

Dimensions of difficulty in children reported to have an autism spectrum diagnosis and features of extreme/'pathological' demand avoidance

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Background: A subset of individuals with autism spectrum disorder (ASD) resemble descriptions of extreme/'pathological' demand avoidance, displaying obsessive avoidance of everyday demands and requests, strategic or 'socially manipulative' behaviour and sudden changes in mood. Investigating challenging presentations using dimensional description may prove preferable to identifying subgroups. However, there remains an imperative to explore which behavioural traits appear most problematic to inform quantitative investigation. This study provides an in-depth exploration of parent perspectives on maladaptive behaviour in children reported to have an autism spectrum diagnosis and features of extreme/'pathological' demand avoidance.

Method: Parents completed a tailored semistructured interview about their child's behaviour, focusing on difficulties relevant to descriptions of extreme/'pathological' demand avoidance. The 26 interviews rated as scoring above threshold for 'substantial' features of extreme/'pathological' demand avoidance on relevant indicators were analysed qualitatively using a general inductive approach. **Results:** New themes that emerged from these data included attempts by the child to control situations and others' activities. Avoidance behaviours in this sample could be described as 'strategic' rather than 'manipulative'. A range of factors, including a negative emotional response to demands, but also phobias, novelty, and uncertainty, were perceived to play a role in triggering extreme behaviour. **Conclusions:** These descriptions highlight the importance of systematically measuring noncompliance, attempts to control situations and others' activities, and extreme mood variability in individuals with ASD. These dimensions represent important targets for intervention, given their considerable impact on daily life.

Key Practitioner Message

- Children with ASD who resemble descriptions of extreme/'pathological' demand avoidance obsessively avoid complying with everyday demands, engage in apparently 'manipulative' behaviour and exhibit sudden changes in mood. We collected interview data from parents of children with ASD (parent-reported) and these features about their child's maladaptive behaviour.
- Subtle differences emerged compared to previous accounts of this profile. These included additional problems relating to the child's frequent attempts to control situations and others' activities (e.g. by insisting that others abide by their rules or meet their demands). In our sample, avoidance behaviours could be described as 'strategic' rather than 'manipulative'. Parents perceived that a range of factors played a role in triggering extreme behaviour, including a negative emotional response to perceived pressure (in line with previous accounts), as well as sensory sensitivities, phobias and anxiety about the unknown.
- These findings add to the literature on extreme noncompliance, mood variability and attempts to control situations and others' activities in ASD, and they motivate examination of similarities and differences in the manifestation and drivers of these behaviours compared with other non-ASD profiles. Our data highlight the importance of these dimensions as intervention targets, given their considerable impact on daily life.

Keywords: Autism spectrum disorder; extreme demand avoidance; pathological demand avoidance; noncompliance; mood variability; meltdowns

Introduction

The term 'pathological demand avoidance' was coined by Elizabeth Newson, a UK-based psychologist, to describe children on the spectrum of pervasive developmental disorders who obsessively resist everyday demands and use 'socially manipulative' behaviour to avoid complying. Other features include a lack of sense of identity, pride or shame; extreme mood variability; a tendency to appear comfortable in role play and pretend; and obsessive behaviour (Newson, Le Maréchal, & David, 2003). Those with ASD who most resemble this profile engage in more socially maladaptive behaviours and have worse anxiety/emotional symptoms than others on the spectrum (O'Nions, Viding, Greven, Ronald, & Happé, 2014; O'Nions et al., 2016). Demand avoidance with 'socially manipulative'/shocking behaviour is thought to occur in around 1 in 25 individuals with ASD, with 1 in 5 displaying some behaviours (Gillberg, Gillberg, Thompson, Biskupsto, & Billstedt, 2015).

Numerous reports describe behavioural features associated with extreme/'pathological' demand avoidance in ASD. Severe noncompliance and rages are usually framed either as comorbid disruptive behavioural disorders (e.g. Agazzi, Tan, & Tan, 2013; Armstrong & Kimonis, 2013) or as an extreme response to stressors (e.g. disrupted routines, nonpreferred activities) that is in line with the heightened sensitivity characteristic of ASD (e.g. DeGrace, 2004; Gray, 1993; Hodgetts, Nicholas, & Zwaigenbaum, 2013; Larson, 2006). Noncompliance with everyday demands is now a major treatment target (e.g. Arnold et al., 2012; Bearss et al., 2015; Chowdhury et al., 2010; Scahill et al., 2012).

Several studies report that some children with ASD experience simple tasks and normative parental expectations as aversive (Lucyshyn et al., 2004, 2015; Moes & Frea, 2000). Anxiety, arousal and escape appear to play a key role in driving avoidance of demands in ASD – distinct from noncompliance in disruptive behaviour disorders (Lucyshyn et al., 2004). However, reports also describe attention- or reward-driven problem behaviour, which does resemble that seen in disruptive behaviour disorders, for example, oppositional defiant disorder (Armstrong, DeLoatche, Preece, & Agazzi, 2015; Barry & Singer, 2001; Lucyshyn et al., 2004).

Investigating challenging presentations of ASD using dimensional description may well prove helpful in developing the evidence base. However, there remains an imperative to explore which behavioural traits are most problematic in individuals with challenging profiles, including those resembling descriptions of extreme/'pathological' demand avoidance, to guide these efforts. This study reports a systematic analysis of semistructured interview data from parents of children reported to have an autism spectrum diagnosis and features of extreme/'pathological' demand avoidance. Exemplars drawn from interviews provide insight into how behavioural features manifest in daily life.

Methods

Setting

The study was part of a larger investigation of extreme demand avoidance in a community sample in the United Kingdom. Ethical approval was obtained from the King's College London

Psychiatry, Nursing and Midwifery Research Ethics review board. The study was conducted in accordance with the declaration of Helsinki. Participants gave informed consent to participate.

Participants and procedure

Eighty-two parents of children recruited from community settings with a range of neurodevelopmental/behavioural difficulties completed a screening questionnaire, including a short version of the Extreme Demand Avoidance Questionnaire (O'Nions, Christie, Gould, Viding, & Happé, 2014), consisting of 11 items (items 1, 3, 9, 10, 11, 14 (Reversed), 15, 18, 21, 22, and 25 of the original version), as part of a larger programme of research. Parents of children reported to have an autism spectrum diagnosis and features of extreme/'pathological' demand avoidance invited to complete the interview study were those recruited via specialist conferences ($N = 6$), enquiries to the team ($N = 4$), educational/child psychologists ($N = 3$), web groups ($N = 13$) or specialist educational provisions ($N = 3$). As such, parents were at least slightly familiar with extreme/'pathological' demand avoidance. We made particular efforts to include parents of girls, given the reported equal gender balance associated with this profile (Newson et al., 2003).

Twenty-nine parents of children with an autism spectrum diagnosis reporting features of extreme/'pathological' demand avoidance aged 7 years 10 months to 16 years 11 months completed the semistructured interview ($N = 12$ by telephone, $N = 17$ using a self-administered electronic or written format). The interview was developed for research purposes to capture behaviours relevant to pathological demand avoidance as described by Newson et al. (2003). It has not been validated and is not presumed to assay behaviours that are necessarily specific to this presentation. It consisted of 22 questions, with 55 subquestions, drawing on content from the Diagnostic Interview for Social and Communication Disorders (DISCO; Wing, Leekam, Libby, Gould, & Locombe, 2002) and from an interview developed by Newson et al. (2003). Parents completed the interview by commenting on whether their child displayed the target behaviours and providing relevant examples of their child's behaviour. For telephone interviews, full verbatim transcripts were not available. Interviews completed in self-administered written format and notes from telephone interviews were transcribed.

Interview responses were first coded against the 11 indicators relevant to extreme/'pathological' demand avoidance from the Diagnostic Interview for Social and Communication Disorders. Interviews were included in the analysis if the 'substantial features' cut-off was met (O'Nions et al., 2016). Ratings were made by two independent raters, with detailed scoring advice provided by J. Gould. Codes agreed on the basis of a consensus discussion are in Table S2 and interrater agreement in Table S3. Difficulties reaching consensus for severity ratings for some items emerged in part because collecting data electronically or in written format precluded clarification. The complexity of reported behaviour also presented coding challenges (e.g. if the child did not routinely target or bully others, but had previously engaged in episodes of acute violence towards peers). The 26 interviews that met the cut-off based on consensus agreement were completed for children with a mean age of 11 years 0 months ($SD = 2$ years 8 months), 13 of whom were boys (50%) and 13 girls (50%). Thirteen parents (50%) had an undergraduate degree (data unavailable for $N = 4$). To the best of our knowledge, none of the 26 children had experienced acute environmental adversity (e.g. maltreatment, neglect).

IQ testing using the 2-subtest Wechsler Abbreviated Scales of Intelligence (Wechsler, 1991) was attempted with 23/26 children (88%) during a research session conducted at the child's home or school. The majority of participants were in the normal range for intellectual ability (Table 1), but only four attended mainstream school with no additional help. Children were reported to exhibit high levels of difficult behaviour, and educational placement breakdown was common in the sample.

Analysis

The purpose of the qualitative analysis was to identify maladaptive behaviours from parental descriptions, focusing particularly on aspects of behaviour relevant to extreme/‘pathological’ demand avoidance. A general inductive approach was used to identify overarching themes and individual exemplars (Thomas, 2006). This involved using a subset of interviews to generate a preliminary coding scheme of behaviours that represent common themes. Additional subthemes were then added and existing ones modified during a full review of the remaining interviews. Very specific subthemes were then collapsed to form more general concepts, and peripheral subthemes (i.e. more idiosyncratic and/or less relevant to emergent thematic structure) were pruned. During this process, 180 identified subthemes were reduced to the 90 described here. Thirty-nine of the subthemes relate specifically to probe questions, the remainder reflect emergent concepts. Subthemes were organised into descriptive themes as they were developed. The thematic structure was reorganised during the writing up of results. Further consideration of the validity of the analysis is provided in the Supplemental Methods (Appendix S1).

Results

Behavioural exemplars organised into subthemes are presented below, together with their frequency in the sample. Not all parents provided information pertinent to all identified constructs. As such, numerical data should be viewed as an estimate of the minimum occurrence in the sample. Readers are reminded that children were aged 7 years 10 months to 16 years 11 months (mean age 11 years 0 months) at the time of the interview and were reported to have an autism spectrum diagnosis.

Noncompliance and insistence that others comply with their wishes

Onset of noncompliance. Parents gave diverging accounts of the development of noncompliance. More than a third of parents (10/26) reported that their child had been a placid, easy baby, but just over a quarter (8/26) had reportedly been difficult and demanding. Nearly half of parents (11/26) noted that extreme stubbornness and refusal to comply was evident at nursery, although not always considered ‘beyond the norm’. Around a quarter of children (7/26) resisted passively or withdrew from activities, while a third (9/26) responded to attempts to impose limits with outrage. More than a third of parents (10/26) perceived that noncompliance had worsened with increasing demands for conformity at primary school. In two cases, things deteriorated at a later stage (‘It was really at age nine when he realised that people can’t make you do something’ [3]).

Avoidance of everyday demands. All parents reported that everyday activities (e.g. getting up, brushing teeth, eating meals with family) provoked resistance. For some, every request was avoided, while for others, avoidance was more specific to particular activities (e.g. refusal to visit new places, reluctance to make transitions: ‘Even going from the house to the car provokes anxiety: he tries to collect things to take with him’ [9]).

Variability in resistance from day to day was noted by around a third of parents (9/26; e.g. ‘A bad day at school where she has not reacted will result in her being ultra-controlling, short tempered, avoidant and manic in her behaviour with me’ [17]). Situational factors, including

novelty, anticipation or something being different reportedly exacerbated noncompliance in a similar proportion (10/26).

Compliance ‘on their terms’. Almost all parents (22/26) noted that their child needed everything to be done ‘on their terms’ (e.g. ‘He must be in control of everything, even the smallest of things such as picking a sweet or brushing teeth’ [23]). More than half (14/26) reported that it was impossible to appeal to their child’s ‘better nature’ – compliance depended on the activity suiting the child’s agenda (e.g. ‘If we want something to happen, we have to plan it carefully, distract him with his interests, or if it is something he absolutely must do (e.g. attend an appointment), bribe him’ [9]). Pressure to conform appeared to provoke frustration (e.g. ‘She cut off a piece of her own hair as she was under extra pressure to get to school on time’ [14]).

Nearly half (11/26) of parents reported that their child appeared to dislike praise or recognition (e.g. ‘once he realised he wasn’t in control the rewards seemed meaningless’ [10]). Two parents recounted an incident where behaviour had worsened after their child had received a prize or earned something for behaving well.

Insistence that others comply with their wishes/attempts to control others’ activities. The vast majority of children (23/26) were reportedly rigid in their insistence that others comply with their wishes. This was evident in a tendency to monopolise conversations (e.g. ‘He did this at a school meeting with three adults present. They kept calm and had a logical discussion, but he shouts you down and takes over’ [3]). Need for control was also evident during games: nearly a third (8/26) had ‘intense reactions’ to losing, changing rules or insisting that play continued until they won. For a similar proportion, family members were forced to take on specific roles in games (e.g. ‘With his younger brothers, he wants them to play with him but they have to do exactly the right thing... He will go ballistic if they don’t do what he wants’ [8]).

More than three quarters (22/26) displayed obsessive or controlling behaviour towards family members or favoured individuals (e.g. ‘She doesn’t want me to do anything without her permission, and wants to know what I’m doing when I’m not with her’ [1]). Nearly half (12/26) displayed jealousy over attention, which often led to disruptive behaviour (e.g. ‘If I’m on the phone, she’ll repeat everything I say so that I can’t concentrate’ [14]).

Over three-quarters of parents (21/26) reported that their child was bossy and controlling towards peers (e.g. ‘His ‘friends’ are those who like his interests and abide by his rules’ [25]). More than a third (11/26) had alienated themselves through controlling or infantile behaviour, and a similar proportion (10/26) resisted engaging with peers at all (e.g. by refusing to go to school or attend class). Ten preferred to interact with younger/less able children, who were easier to control and more accepting of domineering behaviour.

Strategic and disruptive behaviour in response to requests or limits

Strategic behaviour. Parents reported that their child employed a wide range of strategic behaviours to subvert requests (e.g. ‘Having to get ready for school, she would

Table 1. Details of sample ($N = 26$)

ID	Age (year; month)	Diagnoses reported by parents	Educational provision	Reported intellectual disability or estimated IQ	EDAQ short version
1	12; 01	ASC	Mainstream	Vocabulary & Matrices – well above average	27/33
2	15; 04	ASC	Excluded	Vocabulary – poor compliance; Matrices – average	32/33
3	13; 04	ASC	Specialist	Poor compliance	25/33
4	9; 11	ASC	Specialist	Poor compliance/test unsuitable	25/33
5	11; 11	ASC, ADHD	Mainstream ^a	Vocabulary – high average; Matrices – low average	27/33
6	8; 04	ASC, PDA	Mainstream	Poor compliance	29/33
7	8; 10	ASC, PDA, dyslexia	Mainstream ^a	Vocabulary – low average; Matrices – average	32/33
8	8; 03	ASC	Not collected	Vocabulary and Matrices – high average	26/33
9	8; 10	ASC, sensory, challenging behaviour	Excluded	Vocabulary – high average; Matrices – well above average	21/33
10	13; 08	ASC, ADHD, Tourettes	Excluded	Vocabulary – low average; Matrices – average	27/33
11	14; 11	ASC, dyspraxia	Mainstream ^a	Moderate ID (reported)	22/33
12	10; 00	ASC	Mainstream	Vocabulary and Matrices – High average	29/33
13	9; 01	ASC	Mainstream	Vocabulary – upper extreme; Matrices – well above average	21/33
14	16; 11	ASC, epilepsy	Specialist	Moderate ID (reported)	30/33
15	8; 03	ASC, dyspraxia	Home schooled	Vocabulary – low average; Matrices – high average	27/33
16	11; 03	ASC	Home schooled	Vocabulary – average; Matrices – well above average	22/33
17	8; 04	ASC, PDA	Mainstream ^a	Vocabulary – poor compliance; Matrices – well above average	32/33
18	12; 05	ASC, PDA, challenging behaviour	Specialist	Vocabulary and Matrices – Average	25/33
19	9; 07	ASC, ADHD	Specialist	Vocabulary – poor compliance; Matrices – average	29/33
20	14; 10	ASC, paranoia	Specialist	Not collected	17/33
21	10; 01	ASC	Specialist	Test unsuitable	30/33
22	9; 09	ASC, ADHD, PDA	Specialist	Vocabulary – low average; Matrices – average	24/33
23	7; 10	ASC, anxiety, OCD	Mainstream ^a	Not collected	21/33
24	13; 02	ASC, ADHD, PDA	Out of school	Not collected	23/33
25	9; 05	ASC, ADHD	Mainstream ^a	Not collected	24/33
26	14; 00	ASC, immature behaviour and emotions	Specialist	Vocabulary and Matrices – borderline	27/33

^aMainstream school with additional support/modifications, for example 1:1 staff member, restricted timetable. IQ measured using the two-subtest Wechsler Abbreviated Scales of Intelligence (WASI); alternate Wechsler IQ classifications reported (Groth-Marnat, 2009). EDAQ, Extreme Demand Avoidance Questionnaire; ASC, autism spectrum condition (e.g. autism, high functioning autism, Asperger's, atypical autism); PDA, pathological demand avoidance; ID, intellectual disability; OCD, obsessive compulsive disorder.

hit, bite or shout at others or use more passive behaviour such as complaining that her legs ached' [22]). Common strategies included distraction or diversion (20/26), point-blank refusal or arguing (19/26) and passivity (18/26); (e.g. 'I am now realising that I have been physically moving her, when she has not been compliant, for many years' [13]). Five used noise to block out requests, and eight withdrew or ran off (e.g. 'He knows his way home from school. He has also disappeared out of the caravan' [9]).

Three quarters of children (20/26) reportedly made excuses to avoid complying (e.g. 'If you challenge the excuse he will come up with another one and so on' [18]). For a minority (6/26), excuses were spurious or bizarre (e.g. 'I'm a baby' [11]). Two-thirds (17/26) reportedly used charm (e.g. 'He is very subtle and will play on mothering instincts: he is hungry, he is tired, he doesn't feel well, he needs a drink.' [7]). Other strategies included accusations (e.g. claiming that requests were 'killing her', [19]), partial compliance or setting conditions (e.g. agreeing to apologise only if her mother let her hug her first, [13]).

Two-thirds of children (17/26) attempted to 'outmanoeuvre' parents with strategic behaviour, such as attempting to play parents off against each other, manipulating rules or making it impossible for themselves to comply ('He didn't want to do the work so he pushed out the lens of his glasses so he couldn't wear them any longer' [24]). Just over a quarter (7/26) argued that they did not need to comply because the rules did not apply to them (e.g. 'Although we explained to her that children were not allowed to sit there, she still believed that she should be exempt from this as she wasn't a child' [12]).

Aggressive behaviour. All children reportedly resorted to tantrums, rages or outbursts of verbal aggression if pushed to comply or accept limits. This frequently involved disruptive, violent or explosive behaviour towards others (e.g. 'He shocks sometimes by having a complete meltdown in public, punching, kicking, hitting me' [7]) or directed at themselves (e.g. 'He has self-harmed at school, when under stress. This included hitting his head against a wall, pulling out

his hair, and cutting and scratching his face until it bled' [23]).

Around three quarters of parents (19/26) reported that aggressive behaviour had an impulsive quality. However, more than half (15/26) felt that threats were also sometimes used to exert control (e.g. 'If you don't . . . I will hit you a million times' [15]). Bullying or targeting of others, including siblings, was reported in more than half (14/26), and more than a third (9/26) reportedly displayed anger towards peers if they refused to obey. Almost a third of parents (8/26) reported that during a meltdown, there were no limits on what the child might resort to.

Threats or extreme behaviour. Threats to hurt themselves (e.g. by jumping out of windows/vehicles, throwing themselves down the stairs), or actual self-injury, were reported by almost all parents (23/26). Threats were frequently made in the context of being pressed to comply or conform. However, not all parents believed that their child would follow through on threats, or understood the implications of what they were saying. Extreme acts other than violence (urinating on other people's possessions, sexually inappropriate actions, setting off fire-extinguishers) had occurred in just over a quarter (7/26).

Sudden, dramatic changes in mood

Dramatic mood switches to minor events were reported by all parents (e.g. 'He goes from 0 to 100 in seconds, with no obvious triggers. This can be anywhere and at any time' [25]). More than three-quarters (20/26) reportedly switched suddenly from loving or playful to aggressive behaviour (e.g. 'She can be cuddling 1 minute and then kicking you away quite hard the next' [21]). The majority (19/26) also sometimes became overexcited or 'hyper' (e.g. '[He] does loops around the house whooping. If you try to stop it, you may get full blown aggression' [8]).

Demands for conformity or things not being on their terms were identified as common triggers for switches in mood. However, for more than half of the sample (15/26), precise triggers were often hard to identify or appeared to relate to other factors (e.g. 'Triggers can include a new place; an unusual smell; babies and toddlers (he fears them) [23]'; 'Excitement about something, as well as fear about the unknown often provokes a violent outburst' [12]). Aggressive outbursts were also sometimes perceived to be a culmination of distress resulting from real or perceived provocation. Several parents reported that getting wound up easily made the child an 'easy target' (e.g. 'Peers (and their parents) have viewed him as the 'mad child', laughing at him when he gets cross' [9]). Nearly a third (8/26) had reportedly been victims of taunting or bullying.

Lack of constraint by social norms or sense of hierarchy

Behaving as though they had adult status. All parents reported that their child behaved as though they had equal status with adults, which frequently led to arguing with requests or rules, or attempting to tell adults what to do. The vast majority (22/26) were reportedly insensitive to authority, ignoring remonstrations from staff at school. Intriguingly, nearly three quarters (19/

26) of parents also felt that their child would not make allowances for younger children (e.g. 'He thinks a toddler is being nasty to him so will retaliate' [6]). Some children appeared to lack any concept of age or hierarchy, while others seemed to have a theoretical awareness: (e.g. 'Now I would say she has some awareness of hierarchies, and could explain who is in charge of whom, but does not feel much need to be constrained by them' [11]).

Failure to constrain behaviour in public. All but one child reportedly resorted to behaviour that others of their age would find embarrassing in public (e.g. lying down in the supermarket, saying embarrassing things or having a meltdown in front of peers). Just under one-third (8/26) reportedly behaved in embarrassing ways to gain attention, for example, drawing attention to themselves by behaving foolishly or inappropriately (e.g. '[He] presents himself to shock others and likes the reaction he gets by people being shocked/embarrassed by his behaviours' [20]). Intriguingly, despite failing to apply social constraints to themselves, nearly two-thirds of children (16/26) reportedly insisted that other children conform with rules.

Lack of sense of responsibility or sensitivity to others' distress

Blaming circumstances or other people for own misbehaviour. Many parents expressed concern about their child's very limited sense of responsibility. All but two children (24/26) reportedly tried to pass blame for their own behaviour onto others or to circumstances beyond their control. For a large proportion (21/26), this seemed to have an irrational quality (e.g. 'Her accusations are more that anything we have done must have been intentional, it cannot be accidental – so the complete inverse of how she sees things relating to herself' [15]). A minority (6/26) perceived hostile intents in people or objects excessively, and two displayed paranoia about others attempting to injure them, or covertly monitoring them.

Several parents reported that their child used elaborate justifications to blame people for events they were unconnected to. Targets were often particular disliked individuals. Just over three quarters of parents (20/26) felt that they could not trust their child to tell the truth – instead they would get the truth 'as s/he sees it'. Four reportedly denied things even when others had witnessed them, and more than a third (10/26) made false accusations ('She will make things up and tell tales to get her brother into trouble, and then wants me to tell him off in front of her' [17]).

Lack of sensitivity to others' distress. Almost two-thirds of children (17/26) were said to be unconcerned or unaware when others were upset or in pain (e.g. 'He does not always recognise others emotions if he did not instigate the mood himself' [26]). More than three quarters (20/26) responded inappropriately to others' distress, either failing to react or becoming more difficult (e.g. 'If someone was feeling ill or low, it seemed to be picked up by X and taken as a signal to behave with total lack of consideration' [19]). One quarter (7/26) sometimes behaved in a superficially caring manner, but 'on their terms', for example, insisting that this took on a

role-play quality. However, around a fifth (5/26) reportedly displayed concern and liked to be helpful.

Controlling reality: fantasy and role play

Engagement in fantasy activities. Nearly two-thirds (16/26) of children reportedly engaged in fantasy activities (e.g. making up stories/scenarios), which were frequently elaborations of things that they had observed. Several insisted that family members take on specific roles (e.g. 'When she knows she is visiting her cousins she will prepare by writing out scripts for plays that she imagines they will perform' [11]). Just over a third (9/26) re-enacted scenes from TV or real events rigidly ('She has memorised large chunks of the scripts and will get very angry with people when they want to change the story' [12]). One child [16] reportedly found screen-based games 'intoxicating', which was attributed to the fact that he has complete control.

Confusing reality and fantasy. Nearly half of parents (12/26) reported that their child seemed to take engagement with fantasy 'too far', either persisting in adopting a role after play had ended, or confusing what had happened in play with reality. Just over a third of children (9/26) had a strong relationship with a toy or fictional character (e.g. 'No-one is allowed to touch [the toy] or they get really hurt, she has run away from school due to another child moving it' [5]).

Acting out personas and styles. Nearly three-quarters of the sample (19/26) reportedly took on borrowed personas or styles. This included adopting the characteristics of other people (e.g. 'If she gets a group of people together in a room, she acts like a very strict teacher, telling people off and to be quiet' [14]). A minority (6/26) appeared to adopt compliant personas (e.g. 'He has also found that he can control a situation by behaving well, for example, acting a part, not natural behaviour' [8]). Nearly two-thirds (16/26) used borrowed phrases when interacting with others (e.g. 'Her favourite is "NO" with pointed finger' [5]).

Discussion

This study provides an in-depth exploration of parent perspectives on maladaptive behaviours in children reported to have an autism spectrum diagnosis and features of extreme/'pathological' demand avoidance. Parental accounts were broadly consistent with previous descriptions, although several novel themes emerged. First, 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. To the best of our knowledge, this 'controlling' dimension has yet to be systematically researched in the context of ASD.

Second, behaviours used to attempt to avoid demands or exert control have been previously described as 'socially manipulative'. However, we used the term 'strategic' given that they were frequently relatively blunt attempts to distract from demands, take advantage of rules/norms (e.g. behaving in a babyish manner because younger children are not expected to comply with parental expectations), or coerce parents into meeting their

demands (e.g. by threatening problem behaviour). Although requiring social knowledge, this may not require the insight necessary for subtle manipulation.

In line with previous work, parents highlighted the need to conform with expectations as a major flashpoint for meltdowns. Other factors included sensory sensitivities, phobias, real or perceived provocation, and anxiety about the unknown. Extreme rigidity was evident in numerous parental descriptions. One potential explanation is that rigidity motivates the child to exert control to ensure that things happen according to their view of what should happen. Any deviation may be experienced as acutely aversive or anxiety-provoking, resulting in a meltdown. Intriguingly, however, extreme rigidity seems inconsistent with reports that novelty and variety can be helpful when making demands (Newson et al., 2003).

A potential moderating factor could be whether the activity is something the child finds problematic. For nonpreferred activities, or in situations where the child's tolerance for demands is reduced, a demand of any kind may be perceived as aversive. In these instances, strategies to mask demands, capture the child's interest indirectly or boost the child's sense of agency by giving them choice are reportedly helpful (Christie, Fidler, Duncan, & Healy, 2012; Larson, 2006; Lucyshyn et al., 2015). Clinical accounts of extreme/'pathological' demand avoidance highlight the need for a range of strategies, such that the child does not come to 'see through' a particular method. However, this approach has yet to be empirically contrasted with more strictly routinised alternatives.

This study raises important questions for future research. First, noncompliance appeared to affect all external demands for some children, but was more specific for others. Further studies could examine triggers or mitigating factors that make certain demands more tolerable. Large-scale investigations could also explore how these dimensions relate to age, gender, ability level, core ASD symptoms and comorbid features (e.g. anxiety, ADHD), and investigate how intensity varies dynamically with environmental stressors. Such work could also relate to ongoing investigation of intolerance of uncertainty in ASD, and its relationship with anxiety (Boulter, Freeston, South, & Rodgers, 2014; Hodgson, Freeston, Honey, & Rodgers, 2017). Understanding heterogeneity is likely to be essential in developing management approaches adapted for a specific child's level of insight and particular sensitivities.

Because this sample was selected for specific traits, these data do not speak to the validity of extreme/'pathological' demand avoidance as a cluster or entity within ASD. Population-representative samples are needed to examine whether associated dimensions cluster together. Noncompliance and mood variability, in particular irritability, are also evident in other profiles, for example, oppositional defiant disorder (Stringaris & Goodman, 2009). Investigating the manifestation of the traits described here across broader clinical samples is an important topic for future research.

Limitations

This study has a number of significant limitations. These include the small sample size, the fact that not all parents provided in-depth information on all aspects of the

interview, the lack of independent confirmation of clinical diagnoses of ASD, and the lack of a detailed diagnostic assessment of comorbidities. For a minority of interview items used to determine inclusion, interrater agreement for severity scores was low. However, parent-report questionnaire data also highlighted features associated with descriptions of extreme/'pathological' demand avoidance in this sample (Table 1). Future large-scale studies with systematic quantitative assessment of traits by multiple informants are needed.

Further considerations are that the specific behaviours reported by parents were affected by the questions included in the interview, which were chosen based on their apparent relevance to extreme/'pathological' demand avoidance (Table S1). This indirectly influenced the emergent thematic structure. There were undoubtedly numerous other important features that were not measured, in particular anxiety-relevant behaviour. The mode of completing the interviews differed across participants, which may have impacted the amount and nature of information provided.

We sampled only cooperative families who were willing to engage with research, who had some degree of knowledge of extreme/'pathological' demand avoidance. Their particular experiences could have impacted their construal of their child's difficulties. Further research in clinical contexts with a broader cross-section of parents, incorporating observational data, is needed to enhance the representativeness of work on this topic.

Clinical implications

These data highlight the importance of considering dimensions of difficulty that characterise children with ASD resembling extreme/'pathological' demand avoidance, given the substantial impact they have on daily life. Our findings indicate that noncompliance with everyday requests, attempts to control situations and others' activities, and extreme variability of mood are important treatment targets. Parental reports suggest that the apparent drivers of these difficulties include a negative emotional response to perceived pressure to conform with expectations, sensory sensitivities, phobias, real or perceived provocation, and anxiety about the unknown. Further empirical investigation is required to examine similarities and differences in the manifestation and drivers of these behaviours compared with other non-ASD profiles.

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Ethical information

Ethical approval was obtained from the King's College London Psychiatry, Nursing and Midwifery Research Ethics review board. The study was conducted in accordance with the declaration of Helsinki. Participants gave informed consent to participate.

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Supporting information

Additional Supporting Information may be found in the online version of this article:

Appendix S1. Supplemental Methods

Table S1. Parent interview questions.

Table S2. Scoring on the 11 indicators for features of extreme/'pathological' demand avoidance.

Table S3. Interrater reliability for DISCO items ($N = 29$).

References

- Agazzi, H., Tan, R., & Tan, S.Y. (2013). A case study of parent-child interaction therapy for the treatment of autism spectrum disorder. *Clinical Case Studies*, 12, 428–442.
- Armstrong, K., DeLoatche, K.J., Preece, K.K., & Agazzi, H. (2015). Combining parent-child interaction therapy and visual supports for the treatment of challenging behavior in a child with autism and intellectual disabilities and comorbid epilepsy. *Clinical Case Studies*, 14, 3–14.
- Armstrong, K., & Kimonis, E.R. (2013). Parent-child interaction therapy for the treatment of Asperger's disorder in early childhood: A case study. *Clinical Case Studies*, 12, 60–72.
- Arnold, L.E., Aman, M.G., Li, X., Butter, E., Humphries, K., Scahill, L., ... & Stigler, K.A. (2012). Research Units of Pediatric Psychopharmacology (RUPP) autism network randomized clinical trial of parent training and medication: One-year follow-up. *Journal of the American Academy of Child and Adolescent Psychiatry*, 51, 1173–1184.
- Barry, L.M., & Singer, G.H.S. (2001). A family in crisis: Replacing the aggressive behavior of a child with autism toward an infant sibling. *Journal of Positive Behavior Interventions*, 3, 28–38.
- Bearss, K., Johnson, C., Smith, T., Lecavalier, L., Swiezy, N., Aman, M., ... & Scahill, L. (2015). Effect of parent training vs parent education on behavioral problems in children with autism spectrum disorder: A randomized clinical trial. *JAMA Psychiatry*, 313, 1524–1533.
- Boulter, C., Freeston, M., South, M., & Rodgers, J. (2014). Intolerance of uncertainty as a framework for understanding anxiety in children and adolescents with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, 44, 1391–1402.
- Chowdhury, M., Aman, M.G., Scahill, L., Swiezy, N., Arnold, L.E., Lecavalier, L., ... & McDougle, C.J. (2010). The Home Situations Questionnaire-PDD version: Factor structure and psychometric properties. *Journal of Intellectual Disability Research*, 54, 281–291.
- Christie, P., Fidler, R., Duncan, M., & Healy, Z. (2012). *Understanding pathological demand avoidance syndrome in children: A guide for parents, teachers and other professionals*. London: Jessica Kingsley.
- DeGrace, B.W. (2004). The everyday occupation of families with children with autism. *The American Journal of Occupational Therapy: Official Publication of the American Occupational Therapy Association*, 58, 543–550.
- Gillberg, C., Gillberg, I.C., Thompson, L., Biskupsto, R., & Billstedt, E. (2015). Extreme ("pathological") demand avoidance in autism: A general population study in the Faroe Islands. *European Child and Adolescent Psychiatry*, 24, 979–984.
- Gray, D.E. (1993). Perceptions of stigma: The parents of autistic children. *Sociology of Health and Illness*, 15, 102–120.
- Groth-Marnat, G. (2009). *Handbook of psychological assessment* (5th edn). Hoboken, NJ: Wiley.
- Hodgetts, S., Nicholas, D., & Zwaigenbaum, L. (2013). Home sweet home? Families' experiences with aggression in children with autism spectrum disorders. *Focus on Autism and Other Developmental Disabilities*, 28, 166–174.

- Hodgson, A.R., Freeston, M.H., Honey, E., & Rodgers, J. (2017). Facing the unknown: Intolerance of uncertainty in children with autism spectrum disorder. *Journal of Applied Research in Intellectual Disabilities*, 30, 336–344.
- Larson, E. (2006). Caregiving and autism: How does children's propensity for routinization influence participation in family activities? *OTJR: Occupation, Participation and Health*, 26, 69–79.
- Lucyshyn, J.M., Fossett, B., Bakeman, R., Cheremshynski, C., Miller, L., Lohrmann, S., ... & Irvin, L.K. (2015). Transforming parent-child interaction in family routines: Longitudinal analysis with families of children with developmental disabilities. *Journal of Child and Family Studies*, 24, 3526–3541.
- Lucyshyn, J.M., Irvin, L.K., Blumberg, E.R., Laverty, R., Horner, R.H., & Sprague, J.R. (2004). Validating the construct of coercion in family routines: Expanding the unit of analysis in behavioral assessment with families of children with developmental disabilities. *Research and Practice for Persons with Severe Disabilities*, 29, 104–121.
- Moes, D.R., & Frea, W.D. (2000). Using family context to inform intervention planning for the treatment of a child with autism. *Journal of Positive Behavior Interventions*, 2, 40–46.
- Newson, E., Le Maréchal, K., & David, C. (2003). Pathological demand avoidance syndrome: A necessary distinction within the pervasive developmental disorders. *Archives of Diseases in Childhood*, 88, 595–600.
- O'Nions, E., Christie, P., Gould, J., Viding, E., & Happé, F. (2014). Development of the 'Extreme Demand Avoidance Questionnaire' (EDA-Q): Preliminary observations on a trait measure for pathological demand avoidance. *Journal of Child Psychology and Psychiatry*, 55, 758–768.
- O'Nions, E., Gould, J., Christie, P., Gillberg, C., Viding, E., & Happé, F. (2016). Identifying features of 'pathological demand avoidance' using the diagnostic interview for social and communication disorders ('DISCO'). *European Child and Adolescent Psychiatry*, 25, 407–419.
- O'Nions, E., Viding, E., Greven, C.U., Ronald, A., & Happé, F. (2014). Pathological Demand Avoidance (PDA): Exploring the behavioural profile. *Autism: The International Journal of Research and Practice*, 8, 538–544.
- Scahill, L., McDougle, C.J., Aman, M.G., Johnson, C., Handen, B., Bearss, K., ... & Vitiello, B. (2012). Effects of risperidone and parent training on adaptive functioning in children with pervasive developmental disorders and serious behavioral problems. *Journal of the American Academy of Child and Adolescent Psychiatry*, 51, 136–146.
- Stringaris, A., & Goodman, R. (2009). Three dimensions of oppositionality in youth. *Journal of Child Psychology and Psychiatry, and Allied Disciplines*, 50, 216–223.
- Thomas, D.R. (2006). A general inductive approach for analyzing qualitative evaluation data. *American Journal of Evaluation*, 27, 237–246.
- Wechsler, D. (1991). *The Wechsler intelligence scale for children* (3rd edn). San Antonio, TX: The Psychological Corporation.
- Wing, L., Leekam, S.R., Libby, S.J., Gould, J., & Locombe, M. (2002). The diagnostic interview for social and communication disorders: Background, inter-rater reliability and clinical use. *Journal of Child Psychology and Psychiatry*, 43, 307–325.

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