ORIGINAL PAPER

# Is Talent in Autism Spectrum Disorders Associated with a Specific Cognitive and Behavioural Phenotype?

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Abstract Parents of 125 children, adolescents and young adults with autism spectrum disorders completed a newly developed questionnaire aimed at identifying cognitive and behavioural characteristics associated with savant skills in this group. Factors distinguishing skilled individuals were then further investigated in case studies of three individuals with exceptional skills for music, art and mathematics. The findings from the case studies largely confirmed the results from the questionnaire study in showing that special skills are associated with superior working memory and highly focused attention that is not associated with increased obsessesionality. Although intellectual impairment and a local bias have been widely associated with special skills in the savant literature, neither the screening nor case studies provided strong evidence for such associations.

**Keywords** Autism spectrum disorders (ASD) · Special skills · Savants · Weak central coherence · Memory · Intelligence

### Introduction

Although research into autism spectrum disorders (ASD) has largely focused on the socio-communicative

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Psychology Department, Goldsmiths, University of London, New Cross, London SE14 6NW, UK e-mail: ps702eb@gold.ac.uk impairment characterising these disorders, the presence of unusual or "savant" skills in this group has also attracted the attention of researchers. Despite some controversy about definitions of "savant syndrome" (Miller 1998; Heaton and Wallace 2004), the term is typically used to describe individuals with intellectual impairment and one or more surprisingly good skills, often, though not exclusively, in the domains of music, art and calculation (Miller 1999). Treffert (1989, 2009) proposed a three-tier system in which prodigious and talented savant categories are used to describe individuals with extensive abilities, where the term "splinter skill" is used to describe more constrained, low-level types of skills, for example the ability to memorise car number plates.

In the most recent empirical study of special skills in ASD, Howlin et al. (2009) differentiated between parental reports of savant skills and exceptional cognitive skills as measured by performance on subtests of the Wechsler Intelligence Scales. From a sample of 137 individuals with autism, 39 (28.5 %) met criteria for either a savant and/or superior cognitive skill. Howlin et al. (2009) concluded that unusual talents might be found in up to one third of all individuals with ASD. These findings, together with results from recent studies showing increases in the numbers of intellectually unimpaired individuals with a current diagnosis of ASD (Williams et al. 2008; Charman et al. 2010) suggest that some re-definition of the savant syndrome is warranted. Although the term 'idiot-savant' was abandoned for largely ethical reasons, this most recent research suggests that the implied association between intellectual impairment and special skills in autism also lacks scientific validity.

Theoretical accounts of autism that have been relevant to the question of special skills include the Weak Central Coherence (WCC) (Frith 1989; Happé 1999; Happé and Frith 2006), Enhanced Perceptual Functioning (EPF) (Mottron and Burack 2001; Mottron et al. 2006, 2009) and hyper-systematising theories (Baron-Cohen 2006, 2008, Baron-Cohen et al. 2009). The findings from several experimental studies have implicated a local information processing bias in savant skills for art (Mottron and Belleville 1993; Pring et al. 1995), for music (Heaton et al. 1998: Mottron et al. 2000) and for calendar calculating (Heavey et al. 1999). However, calendar calculating and musical improvisation are generative skills that rely on global domain-specific knowledge and it is not obvious how a local bias can account for these. In acknowledgement of this, Mottron et al. (2009) have extended their earlier Enhanced Perceptual Functioning theory (Mottron and Burack 2001) to encompass enhanced pattern detection abilities. According to this account, increased sensitivity to perceptual information and good pattern detection abilities are implicated in the acquisition of domain specific knowledge and the emergence of savant skills in autism. Baron-Cohen et al.'s (2009) latest version of the hypersystematising account also suggests that an autism specific ability to identify and structure repeated patterns in stimuli may help explain the high preponderance of savant skills in this group.

In addition to a local bias and pattern recognition skills, superior memory has been implicated in savant skills. Treffert (2009) who has written extensively about savantism has implicated what he describes as "characteristically very deep" memory that is "exceedingly narrow within the confines of the accompanying special skill" (p. 1353). Several empirical studies have investigated memory skills in groups of savants with autism. For example, O'Connor and Hermelin (1987) presented 8 savant artists (4 with autism) and IQ matched non-savants with a variety of simple tasks involving memory recognition and graphic reproduction of shapes from memory. While the performance of both groups was equal in terms of memory recognition (a matching task), the savants demonstrated superior graphic reproduction from memory and this was independent of IQ. In another study, Hermelin and O'Connor (1990) studied 8 savant artists (4 with autism) and compared their drawings to those of typically developing (TD) gifted artists across 4 conditions, one of which required participants to draw a 3D scene from memory. Although the drawings produced from memory were equal in terms of artistic merit, the TD group actually scored higher than the savants in terms of accuracy. Whilst the findings from these two group studies are contradictory and do not clearly support the view that visual memory is enhanced in savants with autism, case reports invariably implicate enhanced visual memory in these individuals (e.g., Selfe 1977; Sacks 1995). Exceptional musical memory in autism was first described by Kanner (1943) and has subsequently been linked to absolute pitch (Heaton et al. 1998; Heaton 2003) and knowledge about musical structure (e.g., Sloboda et al. 1985; Miller 1989; Young & Nettelbeck 1995; Mottron et al. 2000a, b; Pring et al. 2008) in savants.

Other work on memory has been conducted with calendar calculating savants. Heavey et al. (1999) studied memory function in 8 calendar calculators (7 with autism) and observed superior recall of date (domain-specific) information. However, these participants did not possess unusually increased short- and long-term memory capacities as measured by digit span or recall of words from a list. This finding is surprising given that several studies, some of which included large participant samples (Young & Nettelbeck 1995; Bölte and Pouka 2004), have described increased working memory capacity, measured using the digit span test, in individuals with autism and special skills (Spitz and LaFontaine 1973; Rimland and Hill 1984; Rumsey et al. 1992; Young & Nettelbeck 1995; Bölte and Pouka 2004). Superior digit span does not appear to be a general cognitive strength in ASD. For example, in a recent study implementing the digit span task with adults with autism and TD controls, Poirier et al. (2011) reported an autism-specific short-term memory deficit that was directly related to difficulties in encoding the order of presented items. Bölte and Pouka (2004) compared 33 savants and 26 non-savants with ASD and concluded that deficits in working memory were less apparent in savants. Taken together this evidence suggests that spared, or indeed superior, digit span is specifically associated with talent in ASD. The role of memory in special skills in ASD will be further investigated in the screening and validation studies to be described.

Given the increased prevalence of savant skills in populations of individuals with ASD (Rimland 1978) it is unsurprising that the diagnostic characteristics of autism have been implicated in the development and maintenance of special skills in this group. In the first empirical study to associate savantism and ritualistic, obsessive behaviours, O'Connor and Hermelin (1991) asked caretakers of savants with and without autism, together with non-savant controls matched on diagnosis and intelligence, to complete a 15-item repetitive behaviours questionnaire. The comparison of the savant groups with and without autism failed to reveal significant differences on any of the 13 questions that probed routines and checking and hoarding behaviours. Group differences only emerged on two of the questionnaire items. Savants with autism were more likely to order their possessions and demonstrate an increased interest in one particular topic than non-autistic savants. Whilst a tendency to order possessions is commonly observed in autism, highly focused attention, in response to a special interest, is not uniquely

associated with autism. For example, in a recent study, DeLoache et al. (2007) observed an unusually narrow focus of attention in TD children with special interests. Questions about obsessionality and definitions of talent related focus will be further addressed in the questionnaire and case studies.

Sensory abnormalities, included in DSM-III but not DSM-IV criteria for ASD, have also been implicated in savantism. Rimland (1978) has suggested that sensory sensitivities may coincide with special skills. For example, the intellectually impaired calendar calculating twins George and Charles possessed the unusual ability to pick out their own bedroom slippers using olfaction alone (Horwitz et al. 1969). However, in the limited number of studies examining olfaction in ASD, it has been broadly concluded that the ability to identify odours is impaired (Suzuki et al. 2003; Bennetto et al. 2007). More recently, Baron-Cohen et al. (2009) have included sensory sensitivities as a critical component in their hyper-systematising theory. In line with the EPF theory (Mottron et al. 2009) they implicate atypical low-level perception in the emergence of special skills in ASD. In concluding their paper, Baron-Cohen et al. (2009) suggest that "the search for the association between autism and talent should start with the sensory hypersensitivity, which gives rise to the excellent attention to detail, and which is a prerequisite for hypersystematising" (p. 1382). Sensory processing in talented relative to non-talented ASD individuals will be investigated in the current study.

The phenomenon of special skills in autism has important theoretical implications. For example, it has been suggested that savant skills may play a role in enabling researchers to genetically distinguish subgroups within the autism spectrum (Nurmi et al. 2003) and Mottron et al. (2006) have discussed preliminary ideas for savant syndrome as an autistic model for sub-typing PDD. The main aim of the present research was to devise and pilot a new screening questionnaire for the identification of special skills in ASD, drawing on factors highlighted in the research literature (i.e., local bias, memory skills, obsessionality/repetitive interests and sensory abnormalities). Results from the analysis of the questionnaire data then provided the rationale for carrying out case studies on children with ASD and skills in the classic savant domains of music, art and calculation. In addition to skill validation assessments, the cognitive and clinical characteristics of the savants were tested using an extensive test battery. Should the questionnaire study isolate factors that distinguish skilled from nonskilled participants, we would expect that these same characteristics might also be observed in the subjects of the case studies.

### **Study 1: Ascertainment**

#### Method

### **Participants**

Participants were recruited via the National Autistic Society (NAS) and specialist educational establishments in London, UK. E-mails outlining the research proposal and requesting that the questionnaire be forward on to parents and carers were sent to 81 NAS branch officers in the UK as well as to three autism schools that have previously participated in our research studies. 125 primary carers of individuals with ASD (115 mothers, 9 fathers, 1 permanent foster carer) returned completed questionnaires. Ninetyone percent of the screened population were male with ages ranging between 3 and 20 years (M = 10 years,SD = 4.11 years). Specifically, we screened 88 children (3-12 years), 33 adolescents (13-17 years) and 4 adults (18-20 years). Participants had been diagnosed with either autism (31 %), ASD or PDD (nos) (31 %) or Aspergers Syndrome (25 %) by a range of health professionals, including clinicians, child psychologists and paediatricians. The remaining 13 % were diagnosed with autism, ASD or Aspergers syndrome with co-occurring ADHD, epilepsy, SLD or intellectual impairment.

### Questionnaire Development

The new screening questionnaire was designed in accordance with Kline's (1986) psychological test construction theory and aimed to (1) identify individuals with and without reported special skills and (2) provide preliminary data on basic cognitive and diagnostic profiles distinguishing the two groups.

Forty-two items, testing five factors, were included in the profiling section of the questionnaire. The factors probed (1) socio-communicative deficits (2) memory (3) special interests (4) restricted and repetitive behaviour and (5) sensory sensitivity. Individual items were drawn from various standardised assessments, for example the Social Communicative Questionnaire (SCQ; Rutter et al. 2003) and the Short Sensory Profile (Dunn 1999). The sociocommunication factor (9 items) included statements regarding the individual's ability to understand the feelings of his/her peers, preference to interact alone or with others, responsiveness to peer initiations, imaginative play/creativity and communication with others. Memory (8 items) probed memory for facts, dates, places, autobiographical events, general information, things that have happened to others, things of personal interest and general memory skills. The special interests factor (8 items) probed the age appropriateness and unusualness of any identified special interests as well as the degree of intensity and preoccupation that they elicited. A further item probed the individual's range of interests. Sensory sensitivity (9 items) probed hyper or hypo-sensitivity in response to various sensory stimuli. Finally, restricted and repetitive behaviour (8 items) concerned changes in routine, ordering of objects, rituals, stereotypical mannerisms, obsessions and preoccupation with parts of objects (probing WCC). Three psychologists, blind to the research aims, were asked to match items to factors to check face validity and this yielded 100 % agreement.

The special skills section of the questionnaire comprised 9 structured and open-ended questions that elicited information about current skills. For the purposes of the questionnaire, special skills were defined as 'any marked ability that stands in contrast to his/her (the individual's) overall disability'. In addition to parental response to this item, a further item asked if any person outside of the family unit had ever commented on the skill that the parent might be reporting. Questions probed skill onset, time spent engaging in skill related activity, formal skill-related training, similar skills in family members and the extent that skills were thought to hinder or facilitate development in other areas of functioning. The type, extent and number of skills were also probed. The skilled group was formed of individuals whose parents had agreed or strongly agreed that their child possesses one or more skill; the non-skilled group comprised those whose parents disagreed or strongly disagreed that this were the case. Data from parents who neither agreed nor disagreed to this item were excluded from analysis.

All items were adjusted to a 7-point Likert scale. This scale ranged from 1 (Strongly Disagree) to 7 (Strongly Agree) where 4 represented neither agreement nor disagreement with that statement. Respondents were invited to provide more detailed, open ended information should they wish to by making notes on the questionnaire. In order to achieve psychometric validity, the profiling items were randomised and some reverse phrased. The questionnaire takes approximately 10–15 min to complete and is shown in the appendix.

# Results

### Prevalence

The skilled group comprised 42 % of the total sample screened (52 out of 125). Of these individuals, 93 % were male and they were diagnostically heterogeneous: 32 % autism, 32 % ASD or PDD (including PDD nos), 26 % Aspergers syndrome and 10 % autism, ASD or Aspergers syndrome and a co-morbid disability. Surprisingly, given that definitions of the savant syndrome traditionally assume the presence of intellectual impairment, only 5 % of this group were reported to be intellectually disabled. Of those in the non-skilled group, 90 % were male and of these 10 % had an intellectual disability.

Table 1 details the frequency of skill types reported together in the different diagnostic categories.

### Characteristics of Skilled Individuals

The majority of parents of skilled individuals reported a single skill (55 %). However, multiple skills, up to six in some cases, were not uncommon. For the majority of cases (78 %) the skills had developed from an early age (6 % disagreed that this was the case for their child). In almost 90 % of cases the skills had been remarked upon by people outside of the family unit. The vast majority of skills had not been supported by formal training or tutoring (78 %). Indeed, only

 Table 1 Skill types and diagnosis according to reported frequency (shown in %)

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Skill type	N	Autism (1) (%)	N	Asperser syndrome (2) (%)	Ν	ASD/PDD (3) (%)	N	(1) (2) or (3) and another co-morbid disability or intell. impairment (%)	Total N
Memory/knowledge	9	26	8	23	14	40	4	11	35
Mathematical/numerical	11	58	4	21	3	16	1	5	19
Artistic	2	17	4	33	6	50	0	0	12
Music	4	33	3	25	2	17	3	25	12
Reading/vocabulary	3	25	2	17	6	50	1	8	12
Spatial	3	30	1	10	5	50	1	10	11
ICT	5	45.5	4	36.4	1	9.1	1	9.1	11
Other	2	33	1	17	2	33	1	17	6
Mechanical	1	25	3	75	0	0	0	0	4

N number of participants, ASD participants diagnosed with autism spectrum disorder, PDD participants diagnosed with pervasive developmental disorder

5 % reported positively on this item. However, it was noted that 36 % were reported to have one or more family members with similar skills. Just over 65 % of parents reported that their child spends a lot of time engaged in skill related activity (19 % disagreed). One quarter of parents believed that this time engaged in skill related activity interferes with development (53 % disagreed). The vast majority of parents did not make additional comments on the questionnaire. The few comments that were made were reviewed but did not provide any significant information.

### Factor Analysis of the Profiling Data and Reliability

Factor analysis using promax (oblique) rotation was carried out on the profiling data. Three out of five factors revealed perfect loadings as predicted: socialisation and communication, memory and sensory sensitivity. The other two factors, special interests and restricted, repetitive behaviour, consisted of predicted and mixed items, meaning that some items loaded as expected while other items unexpectedly loaded onto the other factor. In order to account for the factor loadings, these two factors were subsequently renamed obsessions and special interests (items 25, 12, 22, 32, 30) and repetitive behaviour and unusual interests (items 7, 37 and 30). Given the similarity in constructs between the two originally conceived factors (all items correspond to DSM-IV Axis 3 for diagnosis of autism) it is perhaps unsurprising that the items loading onto them were both predicted and mixed. Seven items did not load onto any factor: 2, 10, 17, 20, 21, 35 and 40. Item 17, probed a local processing bias and was retained on theoretical grounds, the others were excluded from further analyses. Reliability analysis was performed to measure the consistency of the questionnaire (excluding items with communalities <.2 and non-loading items). The profiling items (N = 30) achieved a good Cronbach's  $\alpha$  ( $\alpha = .7$ ). Special skills items (N = 8) achieved an excellent Cronbach's  $\alpha$  $(\alpha = .932).$ 

# Traits Distinguishing ASD Individuals with and Without Reported Skills

The five factors and item 17 (the profiling data) were subsequently used as dependent variables (DVs) for 1-tailed *t* tests comparing the skilled to non-skilled group. We justified the use of 1-tailed tests on the grounds that we had a directional hypothesis for the likely association of special skills with increases in these DVs. Between group *t* tests adjusting for multiple comparisons using the Bonferroni method ( $\alpha = 0.05/6 = .0083$ ) were carried out and failed to reveal significant differences on three of the five

factors. These were socialisation and communication (t(114) = -.858, p > .05), repetitive behaviour and unusual interests (t(120) = 1.542, p > .05) and sensory sensitivity (t(113) = .421, p > .05). As item 17 is directly linked to the local information processing bias that has been implicated in savant and splinter skills in ASD, it was analysed as a separate item. However, this item did not significantly distinguish skilled (M = 4.42) from non-skilled individuals (M = 4.01) (t(119) = 1.235, p > .05). This suggests that several of the traits and characteristics, purportedly implicated in the emergence of talent in autism, did not distinguish between those with and without reported skills in the current sample.

The remaining two factors revealed a significant difference and a strong marginal trend towards a significant difference between the skilled and non-skilled groups and a more detailed analysis of these were carried out. Obsessions and special interest scores for skilled individuals (M = 27.19) were significantly higher than those for nonskilled individuals (M = 24.19) (t(115) = 2.614, p < .05). The analysis of the memory factor revealed a strong marginal trend towards significantly higher scores for the skilled group (M = 40.25) relative to the non-skilled group (M = 36.64) (t(113) = 1.961, p > .05).

The False Discovery Rate (FDR) method for correction of multiple comparisons was employed to explore the 13 items nestled within these two factors. This is less conservative than Bonferroni tests but still limits the rate of false discovery (Benjamini et al. 2001). The procedure described by Benjamini et al. (2001) was implemented. This revealed non-significant group differences on the first 8 items but significant differences on the remaining 5 items. As item distributions were not normal a non-parametric version of the t test was used. The p value was smaller than .05 for the ninth observed p (item 28) which is 0.004 (1-tailed). As 0.004 < 0.019 (5/13 × 0.05 = 0.019), item 28 was accepted as significant and all items with smaller p values were then automatically also significant (items 8, 25, 3, 23). Analysis of the obsessions and special interests factor showed that skilled individuals do not show increased levels of rigidity (item 12), obsessionality (item 32) or ritualistic behaviour (item 22) (indeed these items were non-significant), but they do become absorbed in topics that capture their interest (item 25). The analysis of the memory factor showed that skilled individuals are good at remembering dates (item 28), facts (item 3) and things that interest them (item 8). They also appear to show exceptional memory overall (item 23). Memory for places visited (item 33), personally experienced events (item 38), events experienced by others (item 13), or memory for any kind of (non-specific) information (item 18), did not distinguish skill positive and skill negative individuals.

In summary, the ascertainment study identified a number of factors that did and did not distinguish skilled individuals from their non-skilled counterparts, in the current sample. First, severity of social-communicative impairment, sensory abnormalities, obsessionality and a local information processing bias did not appear to differentiate the two groups. In contrast, exceptional general memory abilities, together with specific memory for facts, dates and things of interest were reported for the skilled individuals. Importantly, the questionnaire results indicate that special skills in ASD are not associated with rigid or obsessional tendencies. Instead these individuals show a tendency to become absorbed in topics of interest.

In order to test the preliminary results from the questionnaire piloting, detailed case studies were carried out on children with ASD and validated skills in the classic savant domains of music, art and math. The case studies investigated the factors included in the screening questionnaire and were tested using standardised tests.

## **Study 2: Validation**

## Method

## Participants

The subjects of the case studies were three boys aged between 10 and 11 years. Of these two had participated in previous studies in our lab and a third was identified after he took part in public event showcasing creativity and autism. None had taken part in our original screening study. Each child had a previous clinical diagnosis of ASD. This was confirmed at the time of testing using the Autism Diagnostic Observation Scale (ADOS; Lord et al. 1999) for two of the children. The third child had previously undergone ADOS assessment, by a clinical psychologist. The children were tested individually at their homes over a number of testing sessions and completed both skill validation tests and standardised tests of intelligence, memory and WCC. Psychometric data for the three participants are presented in Table 2.

### Skill Validation

In addition to exceptional drumming ability, D.B. was reported to possess absolute pitch ability (AP). While Profita and Bidder (1988) estimate that 1 in 10,000 TD individuals possess AP, Rimland and Fein (1988) suggest that as many as 1 in 20 individuals with autism possess AP. We presented D.B. with 25 randomly ordered complex tones spanning octaves C3–C5. D.B. rapidly named all tones achieving 100 % accuracy in note naming. J.R.'s Table 2 Participant details for the three case studies

	D.B.	J.R.	A.L.
Age (year, months)	11:10	10:3	11:0
Clinical diagnosis	Autism	Aspergers syndrome	ASD
ADOS total score (autism cut off = $10$ , ASD = $7$ )	12	9	7
Skill type	Music	Mathematics	Art
Onset of skill (months)	6	30	36
FSIQ	104	120	92
VCI	87	106	93
PRI	112	106	90
WMI	138	138	116
PSQ	88	121	83

mathematical skills had not been formally assessed before and were tested using standardised measures. He achieved a scaled score of 13 (IQ equivalent 130) on the Arithmetic subtest of the Wechsler Intelligence Scales for Children (WISC-IV; Wechsler 2004) and scaled scores of 148 (Numerical Operations) and 132 (Mathematical Reasoning) on the two maths subtests of the Wechsler Individual Achievement Test (WIAT-II: Wechsler 2005). His overall Mathematics composite score on the WIAT-II was very superior (149; 99.9th percentile). The third individual, A.L., produces tonal sketches that are well composed, realistic and detailed. During testing it was noted that he begins his drawings from local points and works from one detail to the next until his picture is complete. This style of graphic production has been described in a savant draftsman with autism (Mottron and Belleville 1993). Figure 1 is an example of a drawing produced by A.L.

### Results

The results from the questionnaire study failed to observe higher levels of symptom severity, obsessionality or restricted and repetitive behaviour in skilled ASD participants. The ADOS was completed with each of the children and their parents completed the lifetime version of the SCQ. ADOS Module 3 cut-off scores for J.R. and A.L. were 9 and 7, respectively (meeting ASD criteria) and 12 for D.B. (meeting autism criteria). The screening study had indicated that special skills were not limited to one PDD subtype and the case studies confirmed this. Scores of 0 were recorded for each child on the Stereotyped Behaviours and Restricted Interests component of the ADOS. This is in line with results from the questionnaire indicating that skilled individuals do not show increased levels of rigid behaviour relative to those without reported skills. In order to further measure obsessionality, 6 items were



Fig. 1 Example of a drawing produced by A.L.

drawn from the SCQ and the ADOS. These are shown in Table 3 below.

As can be seen from Table 3, the children do not appear to show exceptionally high levels of obsessional behaviour. With a total of 6 items, D.B. and A.L. showed signs of obsessional behaviour on two items (33 %) and J.R. on three items (50 %). The question of whether high-levels of focus, described in studies of talented TD children, provide a better explanation for special skills in autism will be further considered.

Exceptional memory has been highlighted in savants with autism and the results from the questionnaire study revealed a significant difference between skilled and non-skilled

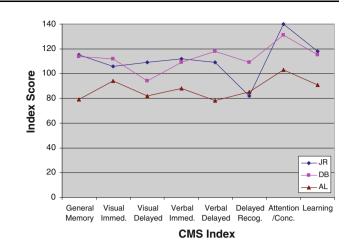


Fig. 2 Children's Memory Scale factor scores for the three cases

participants on this factor. The three children therefore completed a standardised battery of memory tasks (Children's Memory Scales; CMS, Cohen 1997). The results from this test are shown in Fig. 2.

As Fig. 2 illustrates, General Memory factor scores were in the high average range for J.R. and D.B. (115 and 114, respectively) and borderline for A.L. (79). This result, showing that none of the skilled children obtained scores in the superior or very superior range was inconsistent with the results from the questionnaire study. However, it was noted that all three children showed superior working memory. D.B. and J.R. achieved a score of 138 (very superior) on the Working Memory Index (WMI) of the WISC-IV and WMI was in the high average range (116) for A.L. (Table 2). Further it is worth noting that WMI was the highest composite score for each child on the WISC-IV. This peak was largely driven by very superior Digit Span subtest scores. A.L obtained a scaled score of 14 on this task and both D.B. and J.R. achieved scaled scores of 19.

Table 3 Obsessionality results for the three cases

SCQ/ADOS	Item	D.B.	A.L.	J.R.
SCQ	8. Has she/he ever had things she/he seemed to have to do in a very particular way or order, or rituals that s/he insisted you go through?	Ν	Y	Y
SCQ	11. Has she/he ever had any interests that preoccupy her/him and might seem odd to other people (e.g., traffic lights, drainpipes, timetables)?	Y	Ν	Y
SCQ	13. Has she/he ever had any special interests that were unusual in their intensity but otherwise appropriate for her/his age and peer group (e.g., trains, dinosaurs)?	Y	Y	Y
SCQ	18. Has he/she ever had any objects (other than a soft toy or comfort blanket) that s/he had to carry around?	Ν	Ν	Ν
ADOS	D4. Excessive interest in or references to unusual or highly specific topics or objects or repetitive behaviours	0	0	0
ADOS	D5. Compulsions or rituals	0	0	0
	Total	2/6	2/6	3/6

Table 4 Cognitive processing style results for the three cases

Test	D.B.	J.R.	A.L.		
CEFT					
Total score	25/25	21/25	22/25		
SD (above M)	1.4 <sup>a</sup>	0.84 <sup>b</sup>	1.2 <sup>a</sup>		
SCT	0 local endings	0 local endings	1 local ending		
Block design	13	10	9		
Picture completion	11	10	12		
Object assembly	7	11	11		

<sup>a</sup> Children 11–12 years (as stated in the test manual, Witkin et al. 1971)

<sup>b</sup> Children aged 9-10 years

The results of the questionnaire screen indicated that skilled individuals did not show increased levels of obsessional behaviour, rather they exhibited a tendency to become absorbed in topics of interest. This suggests a role for enhanced attention. A measure of attention for each child was taken using the Attention/Concentration factor of the CMS. J.R. and D.B. achieved very superior Attention/ Concentration factor scores of 140 and 131, respectively. For A.L. Attention/Concentration was average (103), yet it this was his highest factor score from a set of eight factors on this battery. Indeed all three skilled children obtained their highest scores on the CMS on this particular factor. Results showing high attention/concentration but not high levels of obsessionality are consistent with the findings from the ascertainment study and challenge current beliefs about the role of obsessionality in the emergence of savant talent.

Although a tendency to focus on parts of objects was not identified in the questionnaire study, a local processing bias has been strongly implicated in savant skills in autism and was therefore tested using a small battery of tasks traditionally used to assess WCC. This included three subtests from the Weschler Intelligence Scales (Block Design, Picture Completion and Object Assembly), the Children's Embedded Figures Test (CEFT; Witkin et al. 1971) and a Sentence Completion Task (SCT) developed by Booth and Happé (2010) (Table 4). For D.B. and A.L., performance on the CEFT was 1 SD higher than would be predicted on the basis of age norms detailed in the test manual. J.R.'s performance score was greater than the mean but not higher than would have been predicted for his age. None of the boys produced significant numbers of local endings on the SCT which suggested that language is processed in a global, not local, fashion. Results for the other three assessments were less congruent. Very superior Block Design performance was noted for one child only, the musician D.B. While J.R. and A.L. completed this task satisfactorily their scores were not higher than average. Picture Completion scores ranged between average and superior, Object Assembly scores ranged between borderline to high average. The results from the screening study failed to suggest increased local processing in skilled relative to non-skilled individuals, and the case study data broadly supports this finding.

# Discussion

Whilst a number of studies have tested cognitive style in savants relatively little is known about the diagnostic, behavioural and other features that differentiate savants and non-savants with autism. Thus, in addressing the need for a new validation methodology, the present study had two main aims. The first of these was to develop and pilot a new screening questionnaire for identifying cognitive characteristics and diagnostic features associated with special skills in ASD, and the second was to begin the process of validating the screening instrument in a series of case studies of talented children with ASD.

Piloted with 125 parents of ASD individuals, the questionnaire data revealed that 42 % of respondents had children with one or more skills that were outstanding given their child's functional skills. While questionnaire studies relying on parental/carer reports run the risk of under or over-estimating the prevalence and extent of special skills in autism, our new questionnaire asked respondents to record whether or not others outside of the family had commented on their child's special skill. For 9 out of 10 of the skilled cases this question elicited a positive response, providing a preliminary measure of skill validation in the absence of formal assessments. The estimate of 42 % is more than four times higher than Rimland's (1978) approximation of 9.8 %, and it may be the case that the low proportion of non-skilled individuals in our study resulted from an understandable reluctance, on the part of their parents, to complete our questionnaire. Whilst conclusions about prevalence rates cannot be made on the basis of the current pilot study we note that Howlin et al. (2009), reported a prevalence rate of approximately 30 % in their study and this does suggest that the prevalence of special skills in current populations of individuals with ASD is far higher than it was when Rimland carried out his study. There have been large increases in the numbers of intellectually unimpaired individuals receiving a diagnosis of ASD in recent years, and it is likely that the

population screened by Rimland included fewer intellectually higher functioning individuals. Our screening study indicated that only 5 % of the skilled group and 10 % of the non-skilled group were intellectually impaired and indeed the children in the case studies had Full-Scale IQ's of average or above. These results are consistent with findings by Howlin et al. (2009) showing that the average non-verbal IQ of their savant group was consistently higher than that of their non-savant group. These findings, showing that intellectual impairment is not universally associated with savant skills and that special skills may be more common amongst intellectually able individuals with autism, raises important questions about how savantism should be defined.

The analysis of the types of skills reported in the screening study is relevant to this question. Although highly idiosyncratic skills, such as the ability to identify objects by their smell, are widely reported in the literature on autism, such reports were extremely rare in the screening study and the large majority of skills described were of the type commonly observed in TD children. Theoretical accounts of savant syndrome assume that special skills in ASD are underpinned by different mechanisms to special skills in TD individuals. However, it is plausible to question whether or not good IT skills in a child with normal or superior intelligence and autism can be satisfactorily explained within a current theoretical account of savant syndrome. We believe that this question will be best addressed empirically in a future study comparing cognitive and behavioural profiles of children with and without autism and exceptional IT skills.

Our case studies were carried out on children with skills in the classic savant domains of number, drawing and music and an important aspect of the study was the validation of the skills described by the parents. Using a standardised test of mathematical ability we showed that J.R's skills are exceptionally high for a child of his age, in the top 99.9 %. The musician D.B. possesses exceptional drumming skills and has performed on national television. There are no current and appropriate assessment measures available for validating the type of drumming skills D.B. possesses. However, we were able to establish the existence of AP, a characteristic that is extremely rare in typical populations (See: Tacheuchi and Hulse 1993). It is interesting, given research showing variability in the extent of note naming skills in AP possessors without ASD, that D.B. achieved 100 % accuracy on the note naming task. While standardised validation assessments of artistic output do not currently exist, A.L.'s art work was sampled and we considered it to be of sufficiently high quality to meet criterion for our study. As these data make clear, our inability to support several commonly held assumptions about the characteristics of savants do not result from a lack of skill in our case studies.

The results from the screening study were important in highlighting a number of behavioural characteristics and traits associated with special skills in the current sample. Analysis of the questionnaire data revealed that talented individuals mostly possessed a single talent which had developed from an early age, had not been supported by formal training, and was not observed in other family members. Further, parents reported that the majority of skilled individuals (65 %) spent a good deal of time engaging in skill related activity, although they did not believe that this negatively impacted on their children's development in other areas. Traits that distinguished skilled from non-skilled individuals largely centred on memory characteristics and the presence of an intense interest.

The results from the ascertainment study suggesting that skilled individuals with ASD possess exceptional general memory skills were consistent with the view that exceptional memory abilities underpin savant skills (Treffert 1989, 2009). However, when memory abilities were directly assessed using a standardised battery of memory tasks the savant children in the case studies showed mostly average general memory abilities and in one case general memory skills verged between impaired and average (borderline). Interestingly, the artist A.L. did not show exceptional visual memory and this was not consistent with theorised associations between artistic talent and enhanced visual memory. Despite unremarkable scores on the memory battery, all three case study participants obtained high digit span scores and this was consistent with results from a number of studies showing peak performance on this test in savants (Spitz and LaFontaine 1973; Rimland and Hill 1984; Rumsey et al. 1992; Young & Nettelbeck 1995; Bölte and Pouka 2004). The potential role of domain general working memory abilities in the emergence of special skills in autism will be further discussed.

In addition to exceptional memory ability the parents who completed the questionnaire reported a tendency to become absorbed in topics of particular interest. However, this was not associated with increased sensory abnormalities, social-communicative impairments or restricted, repetitive behaviours. Whilst questions about the similarities and differences between ASD children with and without special skills can only be addressed in studies directly comparing the two groups, the results from the case studies also failed to reveal high levels of symptom severity, obsessionality, or repetitive behaviour. However, it was noted that all three scored in the definitely different range on the total score of the sensory profile and the existence of increased sensory difficulties in children with special skills cannot yet be ruled out. It was noted that ADOS scores for the three children were not unusually high and all three scored 0 for the item assessing stereotyped behaviour and repetitive interests. The analysis of the results from the six SCQ and ADOS items used to examine obsessionality also revealed low or moderate scores. In contrast, the scores on the measures assessing focused attention were striking. All three children obtained their highest memory battery scores on the attention factor and for two children these scores were in the very superior range. The measures of attention/concentration were unrelated to the children's areas of special interest in two out of three cases, and it may be the case that increased focus/concentration, like superior digit span, is a domain general characteristic of children with ASD and special skills. High scores on the attention/concentration factor are consistent with parental descriptions of highly focussed attention characterising skilled individuals in the ascertainment study and we are currently carrying out a new study comparing groups of skilled and non-skilled children with ASD on a test of obsessionality and on standardised measures of attention and concentration. Should the results from this study support those of the ascertainment and case studies our results will provide parallels with findings showing high levels of focus in TD children with special interests (DeLoache et al. 2007). This work may then motivate future exploration of commonalities between gifted individuals with and without disabilities.

Theoretical accounts of autism have implicated a local bias in the emergence of special skills in autism and this was tested in both the ascertainment and the validation studies. The results from the ascertainment study failed to show higher levels of local bias in the skilled compared to the non-skilled group; however, this was probed with a single item drawn from DSM-IV, concerning preoccupation with parts of objects. Given this, the decision to include a number of tests of WCC was taken. Five measures were used to determine whether or not the three savant children would demonstrate a strong local processing bias. All three children showed enhanced performance on the CEFT but the results from the other local processing tasks were mixed. Although the Block Design subtest has traditionally been considered to be a marker for WCC, only the musician obtained a very superior score on this test, whilst scores for the other two equally talented individuals were in the average range. None of the three children obtained peak performance on the Picture Completion or Object Assembly tests where a local processing style would convey an advantage. The results from the SCT yielded similar findings in that two participants failed to make any local endings and the third made one out of a possible ten local endings. This failure to confirm a local bias in individuals with a diagnosis of ASD and well validated skills is surprising given that such an association has been demonstrated in a substantial number of experimental studies (e.g., Mottron and Belleville 1993; Pring et al. 1995; Heaton et al. 1998; Heavey et al. 1999; Mottron et al. 2000a, b). It is often assumed that the CEFT and the Block Design task both tap a local bias (Shah and Frith 1983; Frith 1989), however Ropar and Mitchell (2001) have shown that scores on these two tests do not correlate with each other. In our case studies, Block Design scores were variable, and scores on the CEFT were uniformly high. Recent promising theories of savant syndrome implicate pattern recognition abilities (EPF; Mottron et al. 2009) and systematising (Baron-Cohen et al. 2009) and we are currently investigating associations between performance on the CEFT, the digit span test, and pattern detection tests in group studies of skilled and non-skilled children with ASD. If betweengroup differences in associations across these tests emerge in these studies, an important next step will be to replicate the studies with skilled and non-skilled TD children. These data will enable researchers to draw distinctions between factors associated with talent and factors associated with ASD and will be important to future debates about definitions of savant syndrome.

### Summary and Conclusions

In summary, the main aim of the current research was to develop methods for identifying differences between skilled and non-skilled individuals with ASD. A number of results emerged in the two studies. First, increased social-communicative impairments, repetitive behaviours, obsessionality and sensory abnormalities did not predict the existence of special skills, but skilled individuals did demonstrate a domain-general capacity for highly focused attention. The questionnaire study suggested that individuals with special skills possess outstanding general memory skills, but the case studies showed that this was largely limited to digit span, a measure of working memory. The results from the questionnaire and case studies failed to reveal a local information processing bias, although enhanced performance on the CEFT was observed in the case studies. Future modification of the questionnaire will be made in the light of the presented case studies and group studies are currently in progress. This current research has provided the first phase in this process.

# Appendix: Special Skills in Autism Questionnaire

Thank you for taking the time to complete this questionnaire. Please answer the following questions. Where there are given choices, please circle as appropriate.

- Today's date: \_\_\_\_
- Child's gender: \_
- Child's date of birth and age: \_\_\_\_\_\_
- Your relation to the child: Mother/Father/Other. If other, please specify: \_
- Are you the parent/guardian whom spends the most time with the child? YES/NO
- What is your child's formal diagnosis? Autism/Autism spectrum/High functioning autism/Aspergers syndrome/PDD/PDD not otherwise specified/MLD/SLD/developmental delay/other. If other please detail:
- Have any other family members ever been diagnosed with a psychological disorder or disability? YES/NO
- If YES, please specify the family member in relation to your child, their diagnosis, and when their diagnosis was made: \_\_\_\_\_\_

Please answer each statement below by putting a circle around the number that best reflects your degree of agreement or disagreement with that statement. There are no right or wrong answers. Rate each item from 1 (strongly disagree) to 7 (strongly agree), where 4 indicates that you neither agree nor disagree.

	SD			Ν			SA
1. My child is under sensitive to auditory stimuli	1	2	3	4	5	6	7
2. My child has had an object (other than a	1	2	3	4	5	6	7
blanket or soft toy) that he/she has had to carry							
around with him/her							
3. My child is good at remembering facts	1	2	3	4	5	6	7
4. My child does not seem able to understand the	1	2	3	4	5	6	7
thoughts and feelings of his/her peers							
5. My child is interested in a range of topics	1	2	3	4	5	6	7
6. My child is over sensitive to olfactory stimuli	1	2	3	4	5	6	7
7. My child has mannerisms or atypical ways of	1	2	3	4	5	6	7
moving his/her fingers (e.g., flapping, moving							
fingers in front of eyes)							
8. My child is good at remembering things that	1	2	3	4	5	6	7
interest him/her							
9. My child enjoys playing/interacting with	1	2	3	4	5	6	7
his/her peers							
10. My child has had an intense interest that was	1	2	3	4	5	6	7
appropriate for his/her age and peer group (e.g.,							
trains, dinosaurs)							
11. My child is over sensitive to visual stimuli	1	2	3	4	5	6	7
12. My child objects to changes in his/her routine	1	2	3	4	5	6	7
13. My child is good at remembering things that	1	2	3	4	5	6	7
happened to other people							
14. My child is good at expressing his/her own	1	2	3	4	5	6	7
feelings							
15. My child has a special interest or hobby but	1	2	3	4	5	6	7
he/she is not preoccupied with it							

	SD			N			SA
16. My child is over sensitive to auditory stimuli	1	2	3	4	5	6	7
17. My child seems more interested in parts of a	1	2	3	4	5	6	7
toy or object rather than whole objects							
18. My child is good at remembering any kind of	1	2	3	4	5	6	7
information							
19. My child is responsive to the initiations of	1	2	3	4	5	6	7
his/her peers to play/interact							
20. My child has had an interest that occupied a	1	2	3	4	5	6	7
great deal of his/her time							
21. My child is under sensitive to tactile stimuli	1	2	3	4	5	6	7
22. My child has had rituals that he/she insisted I	1	2	3	4	5	6	7
go through							
23. My child appears to show exceptional	1	2	3	4	5	6	7
memory							
24. My child prefers to do things on his/her own	1	2	3	4	5	6	7
rather than with peers							
25. My child has a special interest that is unusual	1	2	3	4	5	6	7
in intensity			-		-	-	
26. My child seems to be usually aware of the	1	2	3	4	5	6	7
sight, feel, sound, taste or smell of things or			-		-		
people							
27. My child engages in imaginative activity	1	2	3	4	5	6	7
28. My child is good at remembering dates	1	2	3	4	5	6	7
29. My child enjoys talking to his/her peers	1	2	3	4	5	6	7
30. My child has had a special interest that might	1	2	3	4	5	6	7
seem unusual to other people (e.g., drainpipes,	-	-	U	•	U.	Ū	
bus time-tables, traffic lights)							
31. My child is under sensitive to olfactory	1	2	3	4	5	6	7
stimuli	-	-	U	•	U U	Ũ	
32. My child has a tendency to be obsessional	1	2	3	4	5	6	7
33. My child is good at remembering things	1	2	3	4	5	6	7
about places that he/she has visited	-	-	U	•	U.	Ū	
34. My child initiates play/interaction with	1	2	3	4	5	6	7
his/her peers	1	-	0		U	Ū	,
35. My child's interests are focused on a narrow	1	2	3	4	5	6	7
range of topics	1	2	5	•	5	U	,
36. My child is under sensitive to visual stimuli	1	2	3	4	5	6	7
37. My child makes complex movements with	1	2	3	4	5	6	7
his/her whole body (e.g., repeatedly bouncing up	1	2	5	•	5	0	,
and down, spinning)							
38. My child is good at remembering things that	1	2	3	4	5	6	7
happened to him/her	*	-	5	•	5	0	,
39. My child is sensitive to the feelings of his/her	1	2	3	4	5	6	7
peers	-	_	2	•	2	5	,
40. My child has had an intense interest that was	1	2	3	4	5	6	7
not appropriate for his/her age group	*	-	5	•	5	0	,
41. My child is over sensitive to tactile stimuli	1	2	3	4	5	6	7
42. My child likes to order toys and/or objects	1	2	3	4	5	6	7
T2. IT y china nikes to order to ys and/or objects	1	4	5	Ŧ	5	U	/

Please answer these questions with regards to any outstanding skill that your child may <u>currently</u> have (that is, any marked ability that stands in contrast to his/her overall disability). Please answer each statement below by putting a circle around the number that best reflects your degree of agreement or disagreement with that statement. There are no right or wrong answers. Rate each item from 1 (Strongly Disagree) to 7 (Strongly Agree), where 4 indicates that you neither agree nor disagree. If your child <u>does not</u> currently have a skill that you consider to be outstanding, please leave this section blank and go to the next.

	SD	)		Ν			SA
1. My child currently has a skill that I consider to be outstanding against his/her usual pattern of everyday ability	1	2	3	4	5	6	7
Please detail if you circled 5, 6 or 7:							
2. My child currently has multiple skills that I consider to be outstanding against his/her usual pattern of everyday ability	1	2	3	4	5	6	7
Please list the skills:							
3. My child developed this skill(s) from an early age	1	2	3	4	5	6	7
Please specify at what age your child was when she/he developed t	he s	kill(s	s):				
4. Some people outside of the family have remarked on this skill(s) that my child has	1	2	3	4	5	6	7
5. My child has received formal training or tutoring to encourage this skill	1	2	3	4	5	6	7
If so please specify the type of training or tutoring and the duration	:						
6. My child spends a lot of time in activities related to this skill	1	2	3	4	5	6	7
7. Other family members possess a similar skill	1	2	3	4	5	6	7
If so, how much training has this family member had in this skill as	rea?	Plea	se sp	ecify	:		
8. My child has encountered training indirectly by bearing witness to this relative's own training	1	2	3	4	5	6	7
9. I think that this skill and the time my child currently spends on it interferes with my child's development	1	2	3	4	5	6	7

Please use this section to tell us anything further that you may wish to with regards to any current skill your child may have:

• What is the maximum level of education that you have received? (Please circle)

GCSE/A-level/Undergraduate degree/Postgraduate/Masters degree/Doctoral study/other If other, please specify:

• What is the maximum level of education that your child's other parent has received? (Please circle)

GCSE/A-level/Undergraduate degree/Postgraduate/Masters degree/Doctoral study/other If other, please specify:

• Is your household yearly income:

 Between £0 and £15,000
 YES/NO

 Between £15,000 and £25,000
 YES/NO

 Between £25,000 and £35,000
 YES/NO

 Between £35,000 and £45,000
 YES/NO

 £45,000 plus
 YES/NO

- Please specify your current occupation: \_

Are you willing to be contacted again for future follow -up research purposes: YES/NO

If yes, please provide your details here:

- Your name: \_\_\_\_\_
- Your telephone number: \_\_\_\_\_\_
- Your email address: \_

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