Defining the clinical and cognitive phenotype of child savants with autism spectrum disorder.

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Abstract

Objective: Whilst savant syndrome is most commonly observed in individuals with Autism Spectrum Disorder (ASD), it has historically been associated with intellectual impairment, and little is known about the clinical and cognitive characteristics of intellectually able individuals with ASD and savant skills.

Methods: Participants with ASD and validated savant skills were compared with age and intelligence matched non-savants with ASD using a range of diagnostic and standardised tests.

Results: Although the analysis of the clinical data revealed few differences between the groups, striking differences emerged during cognitive testing. Children with savant skills exhibited highly superior working memory and their scores on tests of analytic skills were also superior to those of non-savants.

Conclusion: We propose that obsessionality, focused attention, superior working memory and analytic skills facilitate veridical mapping and pattern perception abilities characteristic in savant syndrome.

Keywords: Intellectual disability, Savant syndrome, Children.

Accepted January 30, 2017
to routines, they did show that savants with ASD were more likely to order their possessions and show an increased interest in one particular topic than savants without ASD and ASD non-savant controls. In more recent studies comparing savants and non-savants with ASD on clinical measures, Howlin et al. [16] failed to observe increased levels of repetitive behaviour in savants et al. [17] failed to confirm increased levels of rigidity, obsessionality or ritualistic behaviours in savants. However, consistent with the results from the O’Connor et al. [15] study, the carers who completed Bennett & Heaton’s questionnaire reported an increased tendency to become absorbed in topics of interest and when this was directly tested in a series of case studies a superior and domain-general capacity to focus attention was observed in the savant children.

Whilst little is known about the clinical and behavioural correlates of savant syndrome, its cognitive correlates are better understood. For example, Bölte et al. [18] compared the intelligence test profiles of child and adult savants and non-savants with ASD on the Wechsler Intelligence Scales for children or adults (WISC/WAIS) and showed that the only test to statistically differentiate savants from non-savants was the Digit Span subtest. Whilst the study of calendar calculating savants carried out by Heavey et al. [19] failed to reveal superior Digit Span scores, studies from other research groups [17,20-22] have replicated Bölte et al. [18] finding. As research with typical participants has shown that working memory is a strong predictor of reasoning ability, intelligence and attentional control, the suggestion that savants possess superior working memory clearly merits further investigation [23-25]. The Veridical Mapping model of savant syndrome proposes that an enhanced ability to map isomorphic perceptual and non-perceptual information predisposes outstanding pattern detection and it is plausible to suggest that superior rehearsal and storage capacity will support this process. In addition to identifying high working memory capacity, a number of studies have revealed superior performance on the Block Design subtest from the Wechsler Intelligence Scales in samples of individuals with savant talents [5,18,22,26]. This test has traditionally been considered as a measure of local processing capacity and the idea that a local bias and/or enhanced perceptual processing facilitates access to domain-specific materials (e.g. numbers, musical tones) that can then be hierarchically mapped and organised is an important cornerstone of current models of savant syndrome [27-29].

Whilst savant syndrome is closely associated with ASD, many or most individuals with this disorder are not savants [1,4,5]. Therefore the main objective of the study was to address outstanding questions about how savants and non-savants with ASD differ when tested using standardised clinical and cognitive measures. The rationale for the clinical assessments was drawn from claimed links between the clinical symptoms of ASD and savant skills [11-14]. As recent questionnaire and case study data suggests that savants may exhibit highly focused attention this was also measured in the study [19]. The rationale for the cognitive assessments was drawn from studies raising questions about the role of working memory, Block Design performance and fluid intelligence, in the emergence of savant talent. Research suggests that savants may constitute a distinct genetic/behavioural subgroup within the autism spectrum and the final aim of the study was to provide data that informs our understanding of the heterogeneity characterising ASD [30].

Methods

Participants

A questionnaire was initially used to identify and recruit savant and non-savant participants to the study. This questionnaire drew questions from the special isolated skills section from the Autism Diagnostic Interview–Revised and was modelled on the savant screening questionnaire devised by Bennett et al. [17,31]. An initial sample of 43 children, recruited via UK based NAS branches, parent group forums and by word of mouth participated in the skill validation phase of the study. Of this group 7 were unsuitable for the study. Four were unable to complete testing, two obtained high average mathematics scores and did not meet criteria for either experimental group and the third (a female) was excluded on the basis of gender (all validated savants were male). This resulted in a sample of 17 savant and 19 non-savant males, aged between 8 years and 12 years 9 months with a prior clinical diagnosis of ASD (savants M=123.79 months, SD=16.28; non-savants M=123.79 months, SD=16.70). Whilst the focus of the study was a comparison of savant and non-savant children with ASD, comparison data from 17 Typically Developing (TD) children were included in the assessment of obsessional traits. These children were attending mainstream schools in south London and were recruited via word of mouth. They were matched to the total ASD group for chronological age (ASD group M=124.53 months, SD=16.28; TD group M=123.59 months, SD=19.28) and non-verbal intelligence (Raven’s Standard Progressive Matrices raw scores: ASD group M=41.36, SD=9.55; TD group M=45.24, SD=5.9) [32]. The TD children were screened for ASD traits using the Autism Spectrum Quotient questionnaires and all children scored below the cut-off of 76 for the AQ–Child (M=42.79, SD=8.39) or 32 for the AQ–Adolescent (M=7.33, SD=2.31) [33,34].

Measures

Skills validation assessments

Skill validation assessments operationalised Treffert et al. [1] criteria for talented savant status. The participants screened for inclusion in the savant group presented with reported skills in mathematics (10), absolute pitch (3), calendar calculating (2) and art (2). Mathematics skills were formally assessed using the Numerical Operations and
Mathematical Reasoning subtests from the second edition of the Wechsler Individual Achievement Test [35]. This standardised test yields an overall Mathematics composite which can then be compared to norms for TD individuals of the same age. All mathematic savants obtained scores in the superior/very superior range on this test. Absolute pitch was confirmed using a pitch series naming task, previously used by Heaton et al. [36]. Calendar calculating skills were assessed with date questions and revealed a calculating span of up to 4 years forward/backward for one savant and a span of 100 years forward/backward for the second. Following the assessment method detailed by Hermelin et al. [37], a professional artist, blind to the participants disability, assessed art work for 1) liveliness of and sensitivity to the object/subject drawn, 2) vitality and the character of line and texture, 3) presence of a distinct personal style, 4) organisation and composition of the piece and 5) the degree to which a compelling and interesting image had been produced. While Hermelin et al. [15] employed a rating scale covering grades from A+ to E− (i.e., 15 points), the criteria were simplified for the purposes of the current study and the rating scale covered grades A to E (5 points). The two artists were awarded grades of A or B for each criterion for the examples of art work produced by savants and shown in Figures 1 and 2.

Clinical Assessments

Symptom severity: The lifetime form of the Social Communication Questionnaire was completed by parents/caregivers [38]. It is a 40 item instrument that screens for symptoms of ASD. It shows high levels of agreement with ADI–R. For the lifetime form of the SCQ, a score of ≥ 22 indicates a developmental history of autism, while ≥ 15 indicates a history of Pervasive Developmental Disorder (PDD).

The Autism Diagnostic Observation Schedule is a comprehensive, standardised assessment, administered by a trained user that assesses the social, communication and other disabilities characterising ASD [31]. ADOS can be used in research to confirm a prior clinical diagnosis and to provide some measure of social and communication deficits. It includes four modules and module 3, designed for use with verbally fluent children/adolescents, was administered to all ASD participants in the study.

The Short Sensory Profile is a 38 item parent/caregiver questionnaire that probes the frequency, between (1) always and (5) never, of abnormal sensory responses across seven core areas: tactile sensitivity, taste/smell sensitivity, movement sensitivity, under-responsiveness/seeks sensation, auditory filtering, low energy/weak and visual/auditory sensitivity [39]. The test yields a total score and factor scores for each sensory domain, with responses categorised as typical, probable difference (from typical) and definite difference (from typical).
**Obsessionality:** Parents/caregivers also completed an adapted version of the Cambridge University Obsessions Questionnaire [40]. This questionnaire probes twenty categories across six core cognitive domains (physics, mathematics, biology, psychology, language and taxonomy), eight areas of everyday life (e.g. attachments to objects) and one autism-specific clinical domain (sensory phenomena). The original questionnaire was modified in three ways. First, a category termed ‘other’ for obsessions not captured in the other 19 categories was replaced with a new ‘spatial information’ category. Second, obsessionality was specifically defined as “any idea that haunts hovers and constantly invades one’s consciousness” [41]. Finally, respondents were asked to specify the extent of their child’s obsessions by circling either slightly, moderately or highly obsessed. A key was provided to explain the differences in intensity at each of these levels. At the least intense level, slight obsessions were defined as preoccupations that seem quite mild, are greater than an interest but do not interfere with thinking about or doing other things. Moderately intense obsessions were described as preoccupations that were not all encompassing. Highly intense obsessions were defined as all encompassing so that it was extremely difficult to switch attention from the object of the obsession to something else. Where a parent reported that their child had one or more specific obsessions in a particular category, a score of 1 was recorded (0 for no obsession) and scale of 1–3 was applied to the ratings of slightly (1), moderately (2) and highly obsessed (3). The total number of 1’s, 2’s and 3’s were then recorded for each participant and percentage scores were calculated.

**Cognitive Ability Assessments**

**Focussed attention:** The Attention/Concentration factor from the Children’s Memory Scales consists of two core subtests (Numbers, Sequences) and one supplemental subtest (Picture Locations) [42]. Numbers assesses the ability to repeat sequences of numbers that increase in length and Sequences assesses the ability to mentally order verbal information at speed. Picture Locations assesses immediate visual memory for the spatial locations of pictured objects.

**Intelligence:** The Raven’s Standard Progressive Matrices is a test of fluid intelligence that assesses the ability to indentify items which complete patterns presented in $2 \times 2$, $3 \times 3$ or $4 \times 4$ matrices [32]. The problems within each set of matrices become increasingly difficult and require ever greater cognitive capacity to encode and analyse pattern information.

The fourth edition of the Wechsler Intelligence Scales for Children yields five composite scores [43]. In addition to full scale IQ (FSIQ), four composites representing cognitive abilities in more specific domains are obtained: Verbal Comprehension Index (VCI), Perceptual Reasoning Index (PRI), Working Memory Index (WMI) and Processing Speed Quotient (PSQ). Ten core subtests are divided amongst the four indices.

**Procedure**

Tests were administered in the participants’ homes over a maximum of four testing sessions. Standardised instructions were adhered to and participants took breaks as required. The study adhered to British Psychological Society guidelines and approval for the study was granted by the ethics committee at Goldsmiths University of London.

**Results**

**Clinical Assessments**

**Symptom severity:** The means and standard deviations for savants and non-savants on measures of symptom severity are presented in Table 1.

The groups did not differ on total SCQ ($t(34)=0.58$, $p>0.05$) or total ADOS scores ($t(34)=1.52$, $p>0.05$). However, whilst the groups also failed to differ on the stereotyped behaviours and restricted interests subset from the ADOS ($t(34)=0.042$, $p>0.05$), there was a significant difference on the Imagination/Creativity subset ($t(34)=-2.06$, $p<0.05$) with lower levels of impairment in the savant group.

The means, standard deviations and classifications for savant and non-savant groups on the Short Sensory Profile are presented in Table 2.

<table>
<thead>
<tr>
<th>Measure of Symptom Severity</th>
<th>Savants M (SD)</th>
<th>Non-Savants M (SD)</th>
</tr>
</thead>
<tbody>
<tr>
<td>SCQ Total (autism cut-off=22, PDD=15)</td>
<td>18.65 (7.78)</td>
<td>20.05 (6.76)</td>
</tr>
<tr>
<td>ADOS Total (autism cut-off=10, ASD=7)</td>
<td>8.65 (3.30)</td>
<td>6.95 (3.34)</td>
</tr>
<tr>
<td>ADOS Communication (autism cut-off=4, ASD=2)</td>
<td>2.53 (1.46)</td>
<td>2.00 (1.15)</td>
</tr>
<tr>
<td>ADOS Reciprocal Social Interaction (autism cut-off=6, ASD=4)</td>
<td>6.12 (2.15)</td>
<td>4.95 (2.48)</td>
</tr>
<tr>
<td>ADOS Stereotyped Behaviours and Restricted Interests</td>
<td>0.59 (0.71)</td>
<td>0.58 (0.61)</td>
</tr>
<tr>
<td>ADOS Imagination/Creativity</td>
<td>0.47 (0.51)</td>
<td>0.95 (0.85)</td>
</tr>
</tbody>
</table>
Whilst the savants recorded milder sensory symptoms than non-savants, the group difference narrowly failed to reach statistical significance, $t(34)=2.69$, $p=0.01$.

As the auditory filtering and visual/auditory sensitivity scales measure sensory domains implicated in savant skills (music and art), two tailed t-tests with adjustments for multiple corrections were carried out ($p<0.025$). These showed reduced impairments in savants on auditory filtering ($t(34)=2.63$, $p<0.025$) and there was a marginal trend for reduced impairment in the visual/auditory subscales ($t(34)=2.13$, $p=0.04$).

**Obsessionality:** A one-way ANOVA comparing total obsession scores for the savant, non-savant and TD groups revealed a significant main effect, $F(2,50)=18.86$, $p<0.05$. Post hoc comparisons using the Tukey HSD test showed that whilst the number of obsessions recorded for the savant ($M=7.94$, $SD=4.02$) and non-savant group ($M=7.42$, $SD=2.32$) did not differ, the TD comparison group reported significantly fewer obsessions ($M=2.06$, $SD=2.84$) than both ASD groups ($p=0.01$). Intensity ratings for the two ASD groups were calculated and failed to reveal marked differences across the groups. Thirty-five percent of savants and 34% of non-savants reported slight obsessions, 30% of savants and 41% of non-savants reported moderate obsessions and 30% of savants and 25% of non-savants reported strong obsessions. The profile of obsessions (e.g. in the different categories) failed to show consistent and different patterns across groups.

**Cognitive Ability Assessments**

**Focussed attention:** The analysis of the composite scores on the Attention/Concentration factor from the Children’s Memory Scale revealed a highly significant difference between groups: scores were in the superior range for the savant group ($M=129.29$, $SD=12.39$) compared with average scores for the non-savant group ($M=108.32$, $SD=18.48$) ($t(34)=4.04$, $p<0.05$). Independent samples t-tests with corrections for multiple comparisons ($p=0.05/3$; $p≤0.02$) were carried out on the three Attention/Concentration subscales and revealed significantly higher scores for savants on the Numbers subtest (savants $M=17.35$, $SD=2.09$; non-savants $M=13.42$, $SD=3.20$; $t(34)=4.31$, $p<0.02$), the Sequences subtest (savants $M=12.29$, $SD=2.08$; non-savants $M=9.32$, $SD=3.42$; $t(34)=3.19$, $p<0.02$) and the Picture Locations subtest (savants $M=12.94$, $SD=2.05$; non-savants $M=10.53$, $SD=2.89$; $t(34)=2.91$, $p<0.02$).

**Intelligence:** The analysis of RSPM raw scores revealed a significant group difference, with higher scores in the savant group than the non-savant group (savants $M=45.18$, $SD=8.87$; non-savants $M=37.85$, $SD=9.03$; $t(34)=2.42$, $p<0.05$). Scores from the WISC–IV are shown in Figure 3.

An independent samples t-test showed that savants and non-savants did not significantly differ on full scale IQ scores (savants $M=112.35$, $SD=19.13$; non-savants $M=102.95$, $SD=13.90$; $t(34)=1.70$, $p>0.05$). Further independent samples t-tests with corrections for multiple comparisons ($p=0.05/4$; $p≤0.01$) showed that the groups did not differ on verbal IQ (savants $M=101.29$, $SD=17.78$; non-savants $M=97.26$, $SD=9.20$; $t(34)=0.840$, $p>0.01$), performance IQ (savants $M=115.12$, $SD=20.82$; non-savants $M=104.95$, $SD=12.70$; $t(34)=1.75$, $p>0.01$) or processing speed IQ (savants $M=96.94$, $SD=14.72$; non-savants $M=98.84$, $SD=17.27$; $t(34)=0.353$, $p>0.01$). However, savants obtained significantly higher scores than non-savants on the working memory composite of IQ (savants $M=126.76$, $SD=15.87$; non-savants $M=107.63$, $SD=13.90$; $t(34)=3.86$, $p<0.01$).

Independent samples t-tests with corrections for multiple comparisons ($p=0.05/2$; $p≤0.025$) were used to examine performance on the two working memory subtests. This revealed superior savant performance on the Digit Span subtest (savants $M=16.18$, $SD=2.96$; non-savants $M=11.47$, $SD=2.57$; $t(34)=5.10$, $p<0.025$) but not on Letter-Number Sequencing subtest (savants $M=13.12$, $SD=3.30$; non-savants $M=11.42$, $SD=3.04$; $t(34)=1.61$, $p>0.025$). A further two t-tests with corrections for multiple comparisons ($p=0.05/2$; $p≤0.025$) were carried out on the Digit Span data. Savants scored significantly higher than non-savants on Digit Span Forwards (savants $M=14.88$, $SD=2.32$; non-savants $M=12.37$, $SD=2.69$; $t(34)=2.99$, $p<0.025$). Savants also scored significantly higher than non-savants on Digit Span Backwards (savants $M=15.06$, $SD=3.42$; non-savants $M=12.79$, $SD=3.32$; $t(34)=2.94$, $p<0.025$).
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As theoretical accounts of savants have implicated superior Block Design and Matrix Reasoning skills, data for these subtests were also compared using independent samples t-tests with corrections for multiple comparisons ($p=0.05/2$; $p \leq 0.025$). These revealed a savant superiority on both Block Design ($M=12.59$, $SD=3.71$; non-savants $M=10.05$, $SD=2.55$; $t(34)=2.41$, $p<0.025$) and Matrix Reasoning ($M=12.47$, $SD=4.00$; non-savants $M=10.00$, $SD=3.06$; $t(34)=2.10$, $p<0.025$) tests.

Within-group correlations were carried out on the data from the cognitive tests that distinguished the groups. For the savant group the correlation between RSPM and Block Design scores was highly significant ($r=0.563$, $p>0.01$). Non-significant correlations were observed for RSPM and Digit Span scores ($r=0.146$) and Block Design and Digit Span scores ($r=0.010$). For the non-savants, none of the correlations reached significance (RSPM and Block Design, $r=0.297$; RSPM and Digit Span, $r=0.226$; Block Design and Digit Span, $r=0.036$).

**Discussion**

The main aim of the study was to compare clinical and cognitive profiles of child savants and non-savants with ASD. According to Treffert [1] talented savant status depends upon an interpersonal discrepancy across abilities and whilst the individuals in the savant group obtained intelligence test scores that were mostly in the normal range, significant peaks in the domains of maths, calendar calculation, music and art were validated using either standardised tests or pre-existing procedures. The first set of comparisons aimed to identify any behavioural differences distinguishing savants and non-savants with ASD. It was specifically predicted that increased engagement in talent-related activity would be associated with increased social and communication deficits manifested in higher scores on measures of symptom severity from SCQ and ADOS. However, when savants and non-savants were compared, group differences failed to emerge on the measures of socialisation or communication. The only group difference observed was on the imagination factor from the ADOS, where the savants appeared to show lower levels of impairment. Whilst clinical definitions and measures of creativity differ from those typically used to describe abilities in artistic and academic domains, previous work has shown increases in aspects of creativity in savants with ASD and this finding merits further investigation. Small group differences were also observed on the sensory profile and it appeared that the savants experienced less sensory disturbance within specific modalities, related to skill development, compared to non-savants [44]. Obsessionality has been associated with savant syndrome in the literature and was tested using an adapted version of a questionnaire developed by Baron-Cohen and Wheelwright [40]. However, the results from this comparison failed to reveal any difference between the ASD groups. Taken together, the results from the clinical assessments failed to reveal marked differences between the groups and it did not appear that the clinical characteristics of ASD were sufficient to explain savant skills.

In comparison to the clinical assessments, striking differences between savant and non-savant groups emerged on measures of cognitive ability. Whilst groups did not differ on full scale IQ, qualitative differences in cognitive profiles across the different assessments were observed. The first difference to distinguish groups was on the measure used to test focused attention. Our previous screening study had highlighted focussed attention as a factor distinguishing savants from non-savants and when we tested this using the Attention/Concentration factor from the Children’s Memory Scales we observed superior performance, manifested in an outstanding capacity for...
repeating numbers, ordering verbal information at speed and remembering the spatial locations of visually presented objects in savant participants [17]. A related finding emerged in the comparison of the subtest profile analysis of the WISC–IV, where a significant difference between ASD groups emerged on both forward and backward components of the Digit Span test. Whilst deficits in working memory have been reported in individuals with ASD [45] the non-savants in the present study obtained average scaled scores for the Digit Span test. However, savants obtained a group mean scaled score of 16, which, when compared with norms derived from age-matched typical children, is very superior. As savants performed equally well on backward and forward Digit Span tests, these results cannot reflect rote memory, and together with the results from the Children’s Memory Scale suggests that savant syndrome is characterised by superior working memory capacity.

In addition to measures of working memory, savant superiority was observed on the Block Design and Raven’s Matrices tests. Whilst good Block Design performance in ASD has traditionally been taken as a marker for a local processing bias, some studies of savants have failed to observe a cognitive peak on this task [17]. The strong association between performance on the Block Design and Raven’s Matrices tests for savants in the current study suggests that it might instead reflect superior analytic skills or fluid intelligence. Dawson et al. [7,8] have suggested that Raven’s Matrices measures a mechanism that is important for all aspects of cognition in ASD, and our results, showing superior performance on this test in savants, is consistent with this. Whilst savants showed striking performance on tests measuring working memory and analytic skills, scores for these tests did not correlate and we suggest that these are independent processes that work in tandem to facilitate pattern recognition and encoding within talent domains. The Veridical Mapping model of savant skills describes a unique and active learning process and our results provide new information on the cognitive mechanisms implicated in this [25].

Early descriptions of savants report severe and global intellectual impairments. However they pre-date Kanner’s [46] identification of autism, and their authors may have failed to distinguish between social/communication and intellectual deficits in the savants they described. Recent definitions of savant syndrome have de-emphasised the importance of intellectual impairment and our results highlight the value of considering strengths in assessing achievements in ASD. Whilst the savants in our study were not more obsessionally than non-savants, their scores on this measure were significantly higher than those of typical children. We therefore propose that this trait, together with highly focused attention and exceptional analytic and memory abilities, facilitates the veridical mapping and pattern perception abilities characterising savant syndrome.

Acknowledgement

We wish to thank our participants and their parents/carers for their time and support. Thanks also to Michelle Dawson for pointing out that the earliest reported use of the term idiot-savant was by Edouard Seguin in 1866 and not Langdon Down in 1887.

References

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