Exploring the Neurocognitive Correlates of Challenging Behaviours in Young People with Autism Spectrum Disorder

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Abstract

Many young people with autism spectrum disorder (ASD) display ‘challenging behaviours’, characterized by externalising behaviour and self-injurious behaviours (SIB). These behaviours can have a negative impact on a young person’s well-being, family environment and educational achievement. However, the development of effective interventions requires greater knowledge of ASD-specific models of challenging behaviours. ASD populations are found to demonstrate impairments in different cognitive domains, namely social domains, such as theory of mind (ToM) and emotion recognition (ER), but also non-social domains such as executive functioning (EF) and sensory or perceptual processing (PP). Parent-rated SIB and externalising behaviours, and neurocognitive performance were assessed in a population-derived sample of 100 adolescents with ASD. Structural equation modelling was used to estimate associations between cognitive domains (ToM, ER, EF, PP) and SIB and externalising behaviours. Poorer ToM was associated with increased SIB, whereas poorer PP was associated with increased externalising behaviours. These associations remained when controlling for language ability. This is the first analysis to examine how a wide range of neurocognitive domains relate to challenging behaviours, and suggests specific domains that may be important targets in the development of interventions in adolescents with ASD.

Keywords: autism spectrum disorder, cognition, externalising behaviours, self-injurious behaviour, challenging behaviours, SNAP

Introduction

A large body of research demonstrates that individuals with autism spectrum disorder (ASD) are at increased risk of experiencing co-occurring mental health problems (Gjevik et al., 2011; Leyfer et al., 2006; Simonoff et al., 2008). One of the more concerning issues in ASD is a set of behaviours subsumed under the term ‘challenging behaviours’. This umbrella term encompasses a wide range of phenomena including externalising behaviours (including severe non-compliance), and self-injurious behaviour (SIB) (Emerson, 2001). These behaviours have a negative impact upon educational achievement and community participation, and are associated with increased caregiver stress (Lecavalier et al., 2006), and increased risk of hospitalisation and admission to residential care (Emerson, 2001; Mandell, 2008). These behaviours may also increase the likelihood of later negative outcomes (e.g., delinquency, peer rejection), as is found in non-ASD populations (Card et al., 2008). Understanding ASD-specific risk factors for challenging behaviours will allow novel, targeted interventions to be developed, promoting improved quality of life and better long-term outcomes.

Although the term challenging behaviours encompasses a wide range of behaviours, this manuscript considers two types of challenging behaviours, which are often seen in individuals with ASD, separately. These are externalising behaviours, including conduct problems such as aggression and temper tantrums, along with severe non-compliance and refusal to meet demands (e.g. oppositionality), and SIB, which encapsulates a continuum of severity and topography directed at the self. The two domains have been found to have differential correlates, in that SIB, but not externalising behaviours, has been reported to be associated with having lower verbal ability and a specialist educational placement (Maskey et al., 2013), as well as having an IQ<70 (Carroll et al., 2014), supporting the importance of considering these two domains separately.

Both externalising behaviours and SIB are much more prevalent in individuals with ASD, as compared to typically developing individuals. Estimates for externalising behaviours in young people with ASD vary from 22-36% (Kaat and Lecavalier, 2013). Although externalising behaviours present in a somewhat different way in non-ASD populations (where along with core symptoms of oppositionality and aggressive behaviour, behaviours such as theft and deceitfulness are also common), population prevalence rates are estimated at 5-7% in young people (Costello et al., 2003; Meltzer et al., 2000). With regards to SIB, prior work finds a prevalence rate of 14-50% in children and adults with ASD (Baghdadli et al., 2003; Dominick et al., 2007; Maskey et al., 2013; Richards et al., 2012). This is contrast to prevalence rates of 7.3-11.5% in typically developing adolescents (Madge et al., 2008; Taliaferro et al., 2012). It should be noted that much research into SIB and ASD has used populations of individuals with concurrent intellectual disability (ID), and since individuals with ASD and ID are more likely to show SIB (Carroll et al., 2014), prevalence rates may be inflated. Additionally, the type of SIB found in young people without developmental disabilities is usually less stereotyped (e.g., cutting oneself) than that found in individuals with developmental disabilities (e.g., repetitive head banging). Whether these two apparently different forms of SIB are manifestations of the same underlying process remains unclear.

In both typically developing individuals, and in those with ID, having a diagnosis of ASD is associated with increased likelihood of challenging behaviours (Holden and Gitlesen, 2006; Matson and Rivet, 2008; McClintock et al., 2003). This suggests that ASD is a risk factor, over above having ID. There are multiple conceptual frameworks one can consider to understand challenging behaviours in individuals with ASD. One is the functional perspective, which originated from work with individuals with ID, but has since been applied to ASD. Here, challenging behaviours are seen as alternative communication strategies, resulting from comprised communicative ability (characteristic of individuals with ASD), which are then reinforced through interactions with their environment. The functional approach has been used to successfully decrease challenging behaviours in ID populations, however, the antecedents behind challenging behaviour in ASD may differ from that of ID populations (Reese et al., 2005), suggesting the development of more ASD-specific models of challenging behaviour is required. Additionally, the functional perspective cannot account for why the profile and prevalence of challenging behaviours varies across different genetic syndromes (e.g., increased self-injury in Cornelia de Lange and Prader-Willi, but not Angelman Syndrome) with comparable levels of ID (Oliver et al., 2013). This variation suggests that there are other factors, beyond impaired communication and inadvertent environmental reinforcement, to consider. Thus, one alternative approach is to focus upon the neurocognitive profile associated with ASD, which is thought to underpin the core symptoms of social communication difficulties and restricted, repetitive behaviours, and consider how these impairments may also be important in understanding the development of challenging behaviours. The current manuscript takes this approach, although acknowledges there are other, complementary perspectives available.

Neurocognitive correlates of challenging behaviours in ASD populations

Recent calls for a focus upon mapping pathways between cognition and behaviour (rather than associations between cognition and diagnostic categories) suggest this method may better contribute to our understanding of psychopathology (Insel et al., 2010). Research exploring the neurocognitive correlates of challenging behaviours in ASD is sparse. One of the most well documented aspects of the neurocognitive profile associated with ASD is impairment in theory of mind (ToM) ability (Frith, 2001), characterised by difficulties understanding the mental states (e.g., beliefs) of others. Within a nationwide twin study, the strongest predictor of child conduct problems was ASD symptoms, specifically in the domain of social interaction problems (Kerekes et al., 2014), and performance on computerised ToM tasks has been found to predict self-reported aggression in children with ASD (Pouw et al., 2013). Individuals with ASD and co-occurring aggressive behaviour also demonstrate greater parent-reported social and communication problems (Mazurek et al., 2013; Kanne and Mazurek, 2011). With regards to SIB, the literature is more limited. Studies find SIB is associated with impairment in parent-rated social communication (Duerden et al., 2012), and more severe impairment in parent-rated socialization in individuals with ASD and ID (Baghdadli et al., 2003).

Along with difficulties in ToM, impaired emotion recognition (ER) has also been posited as part of the neurocognitive profile found in individuals with ASD (Uljarevic and Hamilton, 2012, but see Jones et al., 2011a for opposing findings). Research finds robust associations between impairments in fear recognition and externalising behaviour in non-ASD populations (Marsh and Blair, 2008). To our knowledge only two studies have examined the link between ER and co-occurring behaviour problems in ASD, using the same sample, to find that difficulty identifying surprise is associated with the presence of additional severe mood problems (Simonoff et al., 2012) and that difficulty identifying fear is associated with co-occurring callous-unemotional traits (Carter Leno et al., 2015).

Executive functioning (EF) impairments are also reported in individuals with ASD across a variety of domains (Hill, 2004; Brunsdon et al., 2015). EF impairments are found in the domains of cognitive flexibility and planning (Ozonoff et al., 2004; Landry and Al-Taie, 2016), response selection/monitoring (Happé et al., 2006) and inhibition (Geurts et al., 2014). In non-ASD populations, associations are reported between impairments in both inhibition and rigidity, and externalising behaviour (Hobson et al., 2011; Toupin et al., 2000). Correspondingly, aggressive behaviour in children with ASD is associated with parent-reported inattention and hyperactivity (Hill et al., 2014) and inflexibility (Lawson et al., 2015; Visser et al., 2014). Similarly, SIB is also associated with significantly higher levels of parent-rated impulsivity in samples of individuals with ASD and ID (Richards et al., 2012).

The final domain of neurocognitive functioning to consider is atypical sensory, or perceptual processing (PP). Many individuals with ASD experience sensory and perceptual abnormalities across a range of modalities, regardless of age and cognitive ability, experiencing both hypo- and hyper-sensitivity to sensory input (Leekam et al., 2007), and process incoming sensory and perceptual information in a different way to typically developing individuals (Gomot et al., 2006). Research finds auditory hyper-sensitivity is associated with externalising behaviours (Lundqvist, 2013), and atypical sensory processing is the strongest single predictor of SIB in large samples of children with ASD (Duerden et al., 2012). Within a sample of individuals with fragile X syndrome, the presence of SIB was higher in individuals with a diagnosis of ASD, and also in those with PP difficulties (Symons et al., 2010).

Current Aims

Prior literature suggests that specific elements of the neurocognitive profile associated with ASD are related to co-occurring challenging behaviours. However, many prior studies rely on parent report to assess both neurocognitive difficulties and challenging behaviours, and have utilized populations with a large proportion of individuals with severe ID. Furthermore, many previous studies have tested the role of a singular neurocognitive domain, whereas in the current paper we take a more systematic, data driven approach to exploring associations between four neurocognitive domains and behavioural outcomes. The current paper tests how performance in tasks tapping specific neurocognitive domains (ToM, ER, EF, PP) relates to two domains of challenging behaviours (externalising behaviours and SIB) within a population-based sample of adolescents with ASD.

Methods

Sample

A total of 100 adolescents with ASD, who had an IQ≥50, were assessed on the relevant measures as part of the Special Needs and Autism Project (SNAP) cohort (Baird et al., 2006). Of the participants, 54 met consensus criteria for childhood autism and 46 for other pervasive developmental disorders (ICD-10). There were 91 males and 9 females, the mean age was 15.48 years (SD = 0.46; range 14.7–16.8), and the mean full scale IQ was 84.31 (SD = 18.03; range 50–119). This cohort, initially assessed as part of an autism prevalence study, was drawn from 56 946 children living in the South Thames area of the UK and born between July 1990 and December 1991. The cohort was assessed at mean ages of 12 and 16 years. Assessment at 16 years focused on the cognitive phenotype of ASD and only those who had estimated IQ≥50 at 12 years were included (Charman et al., 2011). All received a consensus clinical ICD-10 ASD diagnosis, made using the Autism Diagnostic Interview-Revised (ADI-R; Lord et al., 1994) and Autism Diagnostic Observation Schedule – Generic (ADOS-G; Lord et al., 2000) at age 12 years. Written informed consent was obtained from all parents and at age 16 years by the participant if their level of understanding was sufficient. The study was approved by the South East Multicentre Research Ethics Committee (REC) (05/MRE01/67).

Questionnaires

The majority of questionnaires and assessments were administered to parents when participants were aged 16 years.

The Profile of Neuropsychiatric Symptoms (PONS; Santosh et al., 2015) is a 62-item questionnaire that assesses the severity and impact of 31 symptoms commonly reported in children and young people with neurodevelopmental disorders. For each symptom, a brief definition is given, and the respondent is asked to report the overall frequency of that symptom (0–5) and its impact on everyday life (0–5). The two ratings are combined and averaged to provide an overall score for each symptom (0-5). Current analyses include items related to: oppositionality, aggression, explosive rage, antisocial behaviour, labile mood and self-injury.

The Repetitive Behavior Scale-Revised (RBS-R; Bodfish et al., 2000) is a 43-item questionnaire that assesses repetitive behaviours, and consists of six subscales (stereotyped behaviour, SIB, compulsive behaviour, routine behaviour, sameness behaviour and restricted behaviour). Respondents rate each behaviour from not occurring, to occurring and being a severe problem (0–3). Current analyses focused on items within the SIB subscale: hits body, hits self on surface, hits self with object, bites self, pulls at skin, scratches self, inserts items into body and picks skin.

Assessments

Receptive Language Ability

The Test for Reception of Grammar – Electronic Version (TROG-E; Bishop, 2005) was used to estimate standard scores for receptive grammar. The TROG-E requires participants to select pictures that correspond to sentences of increasing grammatical complexity. The TROG-E provides norms for individuals aged four years to adult.

Neurocognitive Measures

Full details of the neurocognitive tasks are given in the Supplementary Materials.

ToM

ToM ability was assessed using four computer based tasks: the Strange Stories task (Happé, 1994), the Frith–Happé animations (Abell et al., 2000), a combined False Belief task based on previous tasks measuring false belief understanding (Sullivan et al., 1994; Hughes et al., 2000), the Reading the Mind in the Eyes task (Baron-Cohen et al., 2001), and the Penny Hiding task (Baron-Cohen, 1992).

ER

The verbal vocal expressions of emotion task (Sauter, 2006; Sauter et al., 2010), played recordings of actors expressing each of the emotions verbally whilst reading out neutral content (three-digit numbers). The total number of correct responses for each of the six emotions (happy, sad, fear, surprise, anger, disgust) served as a measure of ER ability. Data from this task have previously been reported in the SNAP cohort (Jones et al., 2011a).

EF

EF was assessed using four tasks: the Card Sort task indexing cognitive flexibility and response reversal (Tregay et al., 2009), the Trail Making task indexing attentional switching and response reversal (Reitan and Wolfson, 1985), the Opposite Worlds and Score! tasks from the Test of Everyday Attention for Children (Manly et al., 2001) indexing interference inhibition and sustained attention respectively. Data from the majority of the EF tasks, along with ToM tasks, have previously been reported in the SNAP cohort (Carter Leno et al., 2015; Hollocks et al., 2014).

PP

Auditory Processing

Auditory processing was assessed using the “Dinosaur” software programme created by Dorothy Bishop (Oxford University). Participants were shown two cartoon dinosaurs and had to decide which dinosaur made a 1) louder (intensity discrimination) or 2) longer (duration discrimination) sound, respectively.

Visual Processing

The participant had to indicate from two panels which contained the target motion/stimulus. Participants had to decide which panel contained dots that 1) moved in the same way (detection of coherent motion), 2) contained a shape (detection of a form from motion) or 3) contained a man walking (detection of biological motion).

In both the auditory and visual perception tasks, a detection threshold was established using an adaptive staircase procedure, where the task was made easier/harder depending on ongoing performance. Across the tasks, a higher threshold indicated a greater amount of information required to detect the target stimuli. Data from these tasks have previously been reported in the SNAP cohort (Jones et al., 2011b; Jones et al., 2009).

Statistical Analyses

All variables were assessed for normality, and where necessary transformed using Box-Cox transformation (see Table 1). Eight neurocognitive variables were treated as ordinal variables due to extreme skew (Score!, Penny Hiding task, all ER variables) and all SIB items were treated as binary (present/absent) due to low incidence of individual SIBs. For all neurocognitive variables, a higher score was indicative of worse performance.

Table 1. Mean Raw Scores on Neurocognitive Measures

Latent Variable	Task (n of observations)	Mean (SD; range)
ToM	Strange Stories (n=88)*	0.85 (0.53; 0-2)
	Frith–Happé animations (n=87)*	2.87 (0.94; 0-4.75)
	Combined False Belief Task (n=99)*	4.75 (2.42; 0-8)
	Reading the Mind in the Eyes (n=94)*	17.02 (4.44; 6-25)
	Penny Hiding (n=100)^ ordinal categories are as follows 0/1=1, 2/3=2, 4/5=3, ≥6=4	2.32 (2.75; 0-14)
ER	Happiness (n=96)*	3.56 (1.42; 0-5)
	Sadness (n=96)*	4.23 (1.17; 0-5)
	Fear (n=96)*	2.73 (1.69; 0-5)
	Surprise (n=96)*	3.96 (1.23; 0-5)
	Anger (n=96)*	3.38 (1.72; 0-5)
	Disgust (n=96)*	2.46 (1.55; 0-5)
EF	Card Sort (n=98) +	7.24 (6.62; 1-36)
	Trail Making (n=88) +	63.39 (44.00; 13.37-257.09)
	Opposite Worlds (n=98) +	8.37 (7.49; -3.71-47.42)
	Score!(n=96)* ^ ordinal categories are as follows 0/5=3, 6/9=2, 10=1	7.68 (2.51; 0-10)
PP	Auditory Intensity Threshold (n=92) +	9.40 (6.56; 1-27.75)
	Auditory Duration Threshold (n=93) +	7.67 (6.70; 1-28.75)
	Visual Form Threshold (n=91) +	0.29 (0.17; 0.07-0.88)
	Visual Motion Threshold (n=89) +	0.19 (0.14; 0.30-.74)
	Visual Biological Motion Threshold (n=90) +	0.39 (0.14; 0.14-.83)

EF indicates executive functioning; ER emotion recognition; PP perceptual processing; ToM theory of mind

*indicates reverse score used in analysis; + transformed using Box-Cox; ^ transformed to ordinal data

Structural Equation Modelling (SEM) Analysis

Following the generation of outcome variables (see below for details), SEM was used to estimate the association between performance on the four neurocognitive latent variables (ToM, ER, EF, and PP) and the scores on observed variables (SIB and externalising behaviours). Latent variable models for mixed data SEM were conducted in Mplus 7 (Muthén and Muthén, 2012). Given many of our variables were categorical the weighted least squares mean and variance adjusted (WLSMV) estimator was used. Model fit was examined using the relative χ^2, the root mean square error of approximation (RMSEA), the comparative fit index (CFI), and the Tucker-Lewis fit index (TLI). A satisfactorily fitting model should have RMSEA≤0.05, CFI and TLI >0.90 (Bentler, 1990; Tucker and Lewis, 1973).

Creation of Outcome and Predictor Variables

Outcome variables of 'externalising behaviours' and 'SIB' were generated from parent-reported PONS and RBS items. From these measures relevant items were chosen that indexed either domain of behaviour. These were entered into an exploratory factor analysis (EFA) for mixed data, using maximum likelihood and promax rotation. The factor analysis was constrained to two factors. Both factors had eigenvalues greater than 1 (externalising behaviours factor = 4.08, SIB factor = 1.89). All factor loadings were greater than 0.3, and all items loaded on the predicted factor (see Table 2) except the 'picks skin' item from the RBS-R. This item was therefore excluded from the outcome variable formation.

A confirmatory factor analysis (CFA) indicated a two-factor solution in which latent variables were correlated (r=0.48), had good fit (relative χ^2=1.09, RMSEA=0.03, CFA=0.98, TLI=0.97), and was better suited than a one-factor solution (relative χ^2=1.89, RMSEA=0.10, CFA=0.74, TLI=0.69).

Outcome variables were the sum of all items for each factor respectively. This approach was preferred to the EFA factor extracted scores to allow our results to be directly comparable with future samples. Observed sum-scores were used in the SEM model as measurements of the latent variables, as opposed to a full item to latent variable structure, to reduce the number of parameters the model had to estimate, given the modest sample size. The externalising behaviours variable was transformed to a normal distribution using Box-Cox transformation, and the SIB variable was treated as ordinal (scores ranged from 0-8).

For all four neurocognitive latent variables (ToM, ER, EF, PP), EFA was also undertaken not to identify a new structure, for which a large sample would be required to be convincing, but to ensure that our data were not inconsistent with received wisdom, before assuming that structure held for the CFA. All individual neurocognitive tasks loaded significantly onto the proposed latent variable. See Supplementary Materials for details of all neurocognitive latent variables.

Estimation of associations between neurocognitive latent variables and outcome variables

Step 1. Missing data were imputed in Mplus, and results of SEM analyses were aggregated across 20 imputed data sets. See Tables 1 and 2 for number of observations for all neurocognitive tasks and questionnaire items respectively. All latent neurocognitive variables, SIB and externalising behaviours, were placed into a correlational model.

Over a sequence of models the largest significant correlational pathway between the latent neurocognitive variables and the observed behavioural variables was set to a directional path, which in turn led to existing weaker but significant neurocognition-behaviour associations becoming non-significant and thus being removed from the model (Chou and Huh, 2014). Correlations among latent neurocognitive variables and between externalising behaviour and

SIB were retained in all models. To control for underlying ability that could impact on cognitive performance, the effect of controlling for language on the final model was then examined.

Step 2. Exploratory *post-hoc* mediation analyses were run using the sem and estat effects commands in Stata 14 to explore the high correlation between latent neurocognitive variables in the final model. These *post-hoc* analyses were undertaken as mediation could explain the strong correlations between latent variables, since performance in one neurocognitive domain could mediate performance in another domain. A mediation model proposes that one independent variable (here one neurocognitive variable) has an indirect effect on a dependent variable, by influencing another independent variable (the mediator variable, here a different neurocognitive variable), which in turn influences the dependent variable (here our observed outcomes of externalising behaviours and SIB). To test whether the indirect effect of latent variables was significant, factor scores for neurocognitive variables in the final model were extracted using Mplus, and the coefficients of the indirect pathways were tested for significance.

The aim of these analyses was to identify which neurocognitive domains were associated with different symptoms of challenging behaviours. The data were modelled with paths in the direction from neurocognitive to symptom domains. Because the data are cross-sectional, results are unable to discriminate direction of effect, including reciprocal effects, between neurocognitive and symptom factors, and the direction of these paths should not be used to infer a causal association.

Results

For sample raw scores on neurocognitive tasks that made up the latent variables see Table 1. For sample raw scores from the PONS and RBS-R that made up the outcome variables of externalising behaviours and SIB, see Table 2.

Table 2. Sample Raw Scores and Rotated Factor Loadings of Items from the Profile of Neuropsychiatric Symptoms (PONS) and Repetitive Behaviour Scale-Revised (RBS-R) onto Factors of Aggression/Non-Compliance and Self-Injurious Behaviour

Item (n completed)	**Mean Score (SD; Range)**	**Loading on Factor 1. Aggressive/Non-Compliant Behaviour**	**Loading on Factor 2. Self-Injurious Behaviour**
PONS Oppositionality (n=94)	1.86 (1.40; 0-5)	**0.77**	-0.22
PONS Aggression (n=92)	1.33 (1.33; 0-5)	**0.90**	0.01
PONS Explosive Rage (n=94)	1.10 (1.19; 0-5)	**0.88**	0.01
PONS Antisocial Behaviour (n=94)	0.22 (0.64; 0-5)	**0.42**	-0.22
PONS Labile Mood (n=94)	0.91 (1.29; 0-5)	**0.61**	0.23
PONS Self Injury (n=94)	0.56 (1.12; 0-5)	0.37	**0.40**
RBS Hits Body (n=91)	0.41 (0.71; 0-3)	0.05	**0.73**
RBS Hits Self on Surface (n=89)	0.16 (0.50; 0-3)	0.02	**0.75**
RBS Hits Self with Object (n=91)	0.15 (0.47; 0-3)	-0.14	**0.85**
RBS Bites Self (n=90)	0.11 (0.38; 0-3)	0.04	**0.47**
RBS Pulls at Skin (n=91)	0.14 (0.44; 0-3)	0.07	**0.47**
RBS Scratches Self (n=91)	0.18 (0.44; 0-3)	0.16	**0.41**
RBS Inserts Items into Body (n=92)	0.09 (0.41; 0-3)	0.02	**0.46**

PONS indicates Profile of Neuropsychiatric Symptoms; RBS-R Repetitive Behavior Scale-Revised; SIB self-injurious behaviour.

Note: These data represent raw scores. All RBS items and the PONS self-injury item were treated as binary (present/absent) in analyses due to low incidence of SIB.

Step 1. Correlations among latent neurocognitive variables were very strong (see Figure 1). The correlation between SIB and externalising behaviours was moderate (*r*=0.37). The strongest correlation between latent neurocognitive variables and behavioural outcomes was between ToM and SIB (*r*=0.39, *p*<0.01; Figure 1), whereas the correlation between ToM and externalising behaviours was the smallest and non-significant (*r*=0.18, *p*=0.11). The model was re-run, specifying the pathway from ToM to SIB as a predictive pathway, and removing the pathway from ToM to externalising behaviours, and allowing all remaining latent neurocognitive variables to correlate with behavioural outcomes. This model had acceptable fit (relative χ^2=1.23, RMSEA=0.05, CFI=0.90, TLI=0.88). In this model, the next strongest correlation was between PP and externalising behaviours (*r*=0.32, *p*<0.01), whereas the correlation between PP and SIB was non-significant (*r*=-0.05, *p*=.64). Both the correlation between ER and SIB, and the correlation between EF and SIB, were non-significant (*r*=-0.02, *p*=0.79; *r*=0.05, *p*=0.67). The model was re-run, specifying in addition to the pathway from ToM to SIB, the pathway from PP to externalising behaviour as a predictive pathway, and removing the pathway from PP to SIB. The only correlations now estimated were between ER and externalising behaviours, and between EF and externalising behaviours. This model showed acceptable fit (relative χ^2=1.22, RMSEA=0.045 CFI=0.91, TLI=0.89). Both the correlation between ER and externalising behaviours (*r*=0.08), and the correlation between EF and externalising behaviours (*r*=0.14), were non-significant, therefore the latent variables of ER and EF were removed, giving the final model.

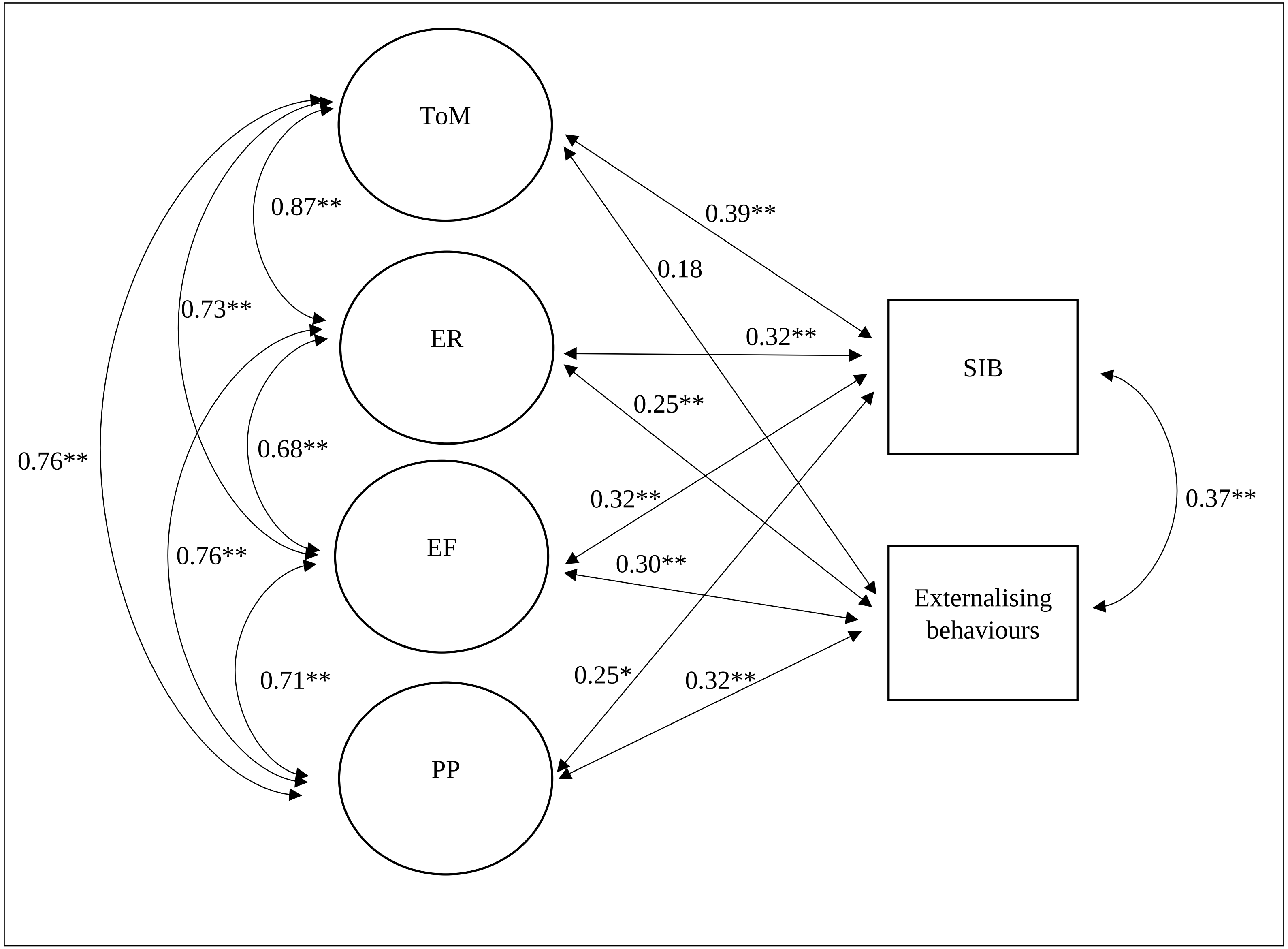

Figure 1. Initial Correlational Model of Associations between Neurocognitive Domains and Challenging Behaviours

EF indicates executive functioning; ER emotion recognition; PP perceptual processing; SIB self-injurious behaviour; ToM theory of mind.
*p<0.05, **p<0.01.

The final model (see Figure 2) continued to demonstrate acceptable model fit (relative χ^2=1.35, RMSEA=0.06, CFI=0.92, TLI=0.90), and indicated a significant association between ToM and SIB (β=0.37, p<0.01) and between PP and externalising behaviours (β=0.29, p<0.01). Significant correlations were found between SIB and externalising behaviours (r=0.33, p<0.01), and between ToM and PP (r=0.74, p<0.01).

Next, a model with directional paths from language ability to both neurocognitive domains and behavioural outcomes was investigated as an additional step, to explore effect of controlling for language on associations between neurocognitive domains and behaviour (Figure 3). The associations between neurocognitive domains and behaviour remained significant, along with the correlations between ToM and PP, and SIB and externalising behaviours (all ps<0.05). This model had poorer fit (relative χ^2=1.64, RMSEA=0.08, CFI=0.87, TLI=0.83).

Since the distribution of the SIB variable was highly skewed, the final model from Step 1 was re-created, treating SIB as a binary variable, and a comparable model was found. The details of this are given in the Supplementary Materials.

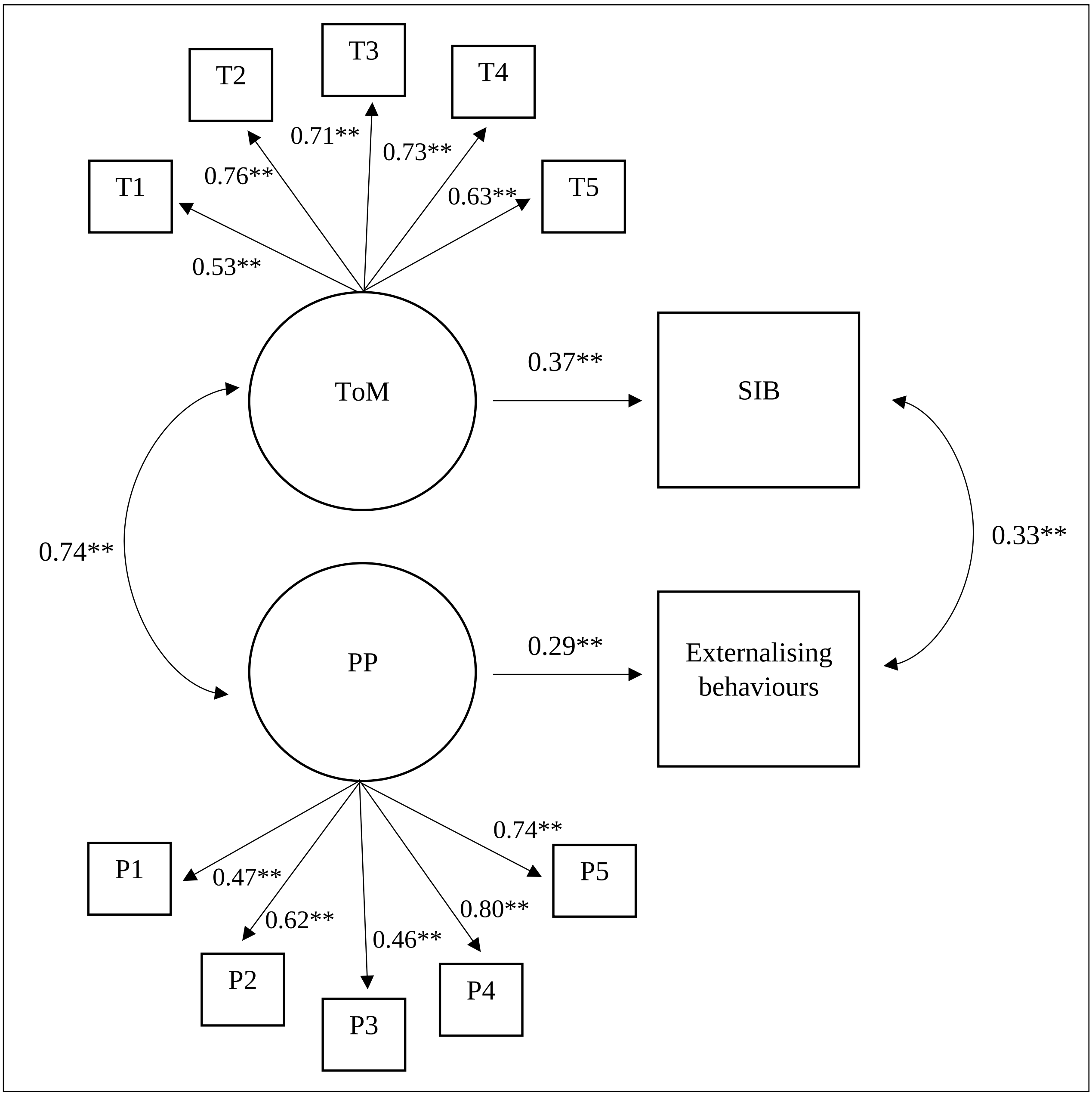

Figure 2. Final Model Depicting Relationship between Neurocognitive Domains and Aspects of Challenging Behaviours.

PP indicates perceptual processing; SIB self-injurious behaviour; ToM theory of mind.

**p*<0.05, ***p*<0.01.

T1: Strange Stories task, T2: Frith–Happé animations task, T3: combined False Belief task, T4: Reading the Mind in the Eyes task, T5: Penny Hiding task, P1: Audio intensity discrimination, P2: Audio duration discrimination, P3: Visual form discrimination, P4: Visual motion discrimination, P5: Visual biological motion discrimination.

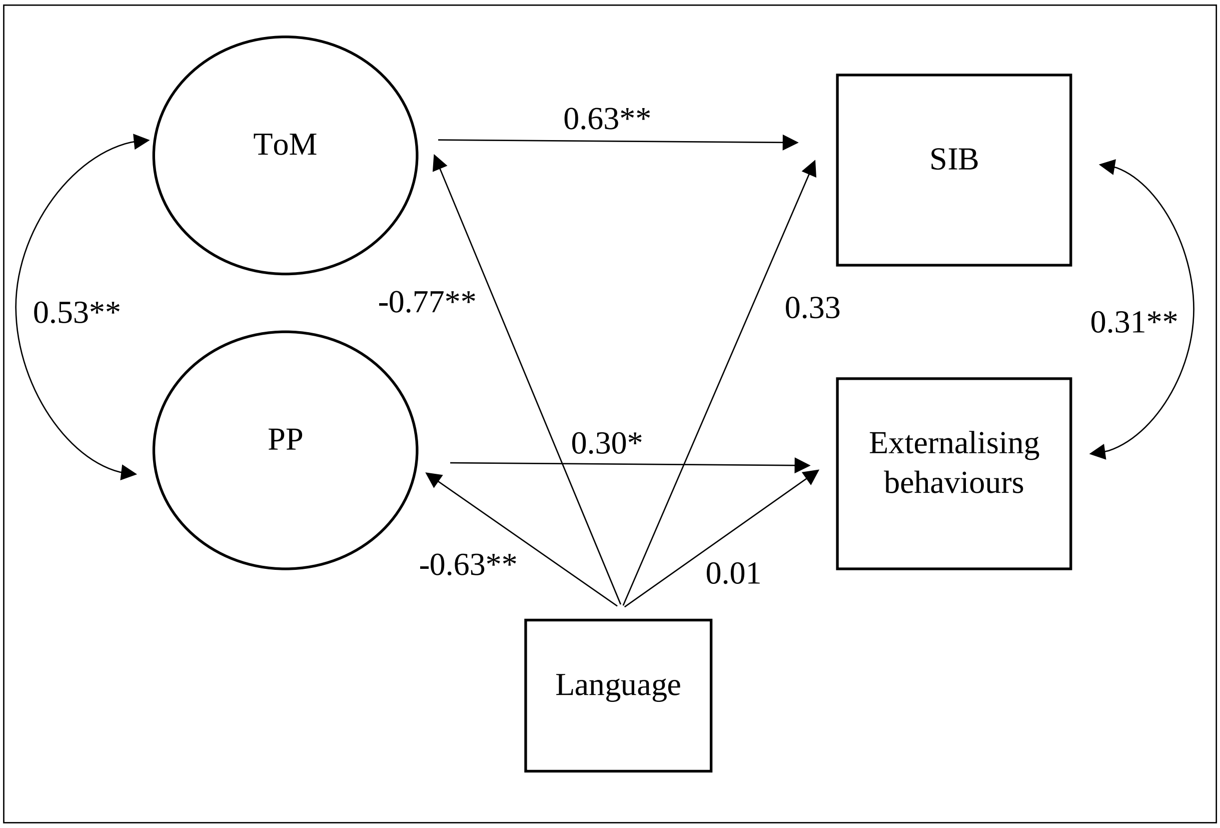

Figure 3. Model Depicting Associations between Neurocognitive Domains and Aspects of Challenging Behaviours Whilst Adjusting for Language

PP indicates perceptual processing; SIB self-injurious behaviour; ToM theory of mind.

**p*<0.05, ***p*<0.01

Step 2. Given the high correlation between the ToM and PP latent variables, exploratory *post-hoc* mediation analyses were conducted. Model 1 tested PP as a mediator of the association between ToM and SIB (ToM→PP→SIB). Model 2 tested ToM as a mediator of the association between PP and externalising behaviours (PP→ToM→ externalising behaviours). In both models the indirect pathway coefficient was non-significant (β=-0.14, p=0.68 and β=0.02, p=0.31 for Model 1 and 2, respectively), indicating that mediation was an unlikely explanation of the observed associations.

Discussion

The current paper tested whether ability in specific neurocognitive domains was associated with externalising behaviours and SIB in a population-based sample of adolescents with ASD. Data-driven SEM, which allows for simultaneous estimation of the association between different domains of cognition and behaviour, indicated poorer PP was associated with increased externalising behaviours, whereas poorer ToM was associated with increased SIB. These associations between cognition and behaviour remained when language ability was controlled for. Non-significant mediation analyses suggested that, despite the high correlation between neurocognitive domains, there was some specificity within the reported associations between neurocognitive domains and aspects of challenging behaviours.

Associations between Neurocognitive Domains and Challenging Behaviours

Sample size requirements for SEM analyses are complex but an obvious concern for analysis of clinical cohort studies of a limited and fixed size. We therefore conducted post-hoc power calculations. Although the calculations for the two paths of primary interest in the final model were satisfactory (94% for the ToM-SIB coefficient and 77% for the PP-externalizing behaviours coefficient at two-tailed 95% significance), nonetheless caution should be taken in interpreting the current results due to a moderate sample size, and strong correlations between neurocognitive domains. However, results suggest there is some specificity in the associations found, as post-hoc mediation analyses found no indirect effect of PP upon SIB through mediation on ToM, or vice versa for ToM upon the association between externalising behaviours and PP. Additionally, within initial correlational analyses, the association between ToM and externalising behaviours was not significant. This is in contrast to prior research that reports an association between parent-reported social functioning and parent-reported aggressive behaviour (Kanne and Mazurek, 2011; Mazurek et al., 2013; Kerekes et al., 2014;

Pouw et al., 2013). However, the majority of these studies, with the exception of Pouw and colleagues, did not specifically measure ToM, instead measuring social functioning or communication, and relied on parent report. Therefore, it may be that some aspects of social functioning (e.g., communication) are related to externalising behaviours in ASD, whereas others, such as ToM, are not. Additionally, respondent differences could be contributing to conflicting results. A further point to consider is that previous studies have only measured aggressive behaviour, and did not specifically test the association between ToM and SIB. However, it should be held in mind that in the current study, reduced power in the context of highly correlated factors could lead to difficulties detecting pathways between cognition and behaviour.

The literature on neurocognitive correlates of SIB in ASD populations is limited and thus current analyses are the first to comprehensively test how ability in specific neurocognitive domains relates to SIB. Prior studies have found more general associations between parent-reported increased SIB and greater social difficulties and communication skills (Duerden et al., 2012; Baghdadli et al., 2003); our finding of poorer ToM performance being associated with increased SIB builds upon these and clarifies that challenging behaviours may not be solely due to difficulties in communication. Recent work with this sample, using the same ToM tasks, found ToM task performance was associated with parent-reported social skills (Jones et al., 2018), suggesting previously reported associations between SIB and social difficulties (e.g., Duerden et al., 2012; Baghdadli et al., 2003) may in part have been driven by impaired ToM.

Two interpretations of results are considered – that SIB may be a ‘distress signal’ in part due to negative emotions caused by lack of social understanding and difficulty communicating. An alternative interpretation is that reduced understanding of other’s thoughts and feelings may mean atypical behaviour is not moderated by social signals to the same degree, and thus

SIB is not inhibited. It also should be noted that ToM is a multi-faced construct, and effective ToM may rely on many abilities (e.g., language skills, abstract/conceptual thinking, and distinguishing self vs. other). Future research should also attempt to disentangle what aspects of ToM might be driving the association with SIB, as this will have direct implications for intervention design.

The finding of poorer PP being associated with increased externalising behaviours is in line with prior research reporting associations between sensory processing and aggressive behaviour in young children with ASD (Hartley et al., 2008), and one study which specifically separated challenging behaviours in individuals with ID into SIB, stereotyped behaviour and aggressive behaviour, and found auditory hypersensitivity was predictive of aggressive behaviour, but not SIB (Lundqvist, 2013). In contrast to prior literature (Duerden et al., 2012; Symons et al., 2010), and although initial correlational analyses indicated poorer PP was significantly related to increased SIB, this association did not remain once the relationship between ToM and SIB was taken into account. A question for future research is whether performance in the kinds of PP tasks used in the current analyses translate to 'real-life' sensory sensitivities. Previous work with this sample found that performance on the auditory processing tasks used in current analyses was associated with self-reported auditory sensory behaviours (e.g., coping with loudness levels) (Jones et al., 2009), however more work is required in this area.

If this hypothesis was supported, it suggests a comprehensive sensory assessment may be informative if an individual with ASD presents with externalizing behaviours. This could be used to tailor interventions to include a focus on identifying sensory-related triggers, or exploring how difficulties processing incoming perceptual information may be linked to behaviour problems. This is in line with current clinical guidelines, which recommend taking into account individual sensory sensitivities when planning support and management of young people with ASD, but also that existing interventions for mental health difficulties, which have been developed in non-ASD populations, may need to be tailored to suit ASD populations (National Institute for Clinical Excellence, August 2013).

Overlap Between Neurocognitive Domains

Current analyses found a strong overlap between the neurocognitive domains of ToM, ER, EF and PP. Although some of these were to be expected (e.g., the overlap between ToM and ER), the association between others is less clear. Prior work using the current sample also found strong correlations between different tasks, which were not found in a non-ASD comparison group (Jones et al., 2011b). Earlier work also reports strong correlations between similar cognitive domains in individuals with ASD, but not in typically developing controls (Ozonoff et al., 2004). Widespread impairments in multiple areas of cognition could be characteristic of ASD (Brunsdon et al., 2015), and perhaps in part help to understand the widespread co-occurring psychopathology reported in young people with ASD (Simonoff et al., 2008). Alternatively, the overlap could be due, in part, to other unmeasured factors which could influence performance across all tasks, such as inattention, motivation or general task understanding. Inattention is likely to be prevalent in individuals with ASD, as studies have found around 30% of this sample also met diagnostic criteria for ADHD (Simonoff et al., 2008), and elsewhere up to 55% of young people with ASD have been found to have sub-threshold ADHD traits (Leyfer et al., 2006).

The strengths of the current work include the wide range of cognitive tasks, tapping different domains, and a population-based sample of well-characterised individuals with ASD, who have a wide range of IQ (50-119). Most studies exploring the neurocognitive profile associated with ASD only include individuals with IQ≥70, and therefore only represent a sub-group of individuals with ASD. A further strength of the current study is the use of SEM, which allows simultaneous estimation of the association between different domains of cognition and two aspects of challenging behaviours, whilst also controlling for the effect of language ability on these associations.

In terms of limitations, strong correlations between neurocognitive domains and a moderate sample size mean associations between cognition and behaviour should be interpreted with caution until replicated. Although final model found poorer ToM and PP ability were significant predictors of SIB and externalising behaviours respectively, EF and ER were still significantly correlated with externalising behaviours and SIB in initial analyses, but were not included in the final model based on the method of model selection. The method of selection based on entering first neurocognitive domains with the strongest association as predictors of behavioural outcomes may lead to inflated specificity in the resulting neurocognition – behaviour associations. It may be the case that if all domains were tested in a full model, using a larger sample, then analyses would have greater power to detect associations between EF and ER and domains of challenging behaviours. Additionally, the cross-sectional nature of the sample also means we cannot draw any conclusions regarding the causality of association between poorer neurocognitive ability and increased challenging behaviours. This is something that should be explored with longitudinal samples, and also with treatment studies specifically targeting cognitive domains.

Findings suggest it may be important to consider PP atypicalities when testing hypotheses regarding potential drivers of challenging behaviours in individuals with ASD, but go one step further to suggest there may be specificity in associations between domains of cognitive functioning and types of challenging behaviours. Although the umbrella term of challenging behaviours is a useful clinical label, results suggest that different types of challenging behaviours are associated with different types of cognitive impairments, and so should be considered separately. Second, although much of the literature in the field aims to draw specific associations between different cognitive domains and behavioural characteristics, our results suggest these cognitive domains are so strongly correlated that the specificity of associations may be over-exaggerated unless studies attempt to use ‘purer’ measures of cognition, and account more widely for overlapping domains. If evidence for a causal association between neurocognitive functioning and co-occurring behaviour problems were found, this would have implications for intervention design, and potentially allow for the identification of individuals at high-risk for developing challenging behaviours.

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