if a participant had selected PDA in the first question (not the dual diagnosis) but had written autism in the text response to the second question, then they would be allocated to the dual diagnosis group.

3.7 Statistical Analyses

IBM's SPSS Statistics package, a commonly used statistical tool, was accessed when analysing the data. The lavaan package (Rosseel, 2012) in R version 4.2.1 (R Core Team, 2021) was also accessed to support mediation and structural equation modelling (SEM) of the data. Data from the whole sample was analysed, as well as being collated and grouped according to diagnostic group which allowed for specific analysis of those who identified with the diagnosis of PDA also so as to explore strengths in this population. Tests of distributions to check for deviations from normality were applied to determine whether relevant parametric or non-parametric tests were to be conducted. Details of specific tests to explore each of the aims of the study are described below:

Aim 1: to describe differences between overall scores on each variable (autistic traits, EDA behaviours, IU, SS and anxiety) between children with different diagnoses including PDA and autism

Descriptive statistics providing the means and standard deviations for each scale split by diagnostic group will be calculated and presented. An overview of differences in mean scale scores between diagnostic groups will then be described.

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Aim 2: to examine the relationships between the variables (autistic traits, EDA behaviours, IU, SS and anxiety) for the whole sample

A correlation matrix of total scores of each scale will be presented, along with descriptions of the significance and strength of the relationships.

Aim 3: to test a Mediation model of EDA predicting anxiety

As previous literature (Stuart et al, 2020) suggested use of a mediation model in making sense of IU and anxiety in PDA, we will undertake mediation modelling on the whole sample to also investigate the any potential mediating role of SS in the model.

Aim 4: to test a Structural Equation Model (SEM) of EDA predicting anxiety

Building on the literature suggesting a) a relationship between PDA and autism, and b) IU and SS have a role of predicting anxiety in autism, Structural Equation Modelling (SEM) will be undertaken on the whole sample to see how well a model of EDA and autistic traits predicting anxiety fits the data.

Aim 5: to investigate any potential areas of relative strength in those identifying with PDA

This part of the analysis will be undertaken only with those identifying with the diagnosis of PDA. A repeated measures ANOVA will be undertaken on the subscales of each measure (the IU-P does not have subscales and therefore will be omitted from this part of analysis) to identify whether there are differences between subscales for each construct. If differences are found, a paired samples t-tests with Bonferonni correction will be undertaken to analyse where differences between subscales occur. This will also allow us to identify areas of relative strength in those identifying with PDA which is lacking in the literature to date concerning this population group.

CHAPTER 4: RESULTS

4.1 Descriptive statistics

To identify differences in scores for each variable across diagnostic groups, means and standard deviations for each scale across the four self-identified diagnostic groups are presented in table 4 below:

Table 4: Means and SD's of all scales across diagnostic groups

Construct Scale	Diagnostic group Mean (SD)			
	PDA (n=530)	Autism (n=171)	Neurodiverse (n=51)	None of the above (n=43)
ASSQ	28.45 (9.53)	28.37 (9.12)	24.04 (9.56)	20.16 (11.32)
EDA-Q	53.22 (11.12)	45.16 (13.22)	43.94 (15.88)	38.42 (18.36)
SCAS-P	48.07 (20.07)	47.08 (21.78)	41.59 (20.76)	34.67 (18.85)
IU-P	91.12 (22.90)	90.37 (24.26)	82.61 (25.78)	73.28 (24.87)
SSP	101.66 (20.82)	101.98 (20.96)	112.16 (18.12)	123.77 (25.56)

ASSQ scores were higher in those identifying with PDA and Autism than Neurodiverse or None of the above. EDA-Q scores were higher in those identifying with PDA than those identifying with Autism, Neurodiverse or None of the above. SCAS-P scores were higher in those with PDA and Autism than Neurodiverse and None of the above. IU-P scores were higher in those with PDA and Autism than those with Neurodiverse or None of the above. SSP scores were lower (lower scores on the SSP mean higher sensory needs) in the PDA and Autism groups than those in Neurodiverse or None of the above.

4.2 Scale Correlations

A series of Pearson correlations were conducted to assess the relationship between each of the five constructs, as shown in Table 5 below.

Table 5: Correlations of total scores from all five measures

	ASSQ	EDA-Q	SCAS-P	IU-P	SSP
ASSQ	1	.61**	.37**	.38**	-.60**
EDA-Q	-	1	.39**	.36**	-.48**
SCAS-P	-	-	1	.63**	-.46**
IU-P	-	-	-	1	-.43**
SSP	-	-	-	-	1

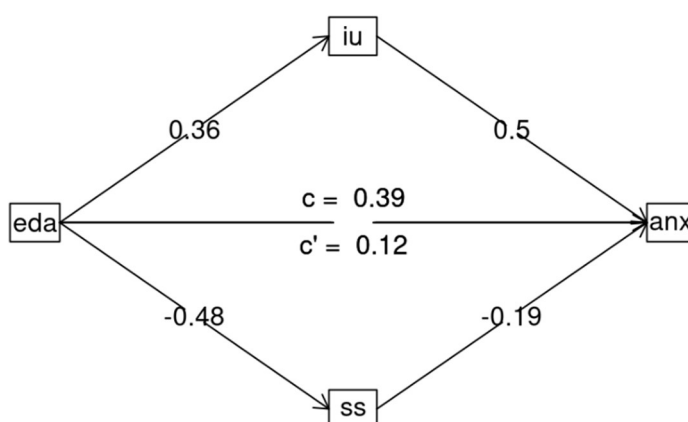
** Correlation is significant at the 0.001 level (2-tailed).

All scales were significantly positively correlated with each other. Lower scores on the SSP mean higher sensory needs, and therefore negative numbers translate to positive correlations. As predicted, autistic traits and sensory needs were highly correlated, and IU and anxiety were highly correlated, each with correlation coefficients above 0.6. In addition, autistic traits and EDA were highly correlated.

4.3 Mediation modelling

The parallel mediating effects of both IU and SS on the relationship between EDA traits and anxiety was tested. The model tested is presented in Figure 2 below along with unstandardised path coefficients:

Figure 2: Parallel Mediation Model



Regression analysis showed the overall model was significant ($F(3,791) = 214.42, p < .001, R^2 = .45$) and explained 45% of the variance in anxiety scores. EDA emerged as a significant

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predictor of anxiety, explaining 12% of the variance; as did IU, explaining 50% of the variance; and SS explaining 19% of the variance. Consistent with Hayes' (2013) approach, the direct effect from EDA traits to anxiety, the indirect effect through IU (EDA traits → IU → Anxiety) and SS (EDA traits → SS → Anxiety) and the total effects were estimated for the whole sample and are presented in Table 6 below.

Table 6: IU and SS mediating the relationship between EDA and Anxiety

			95% Confidence Intervals (CI)		
Mediation effect		Unstandardised coefficient	Standard error (SE)	Lower limit	Upper Limit
Direct effect (c')	Removing effects of IU+SS	0.12*	0.03		
Indirect effect (ab)	Through IU+SS	0.28*	0.02	0.23	0.32
Total effect (c)	Including effects of IU+SS	0.39*	0.03		

The mediation model shows a reduction on the direct effect of EDA on anxiety from .39 to .12 when the mediating effects of IU and SS are accounted for.

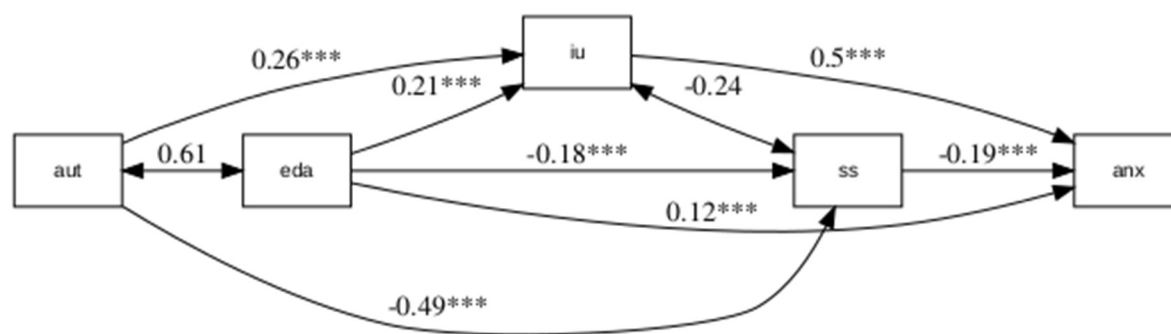
4.4 Structural Equation Modelling

The structural equation model tested in which autism and EDA traits predict anxiety demonstrated a good fit to the data: $\chi^2(1)=0.202, p=.653, cfi=1.000, gfi=1.000, nfi=.1.000, rmsa<.001, srmr=.002$), and model modifications did not improve the fitness or clarity of the model. The model fit indexes are presented in Table 7 and standardized path coefficients and their significance levels are presented in Figure 3 below:

Table 7: SEM model fit indexes

chisq	df	pvalue	gfi	cfi	nfi	rmsea	srmr
0.202	1.000	0.653	1.000	1.000	1.000	0.001	0.002

Figure 3: Structural Equation Model



Note: *** $p < .001$

Results performed using the lavaan package (Rosseel, 2012) in R (R core team, 2021) showed that Autism traits and EDA traits were highly correlated, with a covariance of .61 ($se=5.368, Z=14.765, p<.001$), and that intolerance of uncertainty and sensory sensitivities were also related with a covariance of .24 ($se=13.443, Z=-6.612, p<.001$). Autism traits had no direct relationship with anxiety ($-.036, se=.080, Z=-.443, p>.05$); however, EDA traits did have a significant direct relationship with anxiety (.12, $se=.054, Z=3.552, p<.001$). Autism traits had significant indirect effect on both intolerance of uncertainty (.26, $se=.100, Z=6.293, p<.001$) and sensory sensitivities (-.49, $se=.078, Z=-13.891, p<.001$). Similarly, EDA traits had a significant effect on both intolerance of uncertainty (.21, $se=.074, Z=5.032, p<.001$) and sensory sensitivities (-.18, $se=.058, Z=-5.085, p<.001$).

Of the predictors of anxiety, the strongest was intolerance of uncertainty (.50, $se=.026, Z=16.89, p<.001$), followed by sensory sensitivities (-.19, $se=.030, Z=-6.100, p<.001$).

Finally, the indirect effect of EDA on anxiety through intolerance to uncertainty was significant but the indirect effect through sensory sensitivities were not significant. Indirect effects of intolerance of uncertainty and sensory sensitives on the model are presented in Table 8 below:

Table 8: SEM effects of IU and SS

Variable	Estimate	Std.Err	z-value	P(> z)	Std.lv	Std.all
IU	0.342	0.057	6.011	0.000	0.342	0.219
SS	0.053	0.056	0.941	0.347	0.053	0.025

Of note, the structural equation model remained a good fit even when addressing the model in separate diagnostic groups (PDA, autism and neurodiverse) when was then tested for those identifying with the dual diagnosis of both PDA and autism.

In conclusion, there are a number of advantages of using the SEM model over the mediation model. Firstly, the Mediation model assumes the effect of Autistic traits is not relevant for IU and SS, suggesting that the only source of variation of these two variables is EDA; however, the SEM model shows Autistic traits have a significant effect on IU (.26, $Z=6.293$, $p<.001$) and SS (-.49, $Z=-13.891$, $p<.001$). Secondly, the SEM model accounts for the covariation between Autistic traits and EDA, which is theoretically crucial and the strongest relationship in the whole model (.61, $Z=14.77$, $p<.001$). Thirdly, the SEM model accounts for the covariation between IU and SS, which is important theoretically and the two variables are shown in the SEM to have a strong relationship (-.24, $Z=-6.612$, $p<.001$). Finally, and importantly, the results of the mediation model show a reduction on the direct effect of EDA traits on anxiety from .39 to .12; however, this final total effect is also presented in the SEM model. Furthermore, the SEM model adequately presents the indirect effects of each variable independently, showing IU is stronger (a better mediator) of anxiety (.22, $Z=6.01$, $p<.001$) than SS (.025, $Z=.94$, $p=.35$).

4.5 Strengths and difficulties within the PDA profile

This final analysis was carried out only with those who identified with the diagnosis of pathological demand avoidance (PDA). The data was separated and analysed in terms of subscales for each measure: the ASSQ, EDA-Q, SCAS-P and SSP (there are no subscales for

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the IU-P and therefore it is omitted from this part of the analysis). Means for each subscale are adjusted for the number of items in each. For the subscales of each of the measures, Mauchly's sphericity test was found to be significant, and therefore equal variances across subscales could not be assumed.

4.5.1 ASSQ

Table 9: Means and Standard Deviations of ASSQ subscales

	Mean (SD)
Social Interaction	1.07(.42)
Communication	1.23 (.46)
Restricted & Repetitive Behaviours	1.06 (.48)
Motor Clumsiness	.74 (.45)

A repeated measures ANOVA showed that the ASSQ scores differed across all subscales, $F(2.94, 530)=210.460, p<.001, \text{Eta}=.29$, Greenhouse-Geisser. Paired samples t-tests with Bonferonni correction set to .008 demonstrated that there was no significant difference between Social Interaction and Restricted & Repetitive Behaviours $t(529)=.848, p=.198$; however, there were significant differences in across all other subscales with a range of $t<31.88, p<.001$. Communication difficulties (mean=1.23) were identified as an area of significant concern, whereas Motor Clumsiness (mean=.74) were identified as an area of less concern when compared to other subscales of the ASSQ in this population.

4.5.2 EDA-Q

Table 10: Means and Standard Deviations of EDA-Q subscales

	Mean (SD)
avoidance / controlling interactions	2.14 (.57)
insensitivity to hierarchy praise/reputation with peers	2.03 (.53)
emotional lability in response to demands or perceived pressure	2.56 (.52)
need for control	2.62 (.55)

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lack of responsibility/blaming	2.30 (.64)
mimicry and role play	1.47 (.81)
Distractedness	1.95 (1.01)
Passivity	1.14 (1.13)

A repeated measures ANOVA showed that the EDA-Q scores differed across all subscales, $F(4.44, 530)=324.082, p<.001, \text{Eta}=.38$, Greenhouse-Geisser. Paired samples t-tests with Bonferonni correction set to .001 demonstrated that there was no significant difference between Insensitivity and Distractedness $t(529)=1.75, p=.040$, and Emotional Lability with Need for Control $t(529)=-2.52, p=.006$; however, there were significant differences in across all other subscales with a range of $t < 22.71, p < .001$. Need for Control (mean=2.62) and Emotional Lability in Response to Demands or Perceived Pressure (mean=2.56) are identified as areas of significant concern, whereas Passivity (mean=1.14) and Mimicry and Role Play (mean=1.47) were identified as areas of less concern when compared to other subscales of the EDA-Q in this population.

4.5.3 SCAS-P

Table 11: Means and Standard Deviations of SCAS-P subscales

	Mean (SD)
Panic attack and agoraphobia	.89 (.64)
Separation anxiety	1.72 (.76)
Physical injury fears	1.28 (.65)
Social phobia	1.64 (.79)
Obsessive compulsive	.83 (.67)
Generalized anxiety disorder	1.42 (.66)

A repeated measures ANOVA showed that the SCAS-P scores differed across all subscales, $F(4.16, 530)=296.71, p<.001, \text{Eta}=.36$, Greenhouse-Geisser. Paired samples t-tests with Bonferonni correction set to .003 demonstrated that there was no significant difference

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between Panic and Obsessive Compulsive, $t(529)=2.62, p=.004$, and Separation with Social Phobia $t(529)=2.25, p=.012$; however, there were significant differences in across all other subscales with a range of $t < 28.88, p < .001$. Separation Anxiety (mean=1.72) and Social Phobia (mean=1.64) were identified as areas of significant concern, whereas Obsessive Compulsive traits (mean=.83) and Panic (mean=.98) were identified as areas of less concern when compared to other subscales of the SCAS-P in this population.

4.5.4 SSP

Table 12: Means and Standard Deviations of SSP subscales

	Mean (SD)
Tactile Sensitivity	2.72 (.76)
Taste/Smell Sensitivity	2.02 (1.15)
Movement Sensitivity	3.65 (1.16)
Underresponsive/Seeks Sensation	2.86 (.98)
Auditory Filtering	2.08 (.70)
Low Energy/Weak	3.12 (1.20)
Visual/Auditory Sensitivity	2.48 (.91)

A repeated measures ANOVA showed that the SSP scores differed across all subscales, $F(4.74, 530)=222.427, p < .001, \eta^2=.30$, Greenhouse-Geisser. Paired samples t-tests with Bonferonni correction set to .002 demonstrated that there was no significant difference between Tactile and Under-responsiveness $t(529)=-2.78, p=.003$, Tactile with Low Energy $t(529)=.94, p=.173$, and Auditory with Visual $t(529)=.57, p=.286$; however, there were significant differences in across all other subscales with a range of $t(529) < 34.81, p < .001$. Lower scores on the SSP mean higher sensory needs, and therefore Taste/Smell Sensitivity (mean=2.02) and Auditory Filtering (mean=2.08) were identified as areas of significant concern, whereas Movement Sensitivities (mean=3.65) were identified as an area of less concern when compared to other subscales of the SSP in this population.

A summary of the combined areas of potential strengths from all subscales in those identifying with the diagnosis of PDA is presented in Table 13 below.

Table 13: Summary table presenting relative strengths of those identifying with PDA

Relative Strengths	Mean (SD)
Motor Clumsiness	.74 (.45)
Mimicry and role play	1.47 (.81)
Passivity	1.14 (1.13)
Panic attack and agoraphobia	.89 (.64)
Obsessive compulsive	.83 (.67)
Movement Sensitivity	3.65 (1.16)

We emphasise the importance of addressing the distribution of scores across the measures to highlight variations in the subscales. Importantly this has allowed us to highlight that those identifying with PDA have many strengths that differentiate them from those identifying with Autism without PDA, and it is crucial to emphasise the need for future studies to reflect some of the key strengths of PDA which is currently lacking in the existing literature.

4.6 Summary

Overall, there is no difference in autistic traits between autistic children and those identifying with PDA; however, there are higher EDA traits in those identifying with PDA than those who identify with autism. Using a SEM model of EDA in the context of autistic traits provided a better fit to the data from parents than a mediation model of EDA traits outside the context of Autism. These results suggest that PDA should be considered a distinct construct but within the context of its relationship to autism; ‘a PDA profile of autism’.

EDA traits alone contribute to anxiety directly, whereas autism traits do not. Autism traits have stronger path coefficients through both intolerance of uncertainty (IU) and sensory sensitivities (SS) than EDA traits. The SEM model remained a good fit to the data when applied to those identifying with the diagnosis of PDA.

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Overall, the lack of a significant relationship between autism traits and anxiety suggests that other factors such as EDA traits, IU and SS may help make sense of anxiety in individuals with autism. Furthermore, the importance of researching relative strengths in those identifying with the diagnosis of PDA is emphasised when making sense of this profile in order to move beyond a deficit model of understanding PDA.

CHAPTER 5: DISCUSSION

5.1 Overview

This study aimed to investigate the relationship between autistic traits and extreme demand avoidance (EDA), to exploring the contributing roles of both intolerance of uncertainty (IU) and sensory sensitivities (SS) to levels of anxiety, and to further develop an understanding of those who identify with the diagnosis of pathological demand avoidance (PDA). Parent-report measures were analysed using comparison of scale means, correlational analysis of the variables, structural equation modelling (SEM) and mediation modelling, and repeated measures ANOVA with paired sample t-tests of subscales, to make sense of the relationships between the variables. The results of this study will be discussed in relation to the wider literature on PDA and autism. Furthermore, implications for the theoretical understanding of PDA will be explored along with clinical implications for intervention with this population group. The strengths and limitations arising from the design and methodology of the study will be discussed, along with suggestions for potential future research before conclusions on the study are made.

5.2 Summary of findings

This study employed SEM, applied to all participants regardless of whether they identified with the diagnosis of PDA or not to make sense of the relationships between autistic traits, extreme demand avoidance (EDA), anxiety, intolerance of uncertainty (IU) and sensory sensitivities (SS). SEM analysis described a good model for understanding the relationships between these constructs.

As predicted and synonymous with the previous literature, the results suggested a good model of understanding the EDA-Q as a screening tool for PDA (O’Nions et al., 2014) and the ASSQ as a screening tool for autism (Ehlers et al., 1999). Autistic traits and EDA behaviours

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are very closely related to each other whilst also being distinct in their relationship to anxiety. For example, as the levels of autistic traits increased, there was found to be no direct impact on levels of anxiety; in contrast, as EDA traits increased, so did anxiety directly. For both autistic and EDA traits, as they increased, so did levels of anxiety *indirectly* through the moderating effects of both IU and SS, suggesting both IU and SS to have a key role in the levels of anxiety in PDA. Specifically, whilst autism traits were found to have a stronger relationship to anxiety through SS compared to IU, EDA traits were noted as having a stronger relationship to anxiety through IU compared with SS.

The model continued to show a good fit when applied to those who identify with the diagnosis of pathological demand avoidance, both for those who identified with and without the autism diagnosis.

5.3 EDA-Q and ASSQ

As expected, parents who identified their children with PDA, both with and without autism, showed significantly higher scores on the EDA-Q scale than those who identified their child with autism alone (i.e. without PDA), Neurodiverse, or those who selected ‘none of the above’ diagnostic categories. As expected, those who identified their child with the diagnosis of autism, whether with PDA or not, scored higher on the ASSQ than those who identified their child as either Neurodiverse or those who selected ‘None of the above’ diagnostic groups. This suggested an understanding of the ASSQ as a screening tool for Autism (Ehlers et al., 1999) and the EDA-Q as a screening tool for PDA (O’Nions et al., 2014).

Importantly, those who identified their child with PDA alone (and not with autism) also scored higher on the ASSQ than those who identified their child as Neurodiverse or ‘none of the above’ diagnostic categories. This suggests that understanding of the impact of autistic

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traits in those who identify with PDA, whether they identify with autism or not, could help make sense of their diagnostic and treatment needs.

5.4 Relationships to Anxiety

Consistent with Neil et al. (2016) who found the *direct* relationship between autistic traits and anxiety in autistic individuals to not be significant, this study also found the direct relationship between autistic traits and anxiety was not significant in those who identified with the diagnosis of PDA. The relationship between autistic traits and anxiety only became significant when moderated through either intolerance of uncertainty (IU) or sensory sensitivities (SS), whereas, akin to Stuart and colleagues (2020) findings, EDA traits were found to be *directly* related to anxiety as well as indirectly being moderated by IU and SS.

Consistent with previous research finding a relationship between IU and anxiety in the general population (Dugas et al., 2001) and in autistic individuals (Jenkinson et al., 2020), IU and Anxiety were found to be highly correlated in those who identified with the diagnosis of PDA. The findings from the current research were consistent with previous research finding that IU moderated the relationship between autistic traits and anxiety in autistic individuals (Neil et al., 2016; Boulter et al., 2014), finding that IU moderated the relationship between autistic traits and anxiety in those identifying with PDA. Furthermore, akin to Stuart et al. (2020), IU also moderated the relationship between extreme demand avoidance and anxiety in those identifying with the diagnosis of PDA.

Sensory sensitivities are known as an important underlying factor in the relationship between autism and anxiety (e.g. Boulter et al., 2014); however, no research to date has empirically investigated the role of SS in PDA. Consistent with previous research finding a relationship between SS and anxiety in the general population (Neil et al., 2016) and in autistic individuals (Hwang et al., 2020), the current study found SS to have a moderate positive

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correlation with anxiety in those identifying with PDA. Furthermore, the current research found an indirect effect of SS on both the relationship between autistic traits and anxiety, and the relationship between EDA traits and anxiety. Finally, compared to the moderating effect of IU, SS had a stronger moderating effect on the relationship between autistic traits and anxiety, where IU had a stronger moderating effect on EDA traits and anxiety.

5.5 The PDA profile

Analysis of the subscales of each measure was undertaken from the responses of those identifying with the diagnosis of PDA. This analysis aimed to develop an understanding of the profile of PDA beyond the total scale scores and identify more nuanced areas of concern and areas of less concern in this population.

5.5.1 *Autistic traits*

Autism is characterised by significant difficulties with social communication / interaction, and restricted, repetitive behaviours (American Psychiatric Association, 2013); however, where restrictive, repetitive behaviours have been identified as particular areas of concern for autistic individuals (Boulter et al., 2014; Joyce et al., 2017), analysis of subscales found communication difficulties to be more of a concern for those identifying with PDA as presented in Table 9 in the *Results* section above. Furthermore, motor coordination deficits are described as “a cardinal feature” of autism (Fournier et al., 2010), with Forti et al. (2011) suggesting that motor anomalies in autism might be determined either by a disruption in planning-control integration or by a limited planning process capacity. However, Motor Clumsiness was reported to be less of a concern for those identifying with PDA, suggesting potential strengths in the cognitive capacity of ‘planning’ within this group.

5.5.2 *Demand avoidance traits*

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Consistent with (PDA Society, 2018) survey in which 96% of the children were described as having “a need for control”, this was also reported as the most prominent ‘demand avoidance’ concern within the present study as presented in Table 10 in the *Results* section. With the ‘need for control’ subscale seemingly being of importance to those identifying with PDA, it is suggested that this subscale within the EDA-Q is expanded to be more than one item to explore this construct further. Egan et al. (2020) found EDA traits were predicted by emotional lability, and Mazefsky et al. (2013) described emotional lability being associated with poorer overall functioning in autistic individuals. Consistent with these findings, parents in the present study also reported emotional lability to be of significant concern. It is of note however, that mimicry and role play were seen as relatively less of a concern by parents identifying their children with PDA in this study, which could be a reflection of EDA-Q items being framed as deficits, whereas it has been suggested that mimicry and role can be useful strategies for individuals with PDA to cope as has been described anecdotally by parents of children with PDA e.g. Notes On PDA (2017): “*We’ve come to see role play and pretending to be in an imaginary world as natural coping instincts for certain things she finds difficult*”.

5.5.3 Anxiety

Where up to 50% of autistic individuals have been found to meet the criteria for social phobia (Spain et al., 2018), and separation anxiety has been found to be high in autistic children (Gillott et al., 2001), parents in the present study identifying their child with PDA also reported social phobia and separation anxiety as concerns for their children as presented in Table 11 in the *Results* section. However, where Gillott et al. (2001) also found that autistic children scored highly for obsessive compulsive traits relative to other aspects of anxiety, the present study found obsessive compulsive traits were relatively less of a concern for parents who identify their children with PDA. EDA behaviours in children with PDA has been

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described as “driven by the child’s uncontrolled anxiety which can feel like a panic attack” (Syson & Gore Langton, 2017); however, parents who identified their children with PDA in the present study reported *panic* to be less of a concern when compared to other aspects of anxiety.

5.5.4 Sensory Sensitivities

Autistic children and adults are known to respond to sensory experiences differently to their peers (Tomchek & Dunn, 2007). Consistent with McCormick et al. (2016) finding more sensory symptoms within smell, taste, and auditory domains in children with autism, parents in the present study reported taste/smell sensitivity and auditory filtering as domains of concern relative to other aspects of sensory needs as presented in Table 12 in the *Results* section. It is of note however, that *movement sensitivity* was seen as relatively less of an area of concern for those with PDA. Movement sensitivity as a potential strength could be considered in combination with a desire for ‘role play’, which could support the development of intervention strategies known in autistic children, which could be relevant for children with PDA.

Analysing the subscales provided an opportunity to build a profile of PDA beyond that of deficit and consider potential strengths relative to common difficulties in autism, which could direct intervention and management planning for this population.

5.6 Theoretical implications

To date no research has considered the role of sensory sensitivities as an underlying factor contributing to anxiety in the PDA population. As has been suggested previously by Stuart et al. (2020), intolerance of uncertainty was a significant predictor of anxiety in this population group. However, where Stuart et al. hypothesised PDA as a dependent variable (outcome) of the relationship between IU and Anxiety, our findings showing a strong correlation between

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autism traits and EDA traits, suggests that these constructs may be difficult to separate, and therefore the EDA-Q may be better conceptualised as a screener for PDA rather than a description of a behavioural outcome of (extreme) demand avoidance. The strong positive relationship between autistic traits and EDA is present across the whole sample, meaning the relationships between these constructs are not reserved for only those who identify with pathological demand avoidance.

Whilst the ASSQ scores for those identifying with PDA were similar to those identifying with autism without a PDA profile, exploration of the subscales supports the development of our understanding of the profile of PDA in relation to what is known about autism. For example, one of the subscales of the ASSQ, Restrictive & Repetitive Behaviours (RRBs), is known to be an area of challenge for autistic individuals (e.g. Joyce et al., 2017); however, the subscale from the ASSQ that was of most difficulty for those identifying with PDA was Communication. Moreover, Motor Clumsiness was also identified as being comparatively less of an area of concern for those identifying with PDA, which could indicate not only difficulties but some areas of strengths for PDA and move understanding of the PDA profile away from a deficit-based understanding.

5.6.1 *The diagnosis and the behaviour*

Furthermore, there has been literature exploring behavioural outcomes such as Restrictive, Repetitive Behaviours (RRBs; e.g. Joyce et al., 2017) which are acknowledged as associated with autism but is also present in typical development. However, there has sometimes been a lack of clarity in the PDA literature as to whether what is being described is a neurodevelopmental diagnosis or a behavioural outcome. For example, PDA is described by the PDA Society as a neurodevelopmental diagnosis; however, it is also described as being ‘driven by anxiety’. The research by Stuart et al. (2020) focussed on those who identified with PDA as a diagnosis and conducted a mediator analysis with PDA as the outcome of IU

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(predictor) and anxiety (mediator). Whilst considering our data analysis strategy, we reflected on whether the behaviours described in the EDA-Q should represent a screening tool of PDA as a neurodevelopmental diagnosis similarly to the behaviours described in the ASSQ being used as a screening tool for autism, or whether the items on the EDA-Q measure a behavioural outcome such as those of the Repetitive Behaviour Questionnaire-2 (RBQ-2; Leekam et al., 2007). It was for these reasons we chose a data led approach to analysis, which provided the model of the EDA-Q being a screening tool for the (predictor) diagnostic construct of PDA.

5.7 Clinical Implications

We conceptualise within the model that autism and PDA are related, but at the same time should be considered separate and unique constructs, as their relationship to anxiety and the moderating constructs of intolerance of uncertainty and sensory sensitivities are distinct. This conceptualisation of PDA as related to, but also unique from autism, is consistent with the current conceptualisation of those displaying these traits as having “a PDA profile of autism” as described by both the National Autistic Society and the PDA Society. Also, this is consistent with the current practice guidance describing the need for an autism diagnosis to be concurrent with any diagnosis of pathological demand avoidance.

As noted, literature on PDA suggests an overlap of symptomology with ADHD; however, the development of an understanding of sensory needs in children with PDA could indicate a more nuanced ‘seeking’ of sensory stimulation, which may be currently being misinterpreted as hyperactivity and leading to misdiagnosing of ADHD in these children. Additionally, where parents reported ‘separation anxiety’ as an important factor for their child, clinical interventions such as graded exposure (as described above) may be less useful if considered alongside the autism diagnosis in which the presentation of ‘separation anxiety’ may be about

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having a safe person who understands their cues and needs rather than separation anxiety as presented in diagnostic manuals.

When considering clinical practice, there are known interventions in place to support those with autism in relation to uncertainty. These include interventions such as visual timetables and social stories that can help children with autism to be more aware of upcoming events and situations. However, these interventions may be less effective for those with PDA who are reported to have a significant 'need for control', which may mean they interpret tools such as a visual timetable as a demand on them to undertake the upcoming task on the timetable. It is suggested a more nuanced approach with flexible routines with choices would be required to support uncertainty in those with PDA.

There are also known interventions in place to support those with sensory needs; however, it has been reported that these interventions and approaches are often less effective with those who identify with PDA. The findings that sensory needs may be a significant contributing factor to anxiety in those identifying with PDA suggests a direction for intervention to support this population. Our findings suggest that more emotion regulation work, incorporating sensory processing strategies would be important, utilising a cross-disciplinary approach from psychological therapists and occupational therapy would be a more targeted intervention for those identifying with PDA. Approaches that integrate sensory processing tools as well as cognitive tools, which is the focus of the current literature on autism, could consider threat appraisal and the management of anxiety, and how sensory processing may moderate this relationship in those with PDA. As has been described, cognitive therapies are the most widely used approaches by NHS services supporting people in the UK; however, these findings suggest the need to consider alternatives such as sensory integration therapy which are less widely used as a more targeted intervention for this population to support better clinical outcomes.

5.8 Strengths of Current Study

5.8.1 Stakeholder involvement

A strength of this study was the involvement of relevant stakeholders in the development and implementation of the study. Support from those who work and live with PDA and autism helped ensure wherever possible that steps were taken that were mindful of the needs of this community. This included ease of use of the online survey and clarity over the language used to describe the constructs.

Despite the PDA community being recognised as an underrepresented group in research, there has been research conducted within PDA and autism that this study aimed to build upon. This meant some methodological approaches were replicated for continuity and development of knowledge within PDA research. For example, previous research considering the role of IU in PDA (Stuart et al., 2020) used parent-report measures of the EDA-Q and SCAS-P and therefore these measures were chosen for research continuity. Focus on the broader position of PDA research and how this project could develop knowledge around PDA in line with the research goals of the PDA Society supported the implementation of a ‘research stream’ from which future research projects have begun to taken place. The first of which was a direct request from the PDA community through feedback to the PDA Society social media pages to conduct research with those identifying with PDA as adults.

5.8.2 Theoretical considerations

The systemic literature review within this thesis highlighted a lack of clarity in the PDA literature as to whether what is being described is a neurodevelopmental diagnosis or a behavioural outcome. For example, PDA is described by Stuart et al. (2020) both as a predictor neurodevelopmental diagnosis and as a behavioural outcome that is ‘driven by anxiety’. This led the researchers to sample from those who identify with PDA and conduct a

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mediator analysis with PDA as the outcome of IU (predictor) and anxiety (mediator). Whilst considering our data analysis strategy we reflected on whether the behaviours described in the EDA-Q should represent a screening tool of PDA as a neurodevelopmental diagnosis similarly to the behaviours described in the ASSQ being used as a screening tool for autism, or whether the items on the EDA-Q measure a behavioural outcome such as those of the Repetitive Behaviour Questionnaire-2 (RBQ-2; (Leekam et al., 2007). Due to this complexity, we chose a data led approach to analysis, which provided the model of the EDA-Q being a screening tool for the (predictor) diagnostic construct of PDA.

5.8.3 Ethics and standardisation

Feedback from the PDA society's social media accounts was provided about the survey and suggested some of the wording on the ASSQ was not preferred by the PDA community. There is a greater awareness from clinical populations in the adopted terminology used to describe them in the research, which can in some cases lead to negative effects on the ways in which society views and treats them and also how they view themselves. For example, while more recent research has shown positive changes in the way autism is written and spoken about, "ableist" language is still used. Ableist language refers to language that assumes disabled people are inferior to non-disabled people and encourages a culture of separation; "it defines, excludes and marginalises people" (Bottema-Beutel et al., 2021). While the ASSQ is a reliable and validated measure, its language is reflective of historical misunderstandings, such as item 6 describing "*a deviant style of communication*".

After consultation with the PDA Society, we chose to keep the wording the same to maintain standardisation, but also included a note before the ASSQ to explain our reasoning behind this (see Appendix J). We also chose to include this as 'a note about the language' in the scale rather than a 'trigger warning' due to recent research on trigger warnings suggesting this could result in "people feel[ing] more anxious about the material" (Bridgland et al.,

2022). Future research should reflect on and update some of the current standardised measures in collaboration with those with whom the research is being conducted, adjusting their language choice on items which could cause concern.

5.9 Methodological Limitations

5.9.1 Proxy reporting

Despite our justification for the measures used within this study, the discrepancy between parent and child-report measures of anxiety in the autistic population has been described as a methodological limitation previously (Kaat & Lecavalier, 2015). Having parent-report measures meant we were unsure whether the children who were described as having PDA or any other diagnosis by their parents / caregivers associated with the diagnoses themselves. Therefore, the inferences made from the results of this study must be framed within the knowledge of this limitation. Furthermore, research has shown that when different informants (teachers & parents) rate a child's behaviours using the same measure, discrepancies often occur (De Reyes & Kazdin, 2005; Lopata et al., 2016) which has also been evident in PDA research (Brede et al., 2017). It is suggested that one reason for the lack of agreement may be due to different measures failing to characterise a specific trait across different environments such as home and school. Moreover, discrepancies in rating can also occur due to unique perceptions of the individual being rated (Hoyt, 2000), and there is often a lack of consensus in the ratings for parents and children across internalisation measures. Therefore, it is important that future research takes a more multi-informant approach across these measures.

5.9.2 Diagnostic grouping

The focus of our sampling and data analysis was to make sense of the underlying factors contributing towards anxiety in those who identify with the diagnosis of pathological demand avoidance (PDA). However, some of the participants did not identify their children with

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PDA, and instead with other diagnoses. Where Egan et al. (2019) found ADHD predicted PDA traits in adults, this relationship was not explored within the current study. Both Autism and ADHD are considered neurodevelopmental disorders (APA, 2013), and while they are recognised by their unique features, there is a lot of symptoms overlap and high comorbidity between them (Lecavalier et al., 2018). Therefore, it is suggested that both autism and ADHD should be considered in future research exploring PDA.

Our research intended to explore anxiety in those identifying with a profile of PDA; however, previous research has been critiqued for allocation of participants to a 'PDA group' without validation of the diagnosis e.g. Woods (2020). Notwithstanding the fact that PDA is not recognised in either of the main diagnostic and statistical manuals (the DSM-5 and ICD-11), diagnostic groups are often validated in research through use of a screening measure which was the purpose of the development of the EDA-Q (O'Nions et al., 2014). One option was to comprise a PDA group based on those who scored above the cut off scores for both the ASSQ and PDA group in line with the requirement as recognised by the National Autistic Society and the practice guidelines of the PDA Society that those being diagnosed with PDA also require an autism diagnosis. However, this left the dilemma of how to make sense of those who identified with PDA but who did not identify with autism. Reflecting that the screening tools were developed predominantly for research purposes rather than diagnostic tools in themselves, we focussed our aims on making sense of how the constructs (autistic traits, PDA traits, anxiety, intolerance of uncertainty and sensory sensitivities) related to each other, and how this may or may not be different in the population under investigation: those who *identify* with the diagnosis of PDA.

5.10 Suggestions for future research

Our study investigated the roles IU and SS have as underlying factors contributing to anxiety in autism and PDA; however, there are other known factors underlying anxiety in autistic

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people such as Theory of Mind (e.g. Korkmaz, 2011) or differences in Reasoning (e.g. Brosnan et al., 2016) which may be considered for future research to provide further explanatory power to the model. Furthermore, as making sense of co-morbidity can help differentiate symptoms of autism (Matson & Williams, 2013) it is suggested that relationships to other commonly co-occurring neurodevelopmental diagnoses such as ADHD (Egan et al., 2019) be considered when researching the PDA profile.

The direct relationship between PDA and Anxiety (Stuart et al., 2020) which is not present between autism and anxiety (Neil et al., 2016) suggests an area of further research to explore this unique direct relationship that could help distinguish PDA from autism.

With findings suggesting sensory needs in those with PDA, it is suggested that future research could investigate approaches that integrate sensory processing tools as well as cognitive tools to look at threat appraisal and the management of anxiety, and how sensory processing may moderate this relationship in those with PDA.

Future research could consider validation of the EDA-Q measure for use with this population through exploratory factor analysis. A correlational analysis of the measure used in this study showed items 14 and 20 to have weak correlation with the other items in the measure which is consistent with Stuart et al. (2020)'s findings. Further validation of the items within the EDA-Q is suggested.

Future research should also consider directly capturing the experiences of children identifying with PDA. Research should consider directly self-report rather than proxy report to potentially validate responses against parent-report. Furthermore, future research could consider exploring these constructs within the adult population as suggested in feedback from the PDA community through comments on the PDA Society's online posts publicising this study.

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This study aimed to investigate those who *identify* with the diagnosis of PDA; however, it is suggested that future research could consider differences in those who identify with PDA with self-diagnosis and those who have been given a diagnosis by a clinician or those who meet the screening criteria or PDA.

5.11 Conclusion

The findings of this study contribute to the existing body of literature describing pathological demand avoidance, its relationship to autism, and underlying factors contributing to anxiety in this population. Our findings suggest autistic traits and PDA traits to be distinct but highly related constructs that should be considered in relation to one another. To date, no study has specifically explored the role of sensory sensitivities as a contributing factor to anxiety in the PDA population, and these results suggest sensory sensitivities (SS) *and* intolerance of uncertainty (IU) to be significant factors when making sense of anxiety in those identifying with PDA, supporting previous research (O’Nions et al., 2014) suggesting that PDA presents a clinically relevant, distinct yet related profile of autism.

Research has identified that autistic individuals experience anxiety differently to typically developing individuals (Rodgers & Ofield, 2018); however, more research is needed to make sense of anxiety specifically in those presenting with ‘a PDA profile of autism’. Because individuals with PDA were reported to have overlapping (i.e. separation anxiety) but unique (i.e. less obsessive compulsive traits) experiences of anxiety when compared to what is understood about anxiety in autism suggests a unique experience of the world for individuals with PDA, and the reported relationship between taste, smell and auditory needs and anxiety in this study identifies that more research is needed to be done to demonstrate and understand the way in which individuals with PDA experience and present with anxiety.

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Demand avoidance appears to be a necessary strategy for this population to regulate their emotions and feel in control of their experiences of the world around them. Processing of taste, smell and auditory information, sensing a lack of control, and experiencing feelings of uncertainty are identified within the autism literature as factors contributing to anxiety, which when unmanaged, can result in the presentation of challenging behaviour. This also appears relevant to those with PDA. Demand avoidance itself can therefore be seen as a necessary coping strategy, and challenging behaviour as the result of when coping strategies prove ineffective. Demand avoidance has been described as a coping strategy anecdotally from people with PDA and their parents (e.g. Notes On PDA, 2017) and the challenging behaviours that differentiate PDA from autism are described within the PDA literature as “*Apparently manipulative behaviour, difficulties with other people, harassment of others, fantasising, lying, cheating, stealing and socially shocking behaviour*” (O’Nions et al., 2014).

This presentation of demand avoidance as a necessary coping strategy and other challenging behaviours resulting from coping strategies being ineffective are well documented within research identifying pathological demand avoidance (Newson et al, 2003; O’Nions et al, 2014), with PDA having been coined “a profile of obsessive resistance to everyday demands and requests, with a tendency to resort to ‘socially manipulative’ behaviour” (O’Nions et al, 2014, p.538).

This study has identified sensory needs and feelings of uncertainty as key triggers inducing anxiety in the PDA population, and demand avoidance as a strategy to manage these needs. This study strengthens previous beliefs about the importance of the relationship between anxiety and sensory needs cross-diagnostically (McCormick et al., 2016), where having significant sensory needs can lead to feelings of uncertainty and increased anxiety (Wigham et al., 2015), and the avoidance of demands/situations to regulate their emotions and feel in control (with more of a sense of certainty) can mean the individual is less exposed to sensory

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situations which can exacerbate sensory sensitivities (Neil et al., 2016). The importance of acknowledging sensory needs is further demonstrated within this study and highlights the importance of sensory focussed interventions such as graded expose techniques (Rodgers & Ofield, 2018) for individuals with PDA to reduce anxiety.

Understanding sensory needs alongside intolerance of uncertainty as key underlying mechanisms behind anxiety in those identifying with PDA can support tailored, comprehensive assessment schedules and more individualised behaviour management strategies for this population.

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APPENDICES

Appendix A: Prospero Review Protocol Registration

NIHR | National Institute
for Health Research

PROSPERO
International prospective register of systematic reviews

Investigating the factors underlying anxiety in individuals with pathological demand avoidance across the lifespan: a systematic review

Citation

[REDACTED] et al. Investigating the factors underlying anxiety in individuals with pathological demand avoidance across the lifespan: a systematic review. PROSPERO 2023 CRD42023390500 Available from: https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42023390500

Review question

- What is the relationship between anxiety and demand avoidance?
- Can Intolerance of Uncertainty and/or sensory disturbances relate to anxiety in Pathological Demand Avoidance (PDA)?
- How does the relationship compare to other neurodiverse conditions, and those who have co-occurring symptoms?
- Is the relationship moderated by age, gender, IQ or even informant type (self-report vs proxy-report)?
- What is the relationship between anxiety and demand avoidance?
- Can Intolerance of Uncertainty and/or sensory disturbances relate to anxiety in Pathological Demand Avoidance?
- How does the relationship compare to other neurodiverse conditions, and those who have co-occurring symptoms?
- Is the relationship moderated by age, gender, IQ or even informant type (self-report vs proxy-report)?

Searches

Sources that will be searched are: Scopus, PsycArticles, PubMed, and MEDLINE.

Search terms will be related to:

- The identified phenomenon: pathological demand avoidance; AND
- The underlying factors: anxiety OR intolerance of uncertainty OR sensory sensitivities

An example of a search strategy to capture different terms related to the same construct e.g 'pathological demand avoidance', 'extreme demand avoidance' and 'demand avoidant behaviours' is: "demand avoidan*"

Studies involving participants with PDA, children, and adults (both with and without co-occurring disorders) or caregivers of a child/children with PDA will be included in the review. There are no restrictions to the location of the study or type of methodology that has attempted to address this relationship albeit quantitative or qualitative.

Types of study to be included

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There are no restrictions to the location of the study or type of methodology that has attempted to address this relationship albeit quantitative or qualitative. Studies not in English, or in books, book reviews, commentaries or position papers will be excluded.

Condition or domain being studied

Pathological Demand Avoidance (PDA), first proposed as a pervasive developmental disorder by Newson, Le Marechal, & David (2003), was characterised as “an obsessional avoidance of the demands of everyday life”. PDA was proposed as requiring its own diagnostic category due to the presence of “social manipulation” as seen in all children meeting the criteria for PDA and having a greater degree of difficulty with emotion regulation when compared to those with Autism (O’Nions et al., 2013). Despite its acceptance by the National Autistic Society as part of the autism spectrum, PDA is still highly controversial, with diagnostic manuals including the DSM-5 not currently recognising PDA as either a separate diagnosis or a subgroup within the autism spectrum. There remains an ongoing debate as to whether a requirement for a separate diagnostic category is indicated.

Participants/population

Any paper describing empirical data involving participants with pathological/extreme demand avoidance, children and adults (both with and without co-occurring disorders) or caregivers of a child/children with PDA published in a peer-reviewed journal will be included in the review.

Intervention(s), exposure(s)

Not applicable (this review is investigating the factors underlying anxiety in individuals with pathological demand avoidance across the lifespan).

Comparator(s)/control

Not applicable.

Main outcome(s)

- This review will help to highlight both commonalities and differences in PDA compared to other neurodevelopmental populations where sensory and anxiety related concerns have often been noted.
- To identify any reported overlapping or disorder-specific effects of PDA related to anxiety, sensory sensitivities, and intolerance of uncertainty.

Measures of effect

Not applicable.

Additional outcome(s)

None.

Measures of effect

Not applicable.

Data extraction (selection and coding) [1 change]

Quantitative and Qualitative studies will be included as will intervention-based studies. The PRISMA guidelines will be followed for this review (Page et al., 2021).

Page: 2 / 5

Two authors will independently review the database search results and exclude articles based on title and abstract screening using the inclusion and exclusion criteria. Where there is any ambiguity, the full text will be included for full screening. Both authors will together assess which of the full text articles will be included for the review and where any disagreement the third author will act as an independent moderator for whether the article is included.

The data to be extracted included: details of the environment, the study design, descriptive statistics of the groups in the study (age, gender, diagnosis etc.), the primary and secondary outcomes of the included studies, and the results. The results will be synthesized in a narrative manner in the results and interpreted in the discussion sections.

Risk of bias (quality) assessment

Various critical appraisal tools from the Joanna Briggs Institute will be used depending on the studies.

PECO:

Population: Individuals of all ages, without regard to gender, race or ethnicity.

Environment: Type of environment (e.g home/school)

Comparator: Where placebo or controls groups exist (those without demand avoidance behaviours), they will be reported.

Outcome measure: Changes in demand avoidance behaviours based on anxiety as assessed via, assessment tools or qualitative study. Secondary outcomes changes in sensory and/or intolerance to uncertainty measures based on demand avoidance behaviours

The principal aim of the review is to derive a formal model of the construct of 'demand avoidance' in the context of research involving autistic and non-autistic individuals in order to facilitate a more systematic approach to making sense of the underlying causes of anxiety in this population group in naturalistic settings. The data from included studies will be synthesised as a narrative review and data will be analysed thematically.

The main criterion for data synthesis is therefore that included studies should have the explicit aim of examining participants' anxiety, and any other construct that may underlie this in people identifying with pathological demand avoidance. Data will be organised by invitation method, type of screening programme and strength of evidence.

Furthermore, we aim to develop a classification of these underlying factors, based on the specific their prevalence in relationship to any co-occurring condition such as Autism or ADHD. This will enable us to develop a taxonomy of the different underlying causes / factors of demand avoidance and to associate each factor with valid paradigms to measure it. Reasons for contradictory findings will be also be explored.

The critical data to be extracted from each study will include a careful description of how the construct of 'demand avoidance' was described and how the effects of relevant underlying factors were measured. These descriptions will then be classified in order to identify which facets of the environment or an event are the focus of explanatory factors underlying the behavioural presentation of demand avoidance, and which are expressed as internal to the individual. This data will be presented narratively, with tables and graphical displays (where appropriate).

Analysis of subgroups or subsets

None planned.

Contact details for further information

[Redacted contact details]

Organisational affiliation of the review

University of Hertfordshire
www.herts.ac.uk

Review team members and their organisational affiliations [1 change]

[Redacted] University of Hertfordshire
[Redacted] University of Hertfordshire
[Redacted] University of Hertfordshire

Type and method of review

Diagnostic, Narrative synthesis, Systematic review

Anticipated or actual start date

15 January 2023

Anticipated completion date

01 June 2023

Funding sources/sponsors

This review has not received any financial support

Conflicts of interest

Language

English

Country

England

Stage of review

Review Ongoing

Subject index terms status

Subject indexing assigned by CRD

Subject index terms

Anxiety; Anxiety Disorders; Humans; Longevity; Self Report; Uncertainty

Date of registration in PROSPERO

09 February 2023

Date of first submission

09 January 2023

Stage of review at time of this submission

The review has not started

Stage	Started	Completed
Preliminary searches	No	No
Piloting of the study selection process	No	No
Formal screening of search results against eligibility criteria	No	No
Data extraction	No	No
Risk of bias (quality) assessment	No	No
Data analysis	No	No

The record owner confirms that the information they have supplied for this submission is accurate and complete and they understand that deliberate provision of inaccurate information or omission of data may be construed as scientific misconduct.

The record owner confirms that they will update the status of the review when it is completed and will add publication details in due course.

Versions

09 February 2023

Appendix B: Quality Assessment with Diverse Studies tool

(QuADS; Harrison et al., 2021)

Supplemental Table: Quality assessment of included studies using the quality assessment tool for studies with diverse designs

QuADS item criteria: (1) Theoretical framework (2) Aims/objectives; (3) Description of research setting (4) Appropriate study design; (5) Appropriate sampling to address aims (6) Rationale for data collection tools; (7) Format and content of data collection tool(s); (8) Description of data collection procedure; (9) Recruitment data provided; (10) Justification for analytic method selected; (11) The method of analysis was appropriate to answer the research aim/s; (12) Evidence that the research stakeholders have been considered in research design or conduct; (13) Strengths and limitations critically discussed.

QuADS rating scale: 0=not at all; 1=very slightly; 2=moderately; 3=complete (rate for each of the 13 items in table below)

First coder

QuADS Criteria	Item 1	Item 2	Item 3	Item 4	Item 5	Item 6	Item 7	Item 8	Item 9	Item 10	Item 11	Item 12	Item 13	Total score / 39	% of total score
Schneider et al., 2022	2	3	3	3	3	2	3	3	3	3	3	0	3	34	87
O'Nions et al., 2018	2	0	3	3	3	3	3	3	3	3	3	0	3	32	82
Brede et al., 2017	1	2	3	2	1	1	2	2	2	2	2	0	2	22	56
O'Nions et al., 2021	3	3	3	3	3	3	3	3	3	3	3	0	3	36	92
O'Nions et al., 2016	3	3	2	3	3	3	2	2	3	3	3	0	3	33	85

Anxiety and EDA

Egan et al., 2020	2	3	2	2	3	3	3	3	3	2	2	0	2	30	77
Stuart et al., 2020	3	3	3	3	3	3	3	3	3	3	2	0	3	35	90
O’Nions et al., 2014	3	3	3	3	3	3	3	1	2	2	3	0	3	32	82
Reilly et al., 2014	2	1	3	3	3	2	3	2	2	3	2	0	3	29	74
Doyle & Kenny, 2023	2	1	2	2	2	1	2	2	1	2	2	0	1	20	51
Truman et al., 2021	3	2	3	2	2	2	3	3	2	3	3	0	3	31	79
Trundle et al., 2022	2	3	2	2	2	2	2	2	3	2	2	0	3	27	69
White et al., 2022	3	3	2	3	2	3	3	3	3	2	3	0	3	33	85

Second coder

QuADS Criteria	Item 1	Item 2	Item3	Item 4	Item 5	Item 6	Item 7	Item 8	Item 9	Item 10	Item 11	Item 12	Item 13	Total score / 39	% of total score
Schneider et al., 2022	2	2	3	3	3	3	3	3	3	3	3	0	3	34	87

Anxiety and EDA

O’Nions et al., 2018	2	2	3	3	3	3	3	3	3	3	3	0	3	34	87
Brede et al., 2017	1	2	2	2	2	2	2	2	2	2	2	0	2	23	59
O’Nions et al., 2021	3	3	3	3	3	3	3	3	3	3	3	0	3	36	92
O’Nions et al., 2016	3	3	2	3	2	3	2	2	3	3	3	0	3	32	82
Egan et al., 2020	2	3	2	3	2	3	3	3	3	2	3	0	2	31	79
Stuart et al., 2020	3	3	3	3	3	3	3	3	3	3	3	0	3	36	92
O’Nions et al., 2014	3	3	2	3	2	3	3	3	1	2	3	0	3	31	79
Reilly et al., 2014	2	2	3	2	3	2	3	3	2	3	2	0	3	30	77
Doyle & Kenny, 2023	1	2	2	2	2	1	2	2	2	2	2	0	2	22	56
Truman et al., 2021	2	2	2	2	2	2	3	3	2	3	2	0	3	28	72
Trundle et al., 2022	2	2	2	2	2	2	2	2	2	2	2	0	2	24	62
White et al., 2022	3	3	2	2	2	3	3	3	3	2	2	0	3	31	79

Appendix C: Ethical Approval Notification



HEALTH, SCIENCE, ENGINEERING AND TECHNOLOGY ECDA

ETHICS APPROVAL NOTIFICATION

TO [REDACTED]
CC [REDACTED]
FROM Dr Simon Trainis, Health, Science, Engineering & Technology ECDA Chair
DATE 05/01/2023

Protocol number: **aLMS/PGR/UH/05062(2)**

Title of study: Does intolerance of uncertainty and sensory sensitivity underlie demand avoidant behaviours in autistic and non-autistic children?

Your application to modify and extend the existing protocol as detailed below has been accepted and approved by the ECDA for your School and includes work undertaken for this study by the named additional workers below:

[REDACTED] (Clinical Psychologist) – Field Supervisor

Modification: Detailed in EC2

General conditions of approval:

Ethics approval has been granted subject to the standard conditions below:

Original protocol: Any conditions relating to the original protocol approval remain and must be complied with.

Permissions: Any necessary permissions for the use of premises/location and accessing participants for your study must be obtained in writing prior to any data collection commencing. Failure to obtain adequate permissions may be considered a breach of this protocol.

External communications: Ensure you quote the UH protocol number and the name of the approving Committee on all paperwork, including recruitment advertisements/online requests, for this study.

Invasive procedures: If your research involves invasive procedures you are required to complete and submit an EC7 Protocol Monitoring Form, and copies of your completed consent paperwork to this ECDA once your study is complete.

Submission: Students must include this Approval Notification with their submission.

Validity:

This approval is valid:

From: 05/01/2023

To: 01/05/2023

Please note:

Failure to comply with the conditions of approval will be considered a breach of protocol and may result in disciplinary action which could include academic penalties.

Additional documentation requested as a condition of this approval protocol may be submitted via your supervisor to the Ethics Clerks as it becomes available. All documentation relating to this study, including the information/documents noted in the conditions above, must be available for your supervisor at the time of submitting your work so that they are able to confirm that you have complied with this protocol.

Should you amend any aspect of your research or wish to apply for an extension to your study you will need your supervisor's approval (if you are a student) and must complete and submit a further EC2 request.

Approval applies specifically to the research study/methodology and timings as detailed in your Form EC1A or as detailed in the EC2 request. In cases where the amendments to the original study are deemed to be substantial, a new Form EC1A may need to be completed prior to the study being undertaken.

Failure to report adverse circumstance/s may be considered misconduct.

Should adverse circumstances arise during this study such as physical reaction/harm, mental/emotional harm, intrusion of privacy or breach of confidentiality this must be reported to the approving Committee immediately.

Appendix D: Participant Information Sheet

UNIVERSITY OF HERTFORDSHIRE ETHICS COMMITTEE FOR STUDIES INVOLVING THE USE OF HUMAN PARTICIPANTS (Survey) ('ETHICS COMMITTEE') FORM EC6: PARTICIPANT INFORMATION SHEET

1 Title of study:

Does intolerance of uncertainty and sensory sensitivity underlie demand avoidant behaviours in autistic and non-autistic children?

2 Introduction:

You are being invited to take part in a study. Before you decide whether to do so, it is important that you understand the study that is being undertaken and what your involvement will include. Please take the time to read the following information carefully and discuss it with others if you wish. Do not hesitate to ask us anything that is not clear or for any further information you would like to help you make your decision. Please do take your time to decide whether or not you wish to take part. The University's regulation, UPR RE01, 'Studies Involving the Use of Human Participants' can be accessed via this link: <https://www.herts.ac.uk/about-us/governance/university-policies-and-regulations-uprs/uprs> (after accessing this website, scroll down to Letter S where you will find the regulation) Thank you for reading this.

3 What is the purpose of this study?

As part of fulfilling the requirements for the principal researcher's doctoral course in clinical psychology at the University of Hertfordshire, we wish to examine the relationship between intolerance of uncertainty, sensory sensitivities and anxiety in groups of autistic and non-autistic children with and without demand avoidant behaviours.

4 Do I have to take part?

It is completely up to you whether or not you decide to take part in this study. If you do decide to take part, you will be given this information sheet to keep and be asked to sign a consent form. Agreeing to join the study does not mean that you have to complete it. You are free to withdraw at any stage without giving a reason. A decision to withdraw at any time, or a decision not to take part at all, will not affect any treatment/care that you may receive (should this be relevant).

5 Are there any age or other restrictions that may prevent me from participating?

There are no age restrictions to taking part in this study for yourself; however, this does apply to your child as we are seeking parents and carers of children aged 4–17 years.

6 How long will my part in the study take?

If you decide to take part in this study, you will be involved in it for around 20 minutes to complete the online questionnaires. At the end of the questionnaires there is an option to provide your name and email address if you wish to be contacted about potentially being involved in a follow-up online interview. If you choose to take part, this will be conducted at a later date, online via Microsoft Teams and should last around 45-60 minutes. I will be asking you about questions related to this survey, and more broadly to your lived experiences in relation to demand avoidant behaviours and parenting.

7 What will happen to me if I take part?

The first thing to happen is being asked to read this information sheet and complete the following consent form. You will then be directed to the survey questions. These are multiple choice questions asking about various things you have noticed about your child. There are no right or wrong responses to the questions. You are asked to respond to all the questions before closing this browser.

8 What are the possible disadvantages, risks or side effects of taking part?

We believe there are no known risks associated with this research study; however, as with any online activity the risk of a breach is always possible. We will do everything possible to ensure your responses in this study will remain anonymous. We do not anticipate that you will find any of the questions upsetting. However, if you feel that you need support following completion, we recommend you contact your GP, or the Samaritans can be contacted on 116123, or a family member. I have additionally provided signposts to potential support networks below:

MIND: <https://www.mind.org.uk/>

ACAS: <https://www.acas.org.uk/>

The National Autistic Society's online community for parents and carers: <https://community.autism.org.uk/f/parents-and-carers>

The PDA Society's Enquiry Line: <https://www.pdasociety.org.uk/contact-us/enquiry-line/>

Ambitious About Autism's support for families: <https://www.ambitiousaboutautism.org.uk/information-about->

Anxiety and EDA

autism/early-years/support-for-families

Contact's Listening Ear service <https://contact.org.uk/help-for-families/listening-ear/>

9 What are the possible benefits of taking part?

Taking part in this study will help us gain a greater understanding of some predictors of anxiety in children and what predictors that may be more important for neuro-diverse children. It is hoped that by sharing your experiences, we can create a better understanding of how we could improve support and better meet the needs of children with additional needs.

10 How will my taking part in this study be kept confidential?

We will follow ethical and legal practice and all information will be handled in confidence. Under UK Data Protection laws the University is the Data Controller (legally responsible for the data security) and the Chief Investigator of this study (named above) is the Data Custodian (manages access to the data). This means we are responsible for looking after your information and using it properly. Your rights to access, change or move your information are limited as we need to manage your information in specific ways to comply with certain laws and for the research to be reliable and accurate.

To safeguard your rights, we will use the minimum personally identifiable information possible. Any information that you provide will be anonymised for confidentiality reasons and stored on the secure University of Hertfordshire One-Drive. We would like your permission to use anonymised data in future studies, and to share our research data (e.g. in online databases) with other researchers in other Universities and organisations both inside and outside the European Union. This would be used for research in health and social care. Sharing research data is important to allow peer scrutiny, re-use (and therefore avoiding duplication of research) and to understand the bigger picture in particular areas of research. All personal information that could identify you will be removed or changed before information is shared with other researchers or results are made public. If you have any questions or concerns, please don't hesitate to ask. We can be contacted before and after your participation at the email addresses provided.

11 Audio-visual material

If you would like to take part in a follow-up interview at a later date, we will meet online via Microsoft Teams. This interview will be audio-visual recorded.

12 What will happen to the data collected within this study?

The results of this study will be used to promote future research, understanding anxiety and sensory sensitivities in autistic and non-autistic children with demand avoidance. The results of the study and/or the data collected (in anonymised form) may be deposited in an open access repository and written up for publication. Any information that could be used to identify individuals will be removed from published material. The results will be used for the write up of my major project that forms part of the Professional Doctorate in Clinical Psychology qualification.

13 Will the data be required for use in further studies?

The data will not be used in any further studies.

14 Who has reviewed this study?

This Study has been reviewed by The University of Hertfordshire Health, Science, Engineering & Technology Ethics Committee with Delegated Authority The UH protocol number is aLMS/PGR/UH/05062(2)

15 Factors that might put others at risk.

Please note that if, during the study, any medical conditions or non-medical circumstances such as unlawful activity become apparent that might or had put others at risk, the University may refer the matter to the appropriate authorities and, under such circumstances, you will be withdrawn from the study.

16 Who can I contact if I have any questions?

If you would like further information or would like to discuss any details personally, please get in touch with me, Aaron J. Rai (Trainee Clinical Psychologist) by email at ar20acm@herts.ac.uk or you can contact my research supervisors Dr Amanda Ludlow at a.ludlow@herts.ac.uk and/or Dr Barbara Rishworth at b.rishworth@herts.ac.uk

Although we hope it is not the case, if you have any complaints or concerns about any aspect of the way you have been approached or treated during the course of this study, please write to the University's Secretary and Registrar at the following address: Secretary and Registrar University of Hertfordshire, College Lane, Hatfield, Herts, AL10 9AB.

Thank you very much for reading this information and giving consideration to taking part in this study.

Appendix E: Consent Form

UNIVERSITY OF HERTFORDSHIRE ETHICS COMMITTEE FOR STUDIES INVOLVING THE USE OF HUMAN PARTICIPANTS (ANONYMOUS SURVEY) ('ETHICS COMMITTEE') FORM EC3
CONSENT FORM FOR STUDIES INVOLVING HUMAN PARTICIPANTS

I hereby freely agree to take part in the study entitled: "Does intolerance of uncertainty and sensory sensitivity underlie demand avoidant behaviours in autistic and non-autistic children?" (UH Protocol number aLMS/PGR/UH/05062(2))

1 I confirm that I have been given a Participant Information Sheet (a copy of which is attached to this form) giving particulars of the study, including its aim(s), methods and design, the names and contact details of key people and, as appropriate, the risks and potential benefits, how the information collected will be stored and for how long, and any plans for follow-up studies that might involve further approaches to participants. I have also been informed of how my personal information on this form will be stored and for how long. I have been given details of my involvement in the study. I have been told that in the event of any significant change to the aim(s) or design of the study I will be informed, and asked to renew my consent to participate in it.

2 I have been assured that I may withdraw from the study at any time without disadvantage or having to give a reason.

3 I have been told how information relating to me (data obtained in the course of the study, and data provided by me about myself) will be handled: how it will be kept secure, who will have access to it, and how it will or may be used, including the possibility of anonymised data being deposited in a repository with open access (freely available).

4 I understand that if there is any revelation of unlawful activity or any indication of non-medical circumstances that would or has put others at risk, the University may refer the matter to the appropriate authorities.

5 I have been told that I may at some time in the future be contacted again in connection with this or another study.

Name of (principal) investigator: 

Appendix F: Online Poster for Recruitment

University of Hertfordshire UH

Parents Needed for Research Study on Demand Avoidance

Calling all parents! For my **online research project** entitled '*Does intolerance of uncertainty and sensory sensitivity underlie demand avoidant behaviours in autistic and non-autistic children?*' to better understand demand avoidance in children.

Participation Involves:

- Completing an online survey; providing responses to questionnaires on autistic traits, demand avoidance, sensory behaviors, 'intolerance to uncertainty' and anxiety. It should take around **20 mins to complete**.

Please find the link here:
https://herts.eu.qualtrics.com/jfe/form/SV_a8LtR5cjcfnSVtY

<p>You can participate if:</p> <ul style="list-style-type: none">• You have a child between 4 and 17 years old.	<p>Potential Benefits:</p> <ul style="list-style-type: none">• You can choose to be entered into a raffle at the end with a £50 prize.
--	---

FOR MORE INFORMATION
Please contact [redacted], supervised by [redacted]

My name is [redacted]. Thank you very much for reading this and considering to taking part in this study. The results of this research project will form the basis of the major clinical research project as part of my qualification for the Doctorate in Clinical Psychology at the University of Hertfordshire."

Dr [redacted] is [redacted]

UH ethics protocol number: ~~2~~MS/PGR/UH/05062(2)
UH approving committee: Health, Science, Engineering & Technology ECDA

Appendix G: Raffle prize draw details

Raffle

Excel procedure from original full list of 1569 participants:

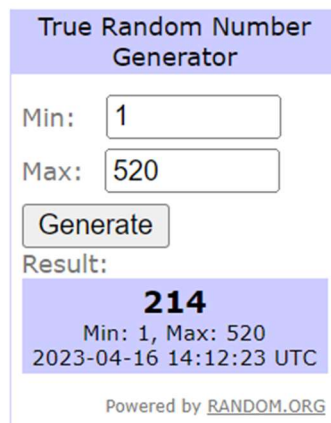
I. Removing Blank Rows with Find & Select

1. Click Find & Select.
2. Click to Go to Special.
3. Choose Blanks.
4. Click OK and then all the blank rows/cells will be highlighted.
5. Choose the Delete under Cells section on the Home Tab.
6. Click Delete Sheet Rows.

The above removed empty cells (missing info so not entered into raffle).

Remainder involved in raffle: n=520.

Random number generator website: <https://www.random.org/>



The image shows a screenshot of the 'True Random Number Generator' website. The interface includes a title bar, input fields for 'Min' (set to 1) and 'Max' (set to 520), a 'Generate' button, and a 'Result' section displaying the number 214. Below the result, it shows the range 'Min: 1, Max: 520' and the timestamp '2023-04-16 14:12:23 UTC'. At the bottom, it states 'Powered by RANDOM.ORG'.

Translates to participant number 304 in SPSS (305 in Excel) in final database

Appendix H: Information sent to High Grange School

Dear High Grange parents,

My name is [REDACTED]. I worked at High Grange School from 2014 to 2016, focusing on how we could help monitor and understand students' behaviours, and then worked in the therapy team as an Assistant Psychologist. Working at High Grange was the first time I heard from students, parents and teachers who told me about PDA (pathological demand avoidance). My work experience has motivated me to try and improve our knowledge of how demand avoidant behaviours may be better understood.

I am now training to be a Clinical Psychologist. I would like to invite you to participate in my research project which forms part of my qualification for the Doctorate in Clinical Psychology at the University of Hertfordshire. Please find a link to the survey

here: https://herts.eu.qualtrics.com/jfe/form/SV_a8LtR5cjcfNSVtY

This survey is entitled '*Does intolerance of uncertainty and sensory sensitivity underlie demand avoidant behaviours in children: A comparison of children with and without a diagnosis of Autism*'. My research aims to address whether intolerance of uncertainty and sensory sensitivity underlie demand avoidant behaviours in children with and without an autism diagnosis. It is hoped that the findings of the project could help could shape schools' strategies working with PDA and autism.

I have provided Gavin Spicer with the ethics approval notification, approved by the Health, Science, Engineering & Technology ECDA (ethics protocol number: **aLMS/PGR/UH/05062(1)**). Should you wish to see this please feel free to ask Gavin.

Thank you very much for reading this information and giving consideration to taking part in this study.

Best wishes,

[REDACTED] supervised by [REDACTED]
[REDACTED]

Trainee Clinical Psychologist

School of Life & Medical Sciences

University of Hertfordshire

Hatfield AL10 9AB

UK

Appendix I: Information sent to Third Sector Organisations

To whom it may concern,

My name is [REDACTED], and I am training to be a Clinical Psychologist. I am contacting about my research project which forms part of my qualification for the Doctorate in Clinical Psychology at the University of Hertfordshire; please find a link to the survey

here: https://herts.eu.qualtrics.com/jfe/form/SV_a8LtR5cjcfNSVtY

This survey is entitled '*Does intolerance of uncertainty and sensory sensitivity underlie demand avoidant behaviours in children: A comparison of children with and without a diagnosis of Autism*', and we hope it will help improve our knowledge of how demand avoidant behaviours may be better understood, which could help could shape schools' strategies working with this population and help make sense of the terminology around demand avoidance.

We would be grateful if you could help us with recruitment for this research project. We welcome all interested parents to participate in the survey, which should take around 20 minutes to complete.

I have attached the ethics approval notification, approved by the Health, Science, Engineering & Technology ECDA, with ethics protocol number: **aLMS/PGR/UH/05062(1)**.

I am happy to discuss the project further with you. Please feel free to contact me if you have any further questions.

Best,

[REDACTED]

Trainee Clinical Psychologist

School of Life & Medical Sciences

University of Hertfordshire

Hatfield AL10 9AB

UK

Appendix J: A note about terminology on the ASSQ

Please note that the following set of questions are extracted from a standardised questionnaire. We recognise that some of the words/terminology are not necessarily what we would choose or deem most suitable.

Appendix K: Debrief at end of online survey

DEBRIEF SHEET: Does intolerance of uncertainty and sensory sensitivity underlie demand avoidant behaviours in autistic and non-autistic children?

Thank you for participating in this research. The current study was interested in us gaining a greater understanding of some predictors of anxiety in your child, and some potential differences for neuro-diverse children. This study hopes to improve our understanding of the relationship between intolerance of uncertainty, sensory sensitivities and anxiety in groups of autistic and non-autistic children. Please note that all information you have provided will remain confidential and has been anonymised by the primary researcher through the creation of your anonymity code.

Your input will help the research team in their efforts to improve knowledge of how demand avoidant behaviours may be better understood, which could help shape schools' strategies working with this population and help make sense of the terminology around demand avoidance.

If taking part in this research has raised any concerns for you, which you would like to discuss further, please contact [redacted] (primary researcher) at the email address below, or his supervisors [redacted] and/or [redacted]. If taking part in this research has raised any concerns for you, you may wish to contact the National Autistic Society for a range of useful support they offer to children and parents (0808 800 4160) and/or Child Autism UK (01344 882248) and/or the Pathological Demand Avoidance Society website <https://www.pdasociety.org.uk/>

Thank you once again for your contribution in the research. Should you have any questions or queries about the study, please contact [redacted] – primary researcher – [redacted]

Appendix L: Autism Spectrum Screening Questionnaire (ASSQ)

This child stands out from other children of his/her age in the following way;		No	Somewhat	Yes
1	is old-fashioned or precocious			
2	is regarded as an 'eccentric professor' by the other children			
3	lives somewhat in a world of his/her own with restricted idiosyncratic intellectual interests			
4	accumulates facts on certain subjects (good rote memory) but does not really understand the meaning			
5	has a literal understanding of ambiguous and metaphoric language			
6	has a deviant style of communication with a formal, fussy, 'old-fashioned' or 'robot-like' language			
7	invents idiosyncratic words and expressions			
8	has a different voice or speech			
9	expresses sounds involuntarily; clears throat, grunts, smacks, cries or screams			
10	is surprisingly good at some things and surprisingly poor at others			
11	uses language freely but fails to make adjustments to fit social contexts or the needs of different listeners			
12	lacks empathy			
13	makes naïve and embarrassing remarks			
14	has a deviant style of gaze			
15	wishes to be sociable but fails to make relationships with peers			
16	can be with other children but only on his/her terms			
17	lacks best friend			
18	lacks common sense			
19	is poor at games; no idea of cooperating in a team, scores 'own goals'			
20	has clumsy, ill coordinated, ungainly, awkward movements or gestures			
21	has involuntary face or body movements			
22	has difficulties in completing simple daily activities because of compulsory repetition of certain actions or thoughts			
23	has special routines; insists on no change			
24	shows idiosyncratic attachment to objects			
25	is bullied by other children			
26	has markedly unusual facial expression			
27	has markedly unusual posture			

Specify reasons other than above:

Appendix M: Extreme Demand Avoidance Questionnaire (EDA-Q)

		Not true	Somewhat true	Mostly true	Very true
1	Obsessively resists and avoids ordinary demands and requests.				
2	Complains about illness or physical incapacity when avoiding a request or demand.				
3	Is driven by the need to be in charge.				
4	Finds everyday pressures (e.g. having to go on a school trip/ visit dentist) intolerably stressful.				
5	Tells other children how they should behave, but does not feel these rules apply to him/herself.				
6	Mimics adult mannerisms and styles (e.g. uses phrases adopted from teacher/parent to tell other children off).				
7	Has difficulty complying with demands unless they are carefully presented.				
8	Takes on roles or characters (from TV/real life) and 'acts them out'.				
9	Shows little shame or embarrassment (e.g. might throw a tantrum in public and not be embarrassed).				
10	Invents fantasy worlds or games and acts them out.				
11	Good at getting round others and making them do as s/he wants.				
12	Seems unaware of the differences between him/herself and authority figures (e.g. parents, teachers, police).				
13	If pressurised to do something, s/he may have a 'meltdown' (e.g. scream, tantrum, hit or kick).				
14	Likes to be told s/he has done a good job.				
15	Mood changes very rapidly (e.g. switches from affectionate to angry in an instant).				
16	Knows what to do or say to upset specific people.				
17	Blames or targets a particular person.				
18	Denies behaviour s/he has committed, even when caught red handed.				
19	Seems as if s/he is distracted 'from within'.				
20	Makes an effort to maintain his/her reputation with peers.				
21	Uses outrageous or shocking behaviour to get out of doing something.				
22	Has bouts of extreme emotional responses to small events (e.g. crying/giggling, becoming furious).				
23	Social interaction has to be on his or her own terms.				
24	Prefers to interact with others in an adopted role, or communicate through props/toys.				
25	Attempts to negotiate better terms with adults.				
26	S/he was passive and difficult to engage as an infant.				

Appendix N: Spence Children’s Anxiety Scale-Parent (SCAS-P)

BELOW IS A LIST OF ITEMS THAT DESCRIBE CHILDREN. FOR EACH ITEM PLEASE CIRCLE THE RESPONSE THAT BEST DESCRIBES YOUR CHILD. PLEASE ANSWER ALL THE ITEMS.

Never	Sometimes	Often	Always
-------	-----------	-------	--------

<ol style="list-style-type: none"> 1. My child worries about things..... 2. My child is scared of the dark..... 3. When my child has a problem, s(he) complains of having a funny feeling in his / her stomach 4. My child complains of feeling afraid..... 5. My child would feel afraid of being on his/her own at home..... 6. My child is scared when s(he) has to take a test..... 7. My child is afraid when (s)he has to use public toilets or bathrooms..... 8. My child worries about being away from us / me..... 9. My child feels afraid that (s)he will make a fool of him/herself in front of people..... 10. My child worries that (s)he will do badly at school..... 11. My child worries that something awful will happen to someone in our family..... 12. My child complains of suddenly feeling as if (s)he can't breathe when there is no reason for this..... 13. My child has to keep checking that (s)he has done things right (like the switch is off, or the door is locked)..... 14. My child is scared if (s)he has to sleep on his/her own..... 15. My child has trouble going to school in the mornings because (s)he feels nervous or afraid..... 16. My child is scared of dogs 17. My child can't seem to get bad or silly thoughts out of his / her head..... 18. When my child has a problem, s(he) complains of his/her heart beating really fast..... 	<ol style="list-style-type: none"> 19. My child suddenly starts to tremble or shake when there is no reason for this..... 20. My child worries that something bad will happen to him/her..... 21. My child is scared of going to the doctor or dentist 22. When my child has a problem, (s)he feels shaky..... 23. My child is scared of heights (eg. being at the top of a cliff)..... 24. My child has to think special thoughts (like numbers or words) to stop bad things from happening..... 25. My child feels scared if (s)he has to travel in the car, or on a bus or train 26. My child worries what other people think of him/her..... 27. My child is afraid of being in crowded places (like shopping centres, the movies, buses, busy playgrounds)..... 28. All of a sudden my child feels really scared for no reason at all..... 29. My child is scared of insects or spiders..... 30. My child complains of suddenly becoming dizzy or faint when there is no reason for this..... 31. My child feels afraid when (s)he has to talk in front of the class..... 32. My child's complains of his / her heart suddenly starting to beat too quickly for no reason 33. My child worries that (s)he will suddenly get a scared feeling when there is nothing to be afraid of..... 34. My child is afraid of being in small closed places, like tunnels or small rooms..... 35. My child has to do some things over and over again (like washing his / her hands, cleaning or putting things in a certain order)..... 36. My child gets bothered by bad or silly thoughts or pictures in his/her head 37. My child has to do certain things in just the right way to stop bad things from happening 38. My child would feel scared if (s)he had to stay away from home overnight..... 39. Is there anything else that your child is really afraid of? <p style="margin-left: 20px;">Please write down what it is, and fill out how often (s)he is afraid of this thing: _____</p> <p style="margin-left: 20px;">_____</p> <p style="margin-left: 20px;">_____</p>
--	--

Appendix O: Intolerance of Uncertainty Scale–Parent (IUS-P)

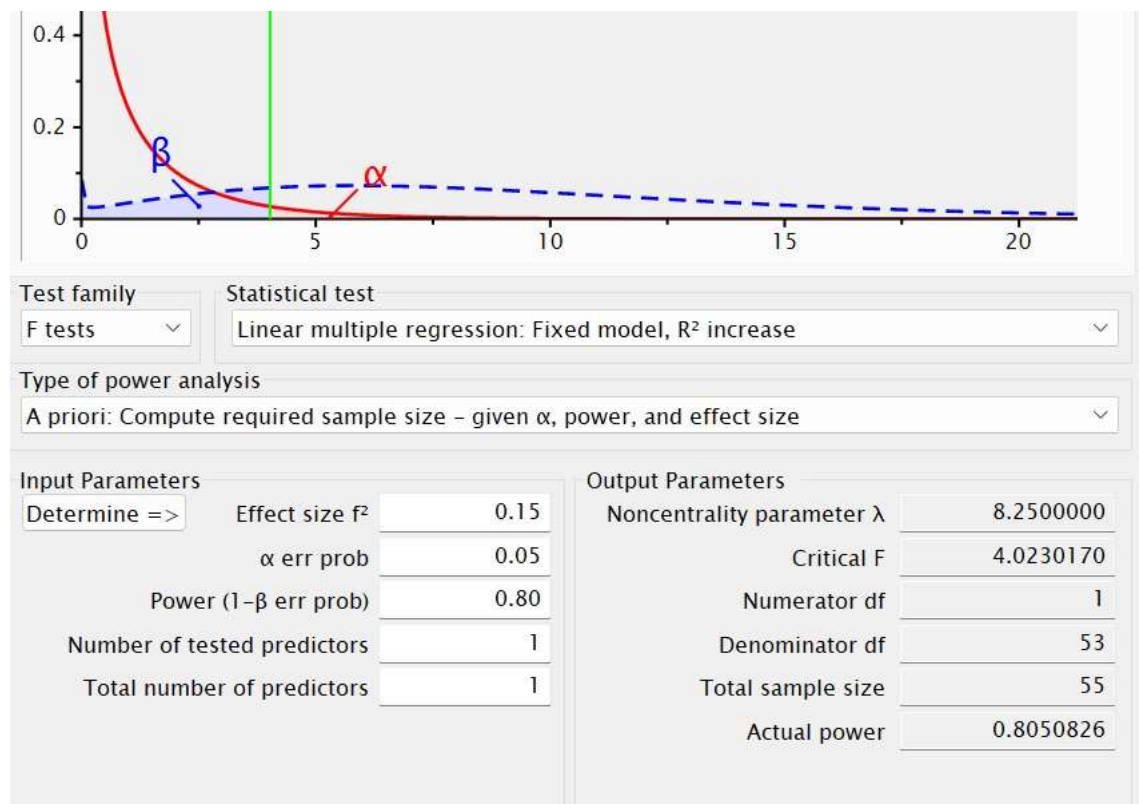
1	Uncertainty stops my child from having strong opinions
2	My child believes that being uncertain means one is mixed-up
3	Uncertainty makes my child's life intolerable
4	My child thinks it's unfair that we can't predict the future
5	My child's mind can't be relaxed if he/she doesn't know what will happen tomorrow
6	Uncertainty makes my child uneasy, anxious, or stressed
7	Unforeseen events upset my child greatly
8	It frustrates my child to not have all the information he/she needs in a situation
9	Uncertainty keeps my child from living a full life
10	My child believes that one should always look ahead so as to avoid surprises
11	My child believes that a small unforeseen event can spoil everything, even with the best planning
12	When it's time to act, uncertainty paralyzes my child
13	My child believes that being uncertain means that he/she is not first rate
14	When my child is uncertain he/she can't go forward
15	When my child is uncertain he/she can't function very well
16	Other children seem to be more certain than my child
17	Uncertainty makes my child unhappy or sad
18	My child always wants to know what the future has in store for him/her
19	My child can't stand being taken by surprise I don't like being taken by surprise
20	The smallest doubt can stop my child from acting
21	My child feels as though he/she should be able to organize everything in advance
22	My child feels as though being uncertain means that he/she lacks confidence
23	My child feels as though it's unfair that other people seem to be sure about their future
24	Uncertainty keeps my child from sleeping soundly
25	My child tries to get away from all uncertain situations
26	The ambiguities of life stress my child
27	My child can't stand being undecided about the future

Appendix P: Short Sensory Profile (SSP)

[removed]

Appendix Q: Details of Statistical Analyses

Power Analysis



Descriptives:

ASSQ and EDA-Q screening groups

ASSQ and EDA-Q screening

		EDA		Total
		Below cut-off	Above cut-off	
ASSQ	Below cut-off	74	22	96
	Above cut-off	210	489	699
Total		284	511	795

Correlation matrix

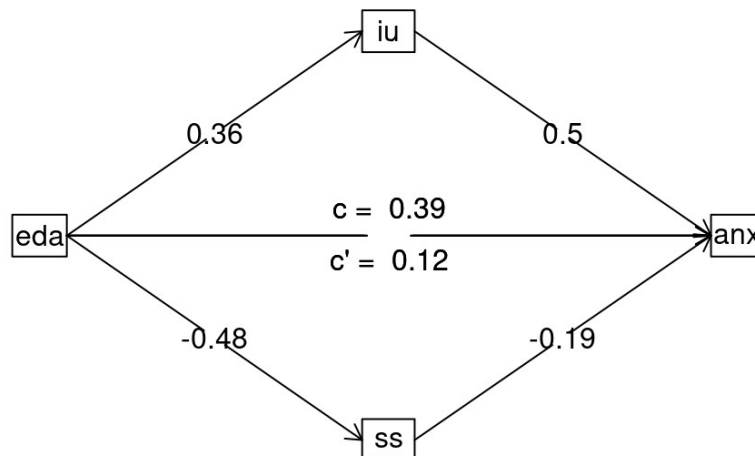
```
##          aut  eda  anx  iu  ss  grops
## aut      1.00
## eda      0.61  1.00
```

Anxiety and EDA

```
## anx      0.37  0.39  1.00
## iu       0.38  0.36  0.63  1.00
## ss      -0.60 -0.48 -0.46 -0.43  1.00
## groups -0.09 -0.08 -0.05 -0.10  0.11  1.00
```

Parallel mediation model

Mediation



```
##
## Mediation/Moderation Analysis
## Call: psych::mediate(y = anx ~ eda + (iu) + (ss), data = pda_data,
##   n.iter = 5000, std = TRUE)
##
## The DV (Y) was  anx . The IV (X) was  eda . The mediating variable(s) =
iu ss .
##
## Total effect(c) of  eda  on  anx  = 0.39  S.E. = 0.03  t = 11.96  d
f= 793  with p = 2e-30
## Direct effect (c') of  eda  on  anx  removing  iu ss  = 0.12  S.E. =
0.03  t = 3.76  df= 791  with p = 0.00019
## Indirect effect (ab) of  eda  on  anx  through  iu ss  = 0.28
## Mean bootstrapped indirect effect = 0.27  with standard error = 0.02
Lower CI = 0.23  Upper CI = 0.32
## R = 0.67  R2 = 0.45  F = 214.42 on 3 and 791 DF  p-value: 1.46e-124
##
## To see the longer output, specify short = FALSE in the print statement
or ask for the summary
## Call: psych::mediate(y = anx ~ eda + (iu) + (ss), data = pda_data,
```

Anxiety and EDA

```
##      n.iter = 5000, std = TRUE)
##
## Direct effect estimates (traditional regression)      (c') X + M on Y
##           anx   se     t  df     Prob
## Intercept  0.00 0.03  0.00 791 1.00e+00
## eda        0.12 0.03  3.76 791 1.85e-04
## iu         0.50 0.03 16.85 791 1.14e-54
## ss        -0.19 0.03 -6.09 791 1.81e-09
##
## R = 0.67 R2 = 0.45   F = 214.42 on 3 and 791 DF   p-value: 9.11e-102
##
## Total effect estimates (c) (X on Y)
##           anx   se     t  df     Prob
## Intercept  0.00 0.03  0.00 793 1e+00
## eda        0.39 0.03 11.96 793 2e-30
##
## 'a' effect estimates (X on M)
##           iu   se  t  df     Prob
## Intercept  0.00 0.03  0 793 1.00e+00
## eda        0.36 0.03 11 793 2.76e-26
##
##           ss   se     t  df     Prob
## Intercept  0.00 0.03  0.00 793 1.00e+00
## eda       -0.48 0.03 -15.47 793 2.15e-47
##
## 'b' effect estimates (M on Y controlling for X)
##           anx   se     t  df     Prob
## iu  0.50 0.03 16.85 791 1.14e-54
## ss -0.19 0.03 -6.09 791 1.81e-09
##
## 'ab' effect estimates (through all mediators)
##           anx boot   sd lower upper
## eda 0.28 0.27 0.02  0.23  0.32
##
## 'ab' effects estimates for each mediator for anx
##           boot   sd lower upper
## eda  0.27 0.02  0.23  0.32
## iu*eda 0.18 0.02  0.14  0.22
```


Anxiety and EDA

```
## ss*eda 0.09 0.02 0.06 0.13
```

SEM modelling in R

All participants

lavaan 0.6-12 ended normally after 47 iterations

Estimator	ML
Optimization method	NLMINB
Number of model parameters	14

Number of observations	795
------------------------	-----

Model Test User Model:

Test statistic	0.202
Degrees of freedom	1
P-value (Chi-square)	0.653

Model Test Baseline Model:

Test statistic	1432.232
Degrees of freedom	10
P-value	0.000

User Model versus Baseline Model:

Comparative Fit Index (CFI)	1.000
Tucker-Lewis Index (TLI)	1.006

Loglikelihood and Information Criteria:

Loglikelihood user model (H0)	-16159.265
Loglikelihood unrestricted model (H1)	-16159.164

Akaike (AIC)	32346.531
Bayesian (BIC)	32412.028
Sample-size adjusted Bayesian (BIC)	32367.570

Root Mean Square Error of Approximation:

RMSEA	0.000
90 Percent confidence interval - lower	0.000
90 Percent confidence interval - upper	0.072
P-value RMSEA <= 0.05	0.863

Standardized Root Mean Square Residual:

SRMR	0.002
------	-------

Parameter Estimates:

Standard errors	Standard
Information	Expected
Information saturated (h1) model	Structured

Regressions:

Anxiety and EDA

	Estimate	Std.Err	z-value	P(> z)	Std.lv	Std.all
ss ~						
eda	-0.294	0.058	-5.085	0.000	-0.294	-0.180
aut	-1.089	0.078	-13.891	0.000	-1.089	-0.491
iu ~						
eda	0.371	0.074	5.032	0.000	0.371	0.206
aut	0.629	0.100	6.293	0.000	0.629	0.257
anx ~						
ss	-0.190	0.032	-5.939	0.000	-0.190	-0.202
iu	0.436	0.025	17.373	0.000	0.436	0.512
eda	0.191	0.054	3.552	0.000	0.191	0.125
aut	-0.036	0.080	-0.443	0.658	-0.036	-0.017

Covariances:

	Estimate	Std.Err	z-value	P(> z)	Std.lv	Std.all
eda ~~						
aut	79.255	5.368	14.765	0.000	79.255	0.615
.ss ~~						
.iu	-88.880	13.443	-6.612	0.000	-88.880	-0.241

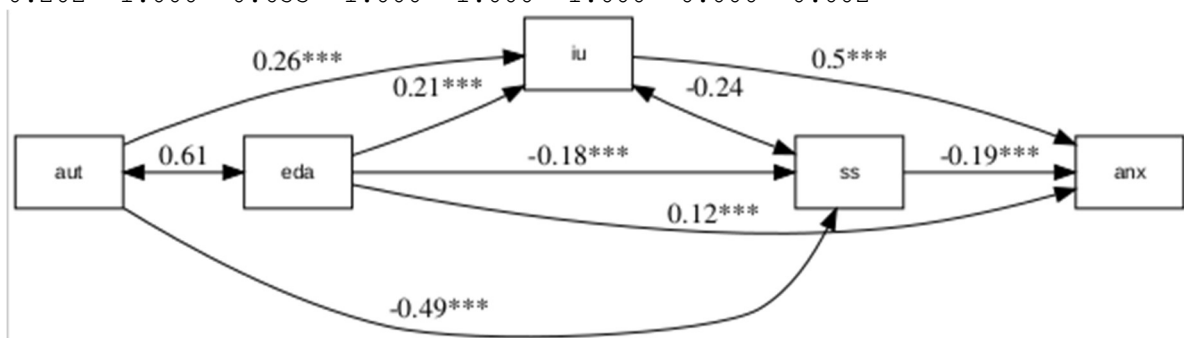
Variances:

	Estimate	Std.Err	z-value	P(> z)	Std.lv	Std.all
.ss	288.890	14.490	19.937	0.000	288.890	0.618
.iu	469.943	23.571	19.937	0.000	469.943	0.826
.anx	235.068	11.790	19.937	0.000	235.068	0.552
eda	174.803	8.768	19.937	0.000	174.803	1.000
aut	95.116	4.771	19.937	0.000	95.116	1.000

Defined Parameters:

	Estimate	Std.Err	z-value	P(> z)	Std.lv	Std.all
tot_iu	0.342	0.057	6.011	0.000	0.342	0.219
tot_ss	0.053	0.056	0.941	0.347	0.053	0.025

chisq df pvalue gfi cfi nfi rmsea srmr
 0.202 1.000 0.653 1.000 1.000 1.000 0.000 0.002



model used - all participants

ANOVA and t-tests

ASSQ

Anxiety and EDA

Mauchly's Test of Sphericity^{a,b}

Measure: MEASURE_1

Within Subjects Effect	Mauchly's W	Approx. Chi-Square	df	Sig.	Epsilon ^c		
					Greenhouse-Geisser	Huynh-Feldt	Lower-bound
ASSQsubscales	.972	14.729	5	.012	.981	.987	.333

Tests the null hypothesis that the error covariance matrix of the orthonormalized transformed dependent variables is proportional to an identity matrix.

a. PDA group = 1.00

b. Design: Intercept
Within Subjects Design: ASSQsubscales

c. May be used to adjust the degrees of freedom for the averaged tests of significance. Corrected tests are displayed in the Tests of Within-Subjects Effects table.

Tests of Within-Subjects Effects^a

Measure: MEASURE_1

Source		Type III Sum of Squares	df	Mean Square	F	Sig.	Partial Eta Squared
ASSQsubscales	Sphericity Assumed	67.334	3	22.445	210.460	<.001	.285
	Greenhouse-Geisser	67.334	2.943	22.879	210.460	<.001	.285
	Huynh-Feldt	67.334	2.961	22.738	210.460	<.001	.285
	Lower-bound	67.334	1.000	67.334	210.460	<.001	.285
Error(ASSQsubscales)	Sphericity Assumed	169.246	1587	.107			
	Greenhouse-Geisser	169.246	1556.886	.109			
	Huynh-Feldt	169.246	1566.544	.108			
	Lower-bound	169.246	529.000	.320			

a. PDA group = 1.00

Paired Samples Test^a

		Paired Differences					Significance			
		Mean	Std. Deviation	Std. Error Mean	95% Confidence Interval of the Difference		t	df	One-Sided p	Two-Sided p
					Lower	Upper				
Pair 1	Social interaction - communication	-.15546	.45936	.01995	-.19466	-.11626	-7.791	529	<.001	<.001
Pair 2	Social interaction - Repetitive and behaviour	.01624	.44085	.01915	-.02138	.05386	.848	529	.198	.397
Pair 3	Social interaction - motor and clumsiness	.33495	.46249	.02009	.29548	.37441	16.673	529	<.001	<.001
Pair 4	communication - Repetitive and behaviour	.17170	.47238	.02052	.13139	.21201	8.368	529	<.001	<.001
Pair 5	communication - motor and clumsiness	.49041	.49721	.02160	.44798	.53284	22.707	529	<.001	<.001
Pair 6	Repetitive and behaviour - motor and clumsiness	.31871	.43603	.01894	.28150	.35592	16.827	529	<.001	<.001

a. PDA group = 1.00

EDA-Q

Anxiety and EDA

Mauchly's Test of Sphericity^a

Measure: MEASURE_1

Within Subjects Effect	Mauchly's W	Approx. Chi-Square	df	Sig.	Epsilon ^b		
					Greenhouse-Geisser	Huynh-Feldt	Lower-bound
EDAQ	.130	1075.673	27	<.001	.634	.640	.143

Tests the null hypothesis that the error covariance matrix of the orthonormalized transformed dependent variables is proportional to an identity matrix.

a. Design: Intercept

Within Subjects Design: EDAQ

b. May be used to adjust the degrees of freedom for the averaged tests of significance. Corrected tests are displayed in the Tests of Within-Subjects Effects table.

Tests of Within-Subjects Effects

Measure: MEASURE_1

Source		Type III Sum of Squares	df	Mean Square	F	Sig.	Partial Eta Squared
EDAQ	Sphericity Assumed	971.011	7	138.716	324.082	.000	.380
	Greenhouse-Geisser	971.011	4.436	218.902	324.082	<.001	.380
	Huynh-Feldt	971.011	4.478	216.842	324.082	<.001	.380
	Lower-bound	971.011	1.000	971.011	324.082	<.001	.380
Error(EDAQ)	Sphericity Assumed	1584.986	3703	.428			
	Greenhouse-Geisser	1584.986	2346.550	.675			
	Huynh-Feldt	1584.986	2368.849	.669			
	Lower-bound	1584.986	529.000	2.996			

Paired Samples Test

		Paired Differences					t	df	Significance	
		Mean	Std. Deviation	Std. Error Mean	95% Confidence Interval of the Difference				One-Sided p	Two-Sided p
					Lower	Upper				
Pair 1	avoidance / controlling interactions - insensitivity to hierarchy praise/reputation with peers	.10472	.59789	.02597	.05370	.15574	4.032	529	<.001	<.001
Pair 2	avoidance / controlling interactions - emotional lability in response to demands or perceived pressure	-.42469	.50460	.02192	-.46774	-.38163	-19.376	529	<.001	<.001
Pair 3	avoidance / controlling interactions - need for control	-.48648	.55146	.02395	-.53353	-.43942	-20.309	529	<.001	<.001
Pair 4	avoidance / controlling interactions - lack of responsibility/blaming	-.16289	.59877	.02601	-.21399	-.11180	-6.263	529	<.001	<.001
Pair 5	avoidance / controlling interactions - mimicry and role play	.67013	.81317	.03532	.60074	.73951	18.972	529	<.001	<.001
Pair 6	avoidance / controlling interactions - distractedness	.18616	1.03364	.04490	.09796	.27436	4.146	529	<.001	<.001

Anxiety and EDA

Pair 7	avoidance / controlling interactions - passivity	.99560	1.14776	.04986	.89766	1.09354	19.970	529	<.001	<.001
Pair 8	insensitivity to hierarchy praise/reputation with peers - emotional lability in response to demands or perceived pressure	-.52940	.60950	.02648	-.58141	-.47739	-19.996	529	<.001	<.001
Pair 9	insensitivity to hierarchy praise/reputation with peers - need for control	-.59119	.57422	.02494	-.64019	-.54220	-23.702	529	<.001	<.001
Pair 10	insensitivity to hierarchy praise/reputation with peers - lack of responsibility/blaming	-.26761	.71459	.03104	-.32859	-.20663	-8.622	529	<.001	<.001
Pair 11	insensitivity to hierarchy praise/reputation with peers - mimicry and role play	.56541	.82502	.03584	.49501	.63581	15.777	529	<.001	<.001
Pair 12	insensitivity to hierarchy praise/reputation with peers - distractedness	.08145	1.06963	.04646	-.00983	.17272	1.753	529	.040	.080
Pair 13	insensitivity to hierarchy praise/reputation with peers - passivity	.89088	1.18255	.05137	.78997	.99179	17.343	529	<.001	<.001
Pair 14	emotional lability in response to demands or perceived pressure - need for control	-.06179	.56570	.02457	-.11006	-.01352	-2.515	529	.006	.012
Pair 15	emotional lability in response to demands or perceived pressure - lack of responsibility/blaming	.26179	.66472	.02887	.20507	.31851	9.067	529	<.001	<.001
Pair 16	emotional lability in response to demands or perceived pressure - mimicry and role play	1.09481	.81824	.03554	1.02499	1.16463	30.803	529	<.001	<.001
Pair 17	emotional lability in response to demands or perceived pressure - distractedness	.61085	1.00288	.04356	.52527	.69643	14.022	529	<.001	<.001
Pair 18	emotional lability in response to demands or perceived pressure - passivity	1.42028	1.18728	.05157	1.31897	1.52159	27.540	529	<.001	<.001
Pair 19	need for control - lack of responsibility/blaming	.32358	.68780	.02988	.26489	.38228	10.831	529	<.001	<.001
Pair 20	need for control - mimicry and role play	1.15660	.83519	.03628	1.08534	1.22787	31.881	529	<.001	<.001
Pair 21	need for control - distractedness	.67264	1.04117	.04523	.58380	.76149	14.873	529	<.001	<.001
Pair 22	need for control - passivity	1.48208	1.16021	.05040	1.38307	1.58108	29.408	529	<.001	<.001
Pair 23	lack of responsibility/blaming - mimicry and role play	.83302	.91224	.03963	.75518	.91086	21.023	529	<.001	<.001
Pair 24	lack of responsibility/blaming - distractedness	.34906	1.03157	.04481	.26103	.43708	7.790	529	<.001	<.001
Pair 25	lack of responsibility/blaming - passivity	1.15849	1.21053	.05258	1.05520	1.26179	22.032	529	<.001	<.001
Pair 26	mimicry and role play - distractedness	-.48396	1.08379	.04708	-.57644	-.39148	-10.280	529	<.001	<.001
Pair 27	mimicry and role play - passivity	.32547	1.23616	.05370	.21999	.43095	6.061	529	<.001	<.001
Pair 28	distractedness - passivity	.80943	1.30417	.05665	.69815	.92072	14.288	529	<.001	<.001

Anxiety and EDA

SCAS-P

Mauchly's Test of Sphericity^{a,b}

Measure: MEASURE_1

Within Subjects Effect	Mauchly's W	Approx. Chi-Square	df	Sig.	Epsilon ^c		
					Greenhouse-Geisser	Huynh-Feldt	Lower-bound
SCASP	.506	359.260	14	<.001	.831	.839	.200

Tests the null hypothesis that the error covariance matrix of the orthonormalized transformed dependent variables is proportional to an identity matrix.

a. PDA group = 1.00

b. Design: Intercept
Within Subjects Design: SCASP

c. May be used to adjust the degrees of freedom for the averaged tests of significance. Corrected tests are displayed in the Tests of Within-Subjects Effects table.

Tests of Within-Subjects Effects^a

Measure: MEASURE_1

Source		Type III Sum of Squares	df	Mean Square	F	Sig.	Partial Eta Squared
SCASP	Sphericity Assumed	372.106	5	74.421	296.707	<.001	.359
	Greenhouse-Geisser	372.106	4.157	89.512	296.707	<.001	.359
	Huynh-Feldt	372.106	4.194	88.722	296.707	<.001	.359
	Lower-bound	372.106	1.000	372.106	296.707	<.001	.359
Error(SCASP)	Sphericity Assumed	663.429	2645	.251			
	Greenhouse-Geisser	663.429	2199.073	.302			
	Huynh-Feldt	663.429	2218.665	.299			
	Lower-bound	663.429	529.000	1.254			

a. PDA group = 1.00

Paired Samples Test^a

		Paired Differences					Significance			
		Mean	Std. Deviation	Std. Error Mean	95% Confidence Interval of the Difference		t	df	One-Sided p	Two-Sided p
					Lower	Upper				
Pair 1	Panic attack and agoraphobia - Separation anxiety	-.83491	.71638	.03112	-.89603	-.77378	-26.831	529	<.001	<.001
Pair 2	Panic attack and agoraphobia - Physical injury fears	-.39258	.71915	.03124	-.45394	-.33121	-12.567	529	<.001	<.001
Pair 3	Panic attack and agoraphobia - Social phobia	-.75189	.69475	.03018	-.81117	-.69260	-24.915	529	<.001	<.001
Pair 4	Panic attack and agoraphobia - Obsessive compulsive	.06164	.54114	.02351	.01546	.10781	2.622	529	.004	.009
Pair 5	Panic attack and agoraphobia - Generalized anxiety disorder	-.52767	.42061	.01827	-.56356	-.49178	-28.882	529	<.001	<.001
Pair 6	Separation anxiety - Physical injury fears	.44233	.72797	.03162	.38021	.50444	13.988	529	<.001	<.001
Pair 7	Separation anxiety - Social phobia	.08302	.84979	.03691	.01051	.15553	2.249	529	.012	.025
Pair 8	Separation anxiety - Obsessive compulsive	.89654	.74538	.03238	.83294	.96014	27.690	529	<.001	<.001
Pair 9	Separation anxiety - Generalized anxiety disorder	.30723	.64983	.02823	.25178	.36268	10.884	529	<.001	<.001

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Pair 10	Physical injury fears - Social phobia	-.35931	.84512	.03671	-.43142	-.28719	-9.788	529	<.001	<.001
Pair 11	Physical injury fears - Obsessive compulsive	.45421	.78417	.03406	.38730	.52113	13.335	529	<.001	<.001
Pair 12	Physical injury fears - Generalized anxiety disorder	-.13509	.71952	.03125	-.19649	-.07370	-4.322	529	<.001	<.001
Pair 13	Social phobia - Obsessive compulsive	.81352	.78841	.03425	.74625	.88080	23.755	529	<.001	<.001
Pair 14	Social phobia - Generalized anxiety disorder	.22421	.70584	.03066	.16398	.28444	7.313	529	<.001	<.001
Pair 15	Obsessive compulsive - Generalized anxiety disorder	-.58931	.58762	.02552	-.63945	-.53917	-23.088	529	<.001	<.001

a. PDA group = 1.00

SSP

Mauchly's Test of Sphericity^a

Measure: MEASURE_1

Within Subjects Effect	Mauchly's W	Approx. Chi-Square	df	Sig.	Epsilon ^b		
					Greenhouse-Geisser	Huynh-Feldt	Lower-bound
SSP	.406	475.438	20	<.001	.790	.798	.167

Tests the null hypothesis that the error covariance matrix of the orthonormalized transformed dependent variables is proportional to an identity matrix.

a. Design: Intercept

Within Subjects Design: SSP

b. May be used to adjust the degrees of freedom for the averaged tests of significance. Corrected tests are displayed in the Tests of Within-Subjects Effects table.

Tests of Within-Subjects Effects

Measure: MEASURE_1

Source		Type III Sum of Squares	df	Mean Square	F	Sig.	Partial Eta Squared
SSP	Sphericity Assumed	1053.073	6	175.512	222.427	<.001	.296
	Greenhouse-Geisser	1053.073	4.740	222.176	222.427	<.001	.296
	Huynh-Feldt	1053.073	4.788	219.945	222.427	<.001	.296
	Lower-bound	1053.073	1.000	1053.073	222.427	<.001	.296
Error(SSP)	Sphericity Assumed	2504.526	3174	.789			
	Greenhouse-Geisser	2504.526	2507.360	.999			
	Huynh-Feldt	2504.526	2532.794	.989			
	Lower-bound	2504.526	529.000	4.734			

Paired Samples Test

		Paired Differences					Significance			
		Mean	Std. Deviation	Std. Error Mean	95% Confidence Interval of the Difference		t	df	One-Sided p	Two-Sided p
					Lower	Upper				
Pair 1	Tactile Sensitivity - Taste/Smell Sensitivity	10.925	6.017	.261	10.411	11.438	41.795	529	<.001	<.001
Pair 2	Tactile Sensitivity - Movement Sensitivity	8.077	5.213	.226	7.632	8.522	35.669	529	<.001	<.001
Pair 3	Tactile Sensitivity - Underresponsive/Seeks Sensation	-1.006	8.319	.361	-1.716	-.296	-2.783	529	.003	.006

Anxiety and EDA

Pair 4	Tactile Sensitivity - Auditory Filtering	6.528	5.786	.251	6.035	7.022	25.976	529	<.001	<.001
Pair 5	Tactile Sensitivity - Low Energy/Weak	.319	7.785	.338	-.345	.983	.943	529	.173	.346
Pair 6	Tactile Sensitivity - Visual/Auditory Sensitivity	6.638	5.111	.222	6.202	7.074	29.901	529	<.001	<.001
Pair 7	Taste/Smell Sensitivity - Movement Sensitivity	-2.847	5.476	.238	-3.314	-2.380	-11.969	529	<.001	<.001
Pair 8	Taste/Smell Sensitivity - Underresponsive/Seeks Sensation	-11.930	7.890	.343	-12.603	-11.257	-34.812	529	<.001	<.001
Pair 9	Taste/Smell Sensitivity - Auditory Filtering	-4.396	5.457	.237	-4.862	-3.931	-18.546	529	<.001	<.001
Pair 10	Taste/Smell Sensitivity - Low Energy/Weak	-10.606	7.794	.339	-11.271	-9.941	-31.327	529	<.001	<.001
Pair 11	Taste/Smell Sensitivity - Visual/Auditory Sensitivity	-4.287	5.492	.239	-4.755	-3.818	-17.970	529	<.001	<.001
Pair 12	Movement Sensitivity - Underresponsive/Seeks Sensation	-9.083	7.651	.332	-9.736	-8.430	-27.329	529	<.001	<.001
Pair 13	Movement Sensitivity - Auditory Filtering	-1.549	4.873	.212	-1.965	-1.133	-7.318	529	<.001	<.001
Pair 14	Movement Sensitivity - Low Energy/Weak	-7.758	6.751	.293	-8.335	-7.182	-26.458	529	<.001	<.001
Pair 15	Movement Sensitivity - Visual/Auditory Sensitivity	-1.440	4.842	.210	-1.853	-1.026	-6.844	529	<.001	<.001
Pair 16	Underresponsive/Seeks Sensation - Auditory Filtering	7.534	6.805	.296	6.953	8.115	25.487	529	<.001	<.001
Pair 17	Underresponsive/Seeks Sensation - Low Energy/Weak	1.325	10.143	.441	.459	2.190	3.006	529	.001	.003
Pair 18	Underresponsive/Seeks Sensation - Visual/Auditory Sensitivity	7.643	7.513	.326	7.002	8.285	23.420	529	<.001	<.001
Pair 19	Auditory Filtering - Low Energy/Weak	-6.209	7.743	.336	-6.870	-5.549	-18.463	529	<.001	<.001
Pair 20	Auditory Filtering - Visual/Auditory Sensitivity	.109	4.456	.194	-.271	.490	.565	529	.286	.572
Pair 21	Low Energy/Weak - Visual/Auditory Sensitivity	6.319	7.725	.336	5.660	6.978	18.832	529	<.001	<.001

Appendix R: Dissemination

An overview of this research project was presented at the LMS research conference at the University of Hertfordshire. Slides are presented below. Further dissemination to journal articles and an accessible summary sheet for the PDA Society and PDA community is planned; however, at the time of writing this is remains ongoing.

1

ANXIETY AND EXTREME DEMAND AVOIDANCE IN CHILDREN AND ADOLESCENTS: THE MODERATING ROLES OF SENSORY SENSITIVITIES AND INTOLERANCE OF UNCERTAINTY

AARON J. RAI
SUPERVISED BY DR. AMANDA LUDLOW & DR. BARBARA RISHWORTH

2

WHAT IS PDA?

- A controversial diagnosis that has been described in clinical practice but not recognised by official diagnostic and statistical manuals
- The National Autistic Society (UK) describes PDA as **'a profile of Autism'**.
- "A profile of obsessive resistance to everyday demands and requests, with a tendency to resort to *socially manipulative* behaviour" (O'Nions et al, 2014)

3

PREVALENCE

- Context of Autism: changes to diagnostic criteria and assessment practices in different countries could explain variance in prevalence studies. However, Baron-Cohen et al. (2009) estimate 1 in 100.
- PDA: one prevalence study from Faroe Islands (Gillberg et al., 2015): 9 of 59 children and adolescents with Autism met criteria for PDA retrospectively. At time, only 1 (one) met full criteria', suggesting behaviours may reduce over time.
 - Is this pervasive (lifelong) like Autism? Or do individuals with PDA learn to live with PDA?
- Notably, there has been a reported 'surge of interest' / requests for assessment etc, since 2010 (O'Nions et al., 2018).

4

PDA: 'DRIVEN BY ANXIETY'

- PDA is described as 'driven by anxiety' by the PDA Society.
- A better understanding of the underlying factors contributing to anxiety may support more effective treatment strategies.
- Why research this?
 - There is some evidence that strategies for supporting those with Autism may be less effective for those identifying with PDA profile (PDA Society, 2018).
 - There is controversy and debate as to what PDA even is. Neurodevelopmental? Behavioural?

Anxiety and EDA

AIMS

- To investigate the relationship between Autistic traits (ASSQ; Ehlers et al., 1999) and Extreme Demand Avoidance (EDA) behaviours (EDA-Q; O’Nions et al., 2014).
- Explore the contributing roles of both Intolerance of Uncertainty (IU) and Sensory Sensitivities (SS) to levels of anxiety in those who identify with Pathological Demand Avoidance (PDA).
- No research to date has empirically investigated the role of Sensory Sensitivities in PDA

5

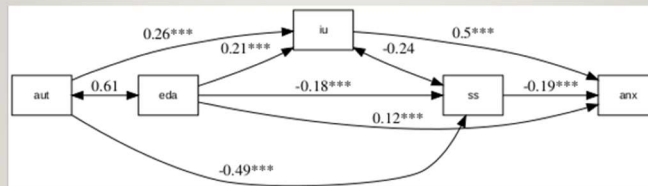
METHODS

- PDA Society support – design and recruitment
- Online survey - Parent report measures: ASSQ, EDA-Q, SCAS-P, IUS-P, SSP.
 - 795 participants aged 4-17 years old: 530 identified with PDA (475 identified with PDA and Autism)
- Data Analysis:
 - Correlational
 - Data driven Structural Equation Modelling (SEM)
 - Analysis of subscales

6

RESULTS

- Autistic traits and EDA behaviours were highly correlated.



7

INFERENCES

- PDA shows discriminant properties to Autism but should be seen within the context of Autism. This supports the concept of PDA as 'a profile of Autism'
- Understanding **sensory needs** as a key underlying mechanism behind Anxiety in those identifying with PDA can support tailored, comprehensive assessment schedules and more individualised behaviour management strategies for this population.

8

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