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Exploring the cognitive features in children with autism spectrum disorder, their co-twins, and typically developing children within a population-based sample

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Background: The behavioural symptoms of autism spectrum disorder (ASD) are thought to reflect underlying cognitive deficits/differences. The findings in the literature are somewhat mixed regarding the cognitive features of ASD. This study attempted to address this issue by investigating a range of cognitive deficits and the prevalence of multiple cognitive atypicalities in a large population-based sample comprising children with ASD, their unaffected co-twins, and typically developing comparison children. **Methods:** Participants included families from the Twins Early Development Study (TEDS) where one or both children met diagnostic criteria for ASD. Overall, 181 adolescents with a diagnosis of ASD and 73 unaffected co-twins were included, plus an additional 160 comparison control participants. An extensive cognitive battery was administered to measure IQ, central coherence, executive function, and theory of mind ability. **Results:** Differences between groups (ASD, co-twin, control) are reported on tasks assessing theory of mind, executive function, and central coherence. The ASD group performed atypically in significantly more cognitive tasks than the unaffected co-twin and control groups. Nearly a third of the ASD group presented with multiple cognitive atypicalities. **Conclusions:** Multiple cognitive atypicalities appear to be a characteristic, but not universal feature, of ASD. Further work is needed to investigate whether specific cognitive atypicalities, either alone or together, are related to specific behaviours characteristic of ASD. **Keywords:** Autism spectrum disorder, cognition, theory of mind, executive function, weak central coherence.

Introduction

Autism spectrum disorder (ASD) is a developmental disorder characterised by impaired social interaction and communication, and restricted and repetitive patterns of behaviour and interests (RRBIs) (American Psychiatric Association, 2013). These behavioural symptoms are thought to reflect underlying cognitive deficits/differences, which have been extensively researched (see Brunsdon & Happé, for review). Findings to date have been somewhat mixed, perhaps due to methodological factors and the inherent heterogeneity within the autism spectrum. This study attempts to address this issue by investigating a range of cognitive atypicalities in a large population-based sample comprising children with ASD, their co-twins, and typically developing comparison children (termed 'controls').

Cognitive accounts of ASD can be broadly divided into domain-specific and domain-general theories. Domain-specific theories situate the primary deficit in social processing. Prominent amongst these is the 'Theory of Mind' (ToM) deficit account, which explains the social and communication impairments of ASD as resulting from difficulty representing mental states (e.g. Frith, Morton, & Leslie, 1991). This account has been influential in psychological research, neuroimaging and intervention, although the universality and specificity of ToM deficits has been questioned (Yirmiya, Erel, Shaked, & Solomonica-Levi, 1998). Whether ToM deficits are primary or result from earlier abnormalities of social orienting or social motivation, is also a topic of much debate (Dawson, Webb, & McPartland, 2005; Jones, Carr, & Klin, 2008).

Domain-general accounts of ASD propose that the primary deficit/difference is not in social cognition specifically but lies in, for example, 'executive functions' (EF; Hill, 2004). Executive dysfunction in ASD has been proposed to underlie RRBIs due to a failure to generate new behaviours or shift set. Executive dysfunction has also been hypothesised to explain social/communicative deficits (Kenworthy, Black, Harrison, Della Rosa, & Wallace, 2009).

A number of domain-general accounts suggest areas of superior processing or differences in cognitive style, such as 'weak central coherence' (CC) (Frith, 1989; Happé & Booth, 2008; Pellicano, 2010),

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a bias towards featural processing and reduced configural processing. Superior local processing, but accompanied by intact global processing, is also proposed by 'enhanced perceptual processing' (Mottron, Dawson, Soulieres, Hubert, & Burack, 2006), 'systemising' (Simon Baron-Cohen, 2009) and enhanced discrimination (O'Riordan & Plaisted, 2001) accounts of ASD.

Traditionally, cognitive accounts of ASD have attempted to explain parsimoniously both sociocommunicative impairments and RRBIs as resulting from a single underlying deficit/difference. However, more recently it has been suggested that multiple cognitive accounts may apply, with each explaining distinct symptoms of ASD (Brunsdon & Happé, 2014; Happé & Ronald, 2008; Happé, Ronald, & Plomin, 2006). Thus, ASD might be seen as the result of a combination of cognitive deficits or atypicalities, with ToM deficits explaining sociocommunicative features, executive dysfunction explaining RRBIs, and detail-focus (e.g. CC) explaining uneven cognitive profile and assets. Previous work has been limited in its scope to examine this hypothesis as most studies have investigated a single cognitive domain, with the noteable exceptions of studies by Pellicano (Pellicano, 2013; Pellicano, Maybery, Durkin, & Maley, 2006) and Charman et al. (2011).

The aim of this study was to address the mixed findings in the literature regarding the cognitive features of ASD and to investigate the prevalence of multiple cognitive atypicalities in ASD. Previous studies, which have reported mixed findings, have typically had sample sizes of 15 to 40 individuals with ASD, and have often given tests of only one area of cognition. We aimed to test weak CC, EF and ToM in the same large sample of individuals with ASD. Mixed findings may also reflect differences in sample selection and recruitment (e.g. through specialist clinics, special schools, parent volunteers). We therefore tested a population-based sample, identified as meeting diagnostic criteria for ASD from a longitudinal study of all twins born in the United Kingdom in 1994–6. In addition, we assessed along with the ASD twins, their unaffected cotwins, who may be expected to share some (subclinical) traits or cognitive characteristics, according to family studies of the 'broader autism phenotype' (e.g. Hughes, Plumet, & Leboyer, 1999). Therefore, this study included individuals across the range of ASD traits as well as typically developing comparison participants.

Method

Participants

Participants were part of the Twins Early Development Study (TEDS), a population-based longitudinal study of all twins born in the United Kingdom between 1994 and 1996. The 12,054 families involved at the start of TEDS were reported to be representative of UK families (Haworth, Davis, & Plomin, 2013).

The Social Relationships Study (SR study) focused on those TEDS families with one or both twins meeting diagnostic criteria for ASD. Twins 'at risk' of ASD were identified a) from a parental report of an ASD diagnosis directly to TEDS (via phone at any point or by ticking boxes about diagnoses on postal questionnaires) and/or b) elevated scores on the Childhood Autism Spectrum Test (CAST) (Scott, Baron-Cohen, Bolton, & Brayne, 2002) at age 8 (data available from 6,736 TEDS families). Two hundred and eleven families reported a previous ASD diagnosis in at least one twin, and an additional 203 families had at least one child who scored above cut-off for suspected ASD on the CAST ($\geq$15). Of these 414 families, 326 families were contactable and consented to take part in the second stage of screening. To address possible selection bias and selective attrition in TEDS, a mail-out to child psychiatrists across the United Kingdom and advertisements through the National Autistic Society and the Twins and Multiple Births Association, were carried out to find any additional twin pairs with ASD born between 1994 and 1996. This yielded an additional five twin pairs. Using the ASD module, families completed the Development and Wellbeing Assessment (DAWBA) (Goodman, Ford, Richards, Gatward, & Meltzer, 2000) via a telephone interview. This identified 235 families with at least one child who met DAWBA criteria for an ASD and so were invited to take part in the SR study. Informed parental consent was obtained from 129 families to complete a home visit, including diagnostic and cognitive testing; other families were not traceable or did not consent to in-person assessments. The 129 families who took part were comparable to those eligible for participation (i.e. CAST$\geq$ 15 or suspected ASD) but who did not take part, CAST score (p = .14), socioeconomic status (p = .25) and zygosity (p = .23), but more girls were in the 'high CAST/suspected ASD group' (36%) than the final sample (17%) (Colvert et al., 2014). Twins in the ASD families who did not meet criteria for ASD comprised the 'unaffected cotwin' group in the following analyses.

Information regarding the ascertainment and diagnostic classification procedure can be found in Colvert et al. (2014). Participants were diagnosed with ASD using gold-standard diagnostic instruments; the Autism Diagnostic Interview-Revised (ADI-R; Lord, Rutter, & Lecouteur, 1994) and the Autism Diagnostic Schedule (ADOS; Lord et al., 2000). Additional cut-offs devised by the Autism Genetic Resource Exchange (AGRE) were implemented to identify family members with more subtle ASD symptoms and assigned cases to 'ASD' (AGRE categories Autism and 'Not Quite Autism'), 'Broad Spectrum Disorder', and 'unaffected'. A 'broad spectrum' diagnosis was permitted for the ADOS and corresponded to just below cut-off for diagnostic criteria for an ASD on the ADOS (−2 points). Participants were classified using available information (ADI-R, ADOS, DAWBA). In 37% of the ASD sample (N = 89), the ADI-R and the ADOS classifications were inconsistent. For these cases, diagnostic consensus was reached by a team of clinicians. One twin pair was excluded from analyses since neither twin reached diagnostic cut-off for ASD, but CAST score >12 rendered them unsuitable for inclusion in the control sample. Children were also excluded if there were known circumstances likely to affect the accuracy of diagnosis (N = 2). For current analyses, ASD diagnoses and broad spectrum diagnoses were combined to create one ASD group to cover the complete autism spectrum from severely impaired individuals through to those with more subtle impairments. In the ASD group, 141 adolescents were diagnosed with ASD and 40 adolescents met the definition for a broad spectrum diagnosis. An unaffected cotwin group was also created consisting of 73 cotwins without an ASD or broad spectrum diagnosis.

A comparison control sample with CAST scores less than 12 was recruited via TEDS and matched to the ASD sample on gender, age, IQ, social economic status and zygosity. 80 control twin pairs were recruited, making a total of 209 families visited in their homes by a team of two trained researchers.

The ASD group contained 181 adolescents (13 years 6 months; 150 males), the unaffected cotwin group contained 73 adolescents (13 years 6 months; 27 males) and the control group contained 160 adolescents (12 years, 10 months; 110 males). Table 1 provides further information regarding the age, IQ, gender, zygosity, ADI and ADOS scores of the ASD, cotwin, and control group.

There was a significant difference between groups (ASD, cotwins, control) in age ($F(2,411) = 32.20$, $p < .001$, $\eta^2 = .135$). Tukey post-hoc tests revealed that the control group was significantly younger than both the ASD and cotwin groups ($p < .001$). There were significant differences in IQ across groups ($F(2,411) = 28.23$, $p < .001$, $\eta^2 = .121$). Overall, the ASD group ($M = 90.02$) had a significantly lower IQ score than both the cotwin group ($M = 104.76$, $p < .001$) and the control group ($M = 101.91$, $p < .001$). There were no significant differences in IQ scores between the co-twin and control groups ($p = .476$).

Measures

Intellectual ability. Intellectual ability was assessed using the Wechsler Abbreviated Scale of Intelligence (WASI) (Wechsler, 1999) to obtain an estimated score for IQ. Fourteen nonverbal adolescents completed the Raven's Coloured Progressive Matrices (Raven, Raven, & Court, 1998) and the British Picture Vocabulary Scales-Revised (BPVS) (Dunn, Dunn, Whetton, & Pintillie, 1997) to obtain an estimated score for verbal and performance IQ. To include the low IQ individuals in the subsequent analyses, the 14 nonverbal children were given a provisional WASI full-scale IQ score of 49 (1 point below the lowest possible score on the WASI). This study used the Block Design subtest as a measure of CC. Therefore, the two-subtest version of the WASI (includes Matrix Reasoning and Vocabulary) was used as an estimate of IQ.

Cognitive task battery. The measures (with the targeted components), key variables, number of trials, and reference to procedure are shown in Table 2.

Procedure

Home visits were made to all ASD and control families by two trained researchers. The ASD families completed two home visits, which lasted approximately 6 hr in total. The ASD families completed gold standard diagnostic assessments; the ADOS (Lord et al., 2000) and the ADI-R (Lord et al., 1994). The control families completed one home visit, which lasted approximately 2 hr. Both the ASD and control families completed an extensive cognitive battery to measure IQ, language ability, CC, executive function (EF) and ToM ability. The batteries were administered in a counterbalanced order with two fixed orders of tasks. A different experimenter assessed each participant within the twin pair in order to reduce possible experimenter bias.

Results

All twins were treated as singletons in the present analyses to allow comparisons between groups of adolescents with ASD (termed ASD group), unaffected cotwins, and a control group. The reaction time from Embedded Figures Test (EFT), total error score in the Sentence Completion Task, the coherence score and planning score from the Planning Drawing Task, reversal errors in ID/ED, and errors in Penny Hiding Game were reflected so that a higher score indicated better performance in all tasks.

Preliminary data analyses indicated that some of the data did not meet assumptions of a normal distribution. Data from six of the cognitive measures were skewed (value > 2) and data from four of the cognitive measures had a leptokurtic distribution (value > 3). All variables were normalised using a Van der Waerden transformation.

Pearson's correlation analyses were carried out to investigate if age and IQ were related to performance on cognitive measures. For all groups, age was not significantly correlated with cognitive measures, except for Block Design Task performance in the ASD and control groups (ASD: $r = -.24$, $p < .01$, controls: $r = -.44$, $p < .001$). In the ASD group, IQ was significantly related to performance on most cognitive measures (12/13, all rs > .21, all ps < .01), except for Homographs Reading Test ($r = .14$, $p = .094$). Correlational analyses revealed fewer significant relationships between IQ and performance on cognitive measures for the unaffected cotwin group (2/13 measures) and the control group (4/13) as compared to the ASD group. Therefore, IQ-

Table 1 Participant characteristics

	ASD			Unaffected Cotwins (CT)			Controls (TD)			Sig.
	N	*M* (*SD*)	Range	*N*	*M* (*SD*)	Range	*N*	*M* (*SD*)	Range	*p*
Age (years)	181	13.49 (0.69)	12.08–16.25	73	13.50 (0.65)	12.25–15.17	160	12.79 (1.10)	10.92–15.58	<.001
IQ (WASI 2-subtest)	153	94.07 (16.91)	55–128	71	104.76 (13.73)	61–130	158	102.00 (15.19)	56–142	<.001
IQ (imputed score)	181	90.02 (20.34)	49–128	73	104.76 (13.54)	61–130	160	101.91 (15.14)	56–142	<.001
ADOS total (raw)[a]	174	11.38 (6.14)	0–26	71	1.83 (2.23)	0–10	–	–	–	<.001
ADI total[a]	177	37.64 (16.19)	3–70	72	5.46 (5.03)	0–23	–	–	–	<.001
Males: Females		4.84:1			1.70:1			2.20:1		<.001
MZ:DZ		1:2.55			1:23.33			1:1.86		.002

ASD, autism spectrum disorder; CT, unaffected cotwins; DZ, dizygotic twin pairs; M, mean average; MZ, monozygotic pairs; N, number of participants; *SD*, standard deviation; TD, typically developing controls.
[a]Higher score = more severe.

Table 2 Battery of cognitive tasks used in Social Relationship Study (SR study) by cognitive domain with references to studies describing task procedure

Cognitive measure	Key variable	Number of trials	Reference for task procedure	Expected direction of group effects
Central coherence				
Embedded figures test (EFT)	Reaction time (seconds)	15 trials; 7 child EFT items, 8 standard EFT items	Shah & Frith (1983)	TD > ASD
Block design task	Accuracy	10 trials	Shah and Frith (1993)	ASD > TD
Homographs reading test	Context effect	16 sentences	Happé (1997)	TD > ASD
Planning drawing task, part A	Coherence score	2 items; house & snowman	Booth et al. (2003)	TD > ASD
Sentence completion task	Error score	10 sentences (plus 5 control)	Booth and Happé (2010)	TD > ASD
Executive function				
Letter fluency task (FAS) (mental initiation)	Number of correct responses	3 trials; F, A, S	Turner (1999)	TD > ASD
Luria hand game (inhibition)	Conflict score	10 trials	Hughes (1996)	TD > ASD
Intradimensional/ Extradimensional task (ID/ED) (cognitive set-shifting)	Reversal errors	9 stages; progress on to next stage after 8 correct trials within 50 trials.	Hughes et al. (1994)	TD > ASD
Planning drawing task, part B (planning)	Planning score	2 items; house & snowman	Booth et al. (2003)	TD > ASD
Theory of mind				
Penny hiding game	Error score	6 trials	Baron-Cohen (1992)	TD > ASD
Triangles animation task	Mentalising score	4 trials; ToM only	Abell, Happé, and Frith (2000)	TD > ASD
False-belief stories	First- and second-order false-belief score	3 stories; 3 first-order, 2 second-order questions	Perner, Frith, Leslie, and Leekam (1989)	TD > ASD

adjusted standardised residuals for cognitive task performance were used in all further analyses (unless otherwise stated). The standardised residuals for the ASD and cotwin group are obtained from the regression line fit when fitting each cognitive measure as a dependent variable in a linear model with IQ as a predictor variable, according to the control group (Thomas et al., 2009).

Table 3 shows the mean performance (raw scores) for each CC, EF, and ToM measure by group. One-way analyses of variance (ANOVA) to investigate group differences (ASD, cotwins, controls) in cognitive task performance are reported in Table 3, with post hoc comparisons using Tukey tests. Figure 1 shows the mean performance of the ASD group and the unaffected cotwin group relative to the control group on all cognitive measures.

Due to the heterogeneity in cognitive performance within the ASD group, means may not fully reflect performance across the groups. To compare performance further, frequencies were calculated for atypical performance on each cognitive measure. Atypical performance was defined as one standard deviation above (EFT and Block Design Task only) or below (all other tasks) the control group mean. The number of cognitive tasks on which participants performed atypically is shown in Table 4. Results indicated that 63% of individuals with ASD performed atypically in three or more cognitive measures, compared to 31% of unaffected cotwins and 23% of controls. The ASD group performed atypically on significantly more tasks than the unaffected cotwin and control groups; $F(2,385) = 36.28$, $p < .001$, $\eta^2 = .159$; post hoc Tukey tests ps < .001. The unaffected cotwin group and control group did not differ in the number of tasks performed atypically ($p = .279$).

We examined how many individuals showed atypicalities across the cognitive domains, by totalling the number of participants performing one standard deviation above (EFT and Block Design only) or below the mean on at least one measure in each cognitive domain. Figure 2 shows how many individuals with ASD, unaffected cotwins and controls had no cognitive atypicalities, single cognitive atypicality, dual cognitive atypicalities, or multiple cognitive atypicalities. The CC domain showed the highest proportion of individuals with atypical performance solely in that domain, perhaps due to more tasks assessing this aspect of cognition. The most frequently cooccurring cognitive atypicalities were in the CC and EF domains. Furthermore, there was a significant relationship between group (ASD, unaffected cotwin, control) and presence of multiple cognitive atypicalities (X^2

Table 3 Performance on cognitive measures for ASD and comparison groups (raw scores) and group differences in cognitive measures (transformed scores)

	ASD		Unaffected Cotwins (CT)		Controls (TD)		Group differences (IQ-adjusted residuals; $p < .05$) ANOVA			
Measure	*N*	*M (SD)*	*N*	*M* (*SD*)	*N*	*M* (*SD*)	*F*	*p*	η^2	Post hoc Tukey
Central coherence										
EFT (reaction time, seconds)[a]	159	20.40 (10.70)	70	17.64 (7.71)	158	17.90 (9.32)	0.31	.733	.002	n.s.
Block design task (score)	154	49.55 (13.02)	71	53.07 (10.60)	151	53.07 (10.58)	0.89	.410	.005	n.s.
Homographs reading test (context effect)	138	1.70 (1.67)	71	2.13 (1.29)	151	2.12 (1.31)	3.91	.021	.021	TD > ASD (CT n.s.)
Sentence completion task (error score, max = 20)[a]	154	3.51 (3.02)	66	2.46 (2.64)	158	2.28 (2.49)	7.38	.001	.038	ASD > TD, CT
Planning drawing A (coherence score, max = 12)[a]	158	1.15 (0.93)	71	0.82 (0.74)	156	0.80 (0.73)	9.89	<.001	.049	ASD > TD, CT
Executive function										
Letter Fluency Task (score)	146	5.26 (2.63)	69	5.23 (2.17)	149	5.53 (2.51)	0.83	.438	.005	n.s.
Luria Hand Game (conflict score, max = 10)	145	8.44 (2.72)	69	9.51 (1.13)	142	9.78 (0.60)	26.95	<.001	.132	TD, CT > ASD
ID/ED (error score)[a]	149	2.68 (2.69)	71	2.19 (2.32)	155	1.92 (1.62)	3.58	.029	.019	ASD > TD (CT n.s.)
Planning drawing B (planning score, max = 4)	158	1.22 (1.01)	71	0.97 (0.89)	156	0.82 (0.82)	5.41	.005	.028	TD > ASD (CT n.s.)
Theory of mind										
Penny hiding game (error score)[a]	148	0.98 (1.93)	68	0.54 (1.11)	152	0.11 (0.50)	19.04	<.001	.094	ASD > CT > TD
Triangles animation task (mentalising score, max = 4)	138	1.38 (1.26)	66	2.56 (1.21)	148	1.68 (1.21)	8.20	<.001	.045	CT > TD > ASD
False-belief stories (score, max = 10)	134	9.22 (1.34)	69	9.88 (0.47)	153	9.77 (0.69)	12.50	<.001	.066	TD, CT > ASD

ASD, autism spectrum disorder; CT, unaffected cotwins; EFT, Embedded Figures Test; M, mean average; N, number of participants; n.s., not significant; *SD*, standard deviation; TD, typically developing controls.
[a]Higher score = poorer performance.

(2) = 41.20, $p < .001$); the ASD group showed the highest proportion of multiple cognitive atypicalities (32% of ASD group) compared to the unaffected cotwins (11%) and control groups (6%).

In the ASD group, correlation analyses indicated that the number of cognitive atypicalities was related to the severity of ASD symptoms (as measured by ADOS calibrated severity scales [ADOS-CSS]; Gotham, Pickles, & Lord, 2009), $r = .27$, $p = .001$. An ANOVA revealed a significant difference in the severity of ASD symptoms (ADOS-CSS) according to the number of cognitive atypicalities (none, single, dual, multiple), $F(3,153) = 3.39$, $p = .020$, $\eta^2 = .062$, with Tukey post hoc comparisons indicating significantly more severe symptoms in ASD individuals with multiple atypicalities ($M = 6.75$) compared to ASD individuals with no cognitive atypicalities ($M = 4.50$, $p = .026$).

Discussion

The aim of this paper was to investigate the pattern of cognitive atypicalities in ASD in a population-based sample to clarify the mixed findings in the literature. Group differences on a cognitive battery devised to assess ToM, EF and CC and the prevalence of multiple cognitive atypicalities were reported for individuals with ASD, their unaffected cotwins, and comparison typically developing individuals. The patterns of results from the group comparisons are discussed in this section.

The 'weak central coherence' account of ASD suggests that individuals with ASD will be better at tasks where a local processing bias is beneficial, such as the EFT (Happé & Frith, 2006) and Block Design Task (Shah & Frith, 1993). However, in this study the ASD group did not significantly outperform the unaffected cotwins or the control group on the EFT or on the Block Design Task. This finding is in contrast to previous studies findings of superior performance on the EFT and Block Design Task in adults with ASD (Jolliffe & Baron-Cohen, 1997; Shah & Frith, 1983) but in line with findings from White and Saldana (2011), who reported that children with ASD performed similarly to typically developing children on the EFT.

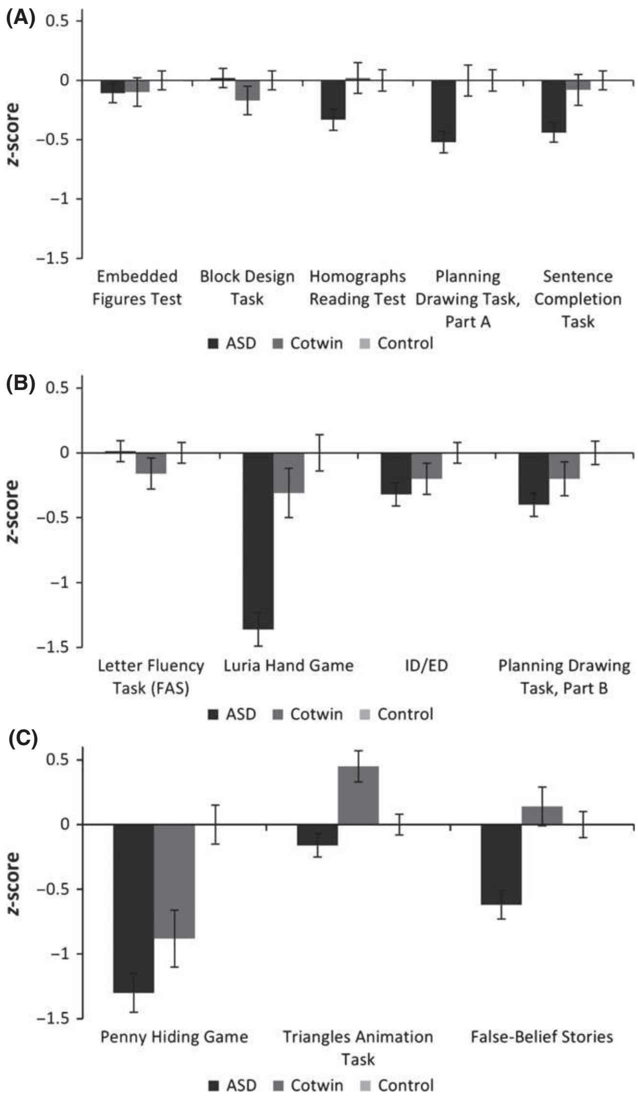

Figure 1 Performance on cognitive measures assessing (A) central coherence, (B) executive function, and (C) theory of mind, for all groups after accounting for IQ. Scores are presented as z-scores relative to the control group. Error bars show standard error

The 'weak central coherence' account of ASD also suggests that individuals with ASD will have poorer performance on tasks which place demands on global processing compared to typically developing children. In this study the ASD group performed below the typically developing control group in all three CC tasks tapping global processing, in support of previous findings that individuals with ASD perform worse than typically developing individuals on the Homographs Reading Test (Happé, 1997), Planning Drawing Task (coherence score; Booth, Charlton, Hughes, & Happé, 2003) and the Sentence Completion Task (Booth & Happé, 2010).

In support of the executive dysfunction account, the ASD group performed below the control group in two tasks measuring EF, specifically those purporting to measure cognitive set-shifting (IDED) and planning (Planning Drawing Task, Part B), and below both comparison groups on a test of inhibition (Luria Hand Game). Previous findings have also reported poor performance by children with ASD in the Luria Hand Game (Hughes, 1996), ID/ED (Ozonoff et al., 2004) and the Planning Drawing task (Booth et al., 2003). No group differences were found for the test of generativity used in this study (Letter Fluency Task).

The ASD group performed significantly below both comparison groups in the Penny Hiding Game, Triangles Animation Task and the False-Belief Stories. These findings provide additional support for a ToM deficit in ASD.

There was a mixed pattern of results regarding whether the unaffected cotwins of those with ASD shared cognitive features with their affected siblings. The unaffected cotwins outperformed the ASD group in the Sentence Completion Task (CC), Luria Hand Game (EF) and all three ToM tasks. However, on all other cognitive tasks (exception; Penny Hiding Game) the unaffected cotwins were not significantly better than the ASD group, nor significantly worse than the control group, even when significant differences were found between the ASD and control group. This may reflect an intermediate cognitive profile in siblings of those with ASD, or it could be due to a lack of statistical power to detect group differences; this group was approximately half the

Table 4 Number (percentage) of individuals with ASD, their unaffected cotwins, and controls performing atypically on cognitive measures (defined as 1 *SD* above/below the control group mean)

Number of cognitive measures in the atypical range	ASD (*N* = 158) *N* (%)	Unaffected Cotwins (*N* = 71) *N* (%)	Controls (*N* = 159) *N* (%)
0	12 (7.6)	7 (9.9)	19 (11.9)
1	19 (12.0)	16 (22.5)	56 (35.2)
2	27 (17.1)	26 (36.6)	47 (29.6)
3	41 (25.9)	13 (18.3)	25 (15.7)
4	25 (15.8)	6 (8.5)	9 (5.7)
5+	34 (21.5)	3 (4.2)	3 (1.9)

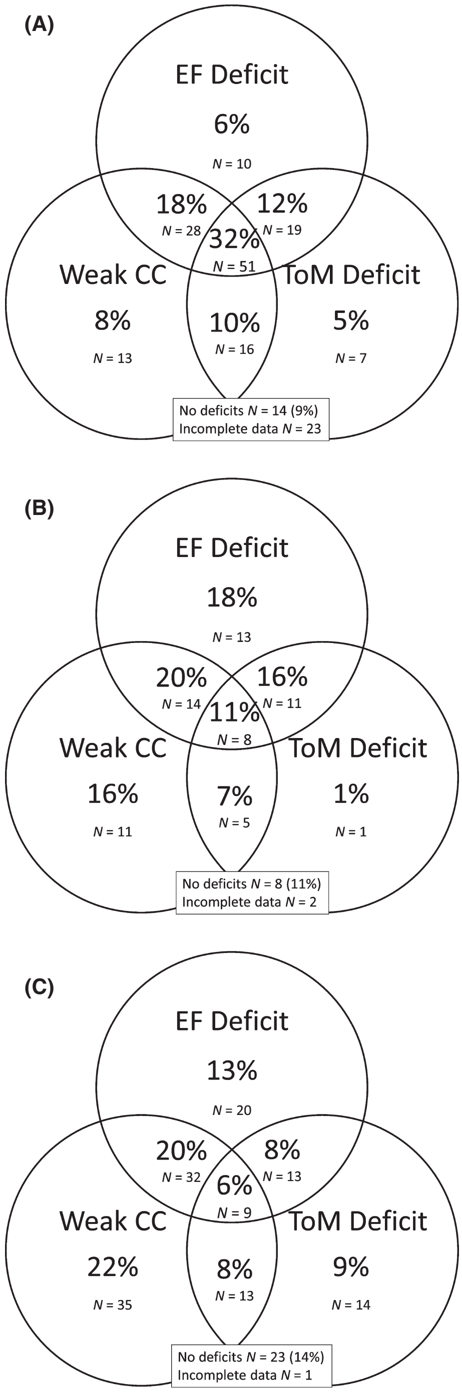

Figure 2 Venn diagrams showing the number and percentage of participants (A) in the ASD group, (B) the unaffected cotwin group, and (C) the typically developing control group, with atypical performance (1 *SD* above/below control group mean) in the three cognitive domains. The central region indicates atypicalities in all three cognitive domains

size of the other two groups. In contrast to the findings of Hughes, Russell, and Robbins (1994), we did not find evidence of EF deficits in siblings of children with ASD, nor did the siblings show weak CC on the present tasks. There was evidence that the broader autism phenotype included ToM deficits, but only in the Penny Hiding Task. It should be noted that the unaffected cotwins in fact performed substantially better in one mentalising task (Triangles Animation Task) than both the ASD and control groups, possibly indicating compensatory skills or protective factors.

The ASD group had a greater number of cognitive deficits/differences overall than both of the other groups. This finding supplements Pellicano (2010) study, in which children with ASD showed difficulties in false-belief understanding, higher-order planning and cognitive flexibility at ages 4–7 years and 7–10 years old relative to typically developing controls. Additionally, in this study, nearly a third of the adolescents with ASD had multiple cognitive atypicalities, i.e. they had atypical performance in tasks across cognitive domains. Pellicano (2010) also found that at age 4–7 years, over half of individuals with ASD had multiple cognitive atypicalities, which declined to 19% by age 7-10 years. However, multiple cognitive atypicalities were not exhibited by every individual with ASD, as might be predicted from a strong version of the fractionated triad/ multiple deficit account proposed by Happé et al. (2006). Instead, multiple cognitive atypicalities seem to be characteristic, but not a universal feature, of ASD.

In this study the individuals with ASD who had multiple cognitive atypicalities also had more severe ASD symptomatology than those with no cognitive atypicalities. As suggested by Happé et al. (2006), this highlights the need to move away from single cognitive accounts of ASD that reduce the behavioural symptoms of the condition to a single underlying cognitive deficit. Instead, a multiple cognitive account of ASD, incorporating several cognitive functions, could provide an explanation for the symptomatology of ASD (Brunsdon & Happé, 2014; Happé & Ronald, 2008; Happé et al., 2006). Previous work has attempted to address whether cognitive atypicalities, either alone or together, are related to the behavioural features of ASD (reviewed in Brunsdon & Happé, 2014). Only a handful of studies have specifically investigated the relationship between test performance in multiple cognitive tasks and the various symptom domains of ASD (Joseph & Tager-Flusberg, 2004; Pellicano, 2013; Pellicano et al., 2006). Joseph and Tager-Flusberg (2004) reported that much of the relationship between ToM, EF and symptom severity in ASD could be accounted for by language ability. However, ToM ability and higher level EF were directly related to the severity of communication symptoms in ASD, but not to reciprocal social interaction and RRBIs. Contrary to Joseph and Tager-Flusberg's (2004) findings and their own predictions, Pellicano et al. (2006) found that performance on CC, EF and ToM tasks failed to correlate with any of the three symptom domains in ASD (Pellicano et al., 2006). In a longitudinal analysis, ToM ability was related to social-communication symptoms, and EF was related to both social-communication symptoms and RRBIs, and CC did not relate to any symptom domains (Pellicano, 2013). Future work is needed to resolve conflicting results and to investigate further whether cognitive atypicalities, either alone or together, are related to the behavioural features of ASD contemporaneously or developmentally.

The SR study has many strengths; it is a large population-based study, with an ASD group that covers the whole ASD spectrum from those with broader spectrum diagnoses through to those who are severely affected, along with a large typically developing comparison group. As the sample contained siblings (i.e. the unaffected cotwins), it was possible to investigate whether cognitive deficits are part of the broader autism phenotype. The study included a wide range of cognitive tasks as well as IQ, allowing us to establish which group differences in ToM, EF or CC survive correction for differences in general intellectual functioning between the groups.

Several limitations need to be considered when reflecting upon the results of the study. First, some potentially eligible families did not enrol in the SR study, and as such the sample, while population-based, is self-selected. Secondly, the adolescents were approximately 13 years of age when they were tested, but many of the tasks are more commonly used to assess younger children. The task battery was designed to assess a wide range of abilities, given the variability of IQ in the ASD group. However, as a result, many adolescents scored close to ceiling on the Luria Hand Game and False-Belief Stories and close to floor (in error scores) on the Planning Drawing Task and Penny Hiding Game. In principle, floor and ceiling effects constrict range and may therefore mask true group differences. In the present analyses, IQ was regressed out and a transformation applied prior to analysis to reduce skewness in the cognitive task data. Our results showed significant group differences even in cognitive tasks that showed some floor/ceiling effects. Thirdly, the tasks may not have fully encapsulated the cognitive ability that they purport to measure, and may not have been equally discriminating across domains. For example, there is no single task/battery that can exhaustively measure all aspects of EF, and tests of individual EFs are rarely ‘process pure’.

Conclusion

The present results suggest that multiple cognitive atypicalities are characteristic, but not a universal feature, of ASD. Several group differences were found in cognitive tasks that are purported to test CC, EF, and ToM. Analysis of individual performance showed that no one deficit was universal in the ASD group. However, participants with ASD had more cognitive atypicalities overall than either unaffected cotwins or typically developing control participants. Furthermore, nearly a third of the ASD group had multiple cognitive atypicalities, i.e. they showed atypical performance in CC, EF and ToM. The next step will be to investigate in this large, population-based sample whether specific cognitive atypicalities, either alone or in combination, are related to specific behaviours characteristic of ASD.

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Key points

- The findings in the literature are somewhat mixed regarding the cognitive features of ASD.
- This study investigated a range of cognitive atypicalities and the prevalence of multiple cognitive atypicalities in a large population-based sample comprising children with ASD, their nonclinical cotwins and typically developing comparison children.
- The ASD group showed atypical performance in significantly more cognitive tasks than the unaffected cotwin and control groups.
- Nearly, a third (32%) of the ASD group had multiple cognitive atypicalities compared to 11% of the unaffected cotwins and 6% of the control group.

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