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Is Synaesthesia More Prevalent in Autism Spectrum Conditions? Only Where There Is Prodigious Talent

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Abstract

Savant syndrome is a condition where prodigious talent co-occurs with developmental difficulties such as autism spectrum conditions (ASC). To better understand savant skills, we previously proposed a link with synaesthesia: that savant syndrome may arise in ASC individuals who also happen to have synaesthesia. A second, unrelated claim is that people with autism may have higher rates of synaesthesia. Here we ask whether synaesthesia is indeed found more often in autism *per se*, or only in cases where autism co-occurs with savant skills. People with autism in previous studies when tested for synaesthesia were not differentiated into those with and without savant abilities. Here we tested three groups: people with autism who also have savant skills ($n = 40$), people with autism without savant skills ($n = 34$), and controls without autism ($n = 29$). We used a validated test to diagnose grapheme–colour synaesthesia. Results show a significantly higher prevalence of synaesthesia in people with ASC, but only those who also have savant skills. This suggests that synaesthesia in autism is linked to those with savant abilities rather than autism *per se*. We discuss the role of synaesthesia in the development of prodigious talent.

Keywords

Synaesthesia, savant syndrome, talent, autism, perception, sensory

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1. Introduction

Synaesthesia is a condition in which certain kinds of stimuli trigger unusual, automatic and involuntary additional experiences. For example, *sound–colour synaesthetes* 'see' colours when they hear particular sounds, tones, or timbres (Ward *et al.*, 2006). Here we use the conventional term *inducer* to refer to the stimulus that triggers the synaesthesia (e.g., music) and the term *concurrent* to refer to the synaesthetic sensation (e.g., colours) (Grossenbacher and Lovelace, 2001). One of the most widely studied types of synaesthesia is *grapheme–colour synaesthesia*, in which the concurrent of colour occurs as a result of reading, hearing, or just thinking about letters or numbers (Simner *et al.*, 2006a, b). In the present study we test the relationship between grapheme–colour synaesthesia and autism spectrum conditions (ASC). We ask whether the two conditions significantly co-occur and if so, whether this is related to the emergence of prodigious talent.

ASC are characterised by difficulties in social communication alongside unusually narrow interests, repetitive behaviour, and a strong need for routines, as well as sensory hyper-sensitivity (American Psychiatric Association, 2013). Previous studies have suggested that people with ASC might also have elevated rates of synaesthesia, and a link between these two conditions has been made at several levels. First, studies have suggested a possible genetic and phenotypic overlap between the two conditions (Asher *et al.*, 2009; Cytowic, 1995). Genetically, one region of chromosome 2 implicated in synaesthesia (2q24.1; Asher *et al.*, 2009) has also been found in genome-wide studies of ASC (IMGSAC, 2001). This region contains several hundred genes, so this potential overlap in genetic architecture requires further study. Gregerson *et al.* (2013) also show that a close genetic relationship exists between synaesthesia and absolute pitch (AP), and AP occurs more often in people with ASC (DePape *et al.*, 2012; Dohn *et al.*, 2012). However, there is a large degree of genetic heterogeneity in the development of synaesthesia, with potential links to many other conditions. The genetic bases of synaesthesia and ASC are still not well understood, so it is premature to suggest a definitive genetic link between these two conditions.

Behaviourally, both conditions are characterised by unusual sensory experiences (Baron-Cohen *et al.*, 2009; Marco *et al.*, 2011; Neufeld *et al.*, 2013; Rogers and Ozonoff, 2005; Tavassoli *et al.*, 2014a, b). Sensory hyposensitivities and hyper-sensitivities are frequently found in ASC, with individuals reporting difficulties adjusting to changes in lighting conditions, sounds, smells, tactile stimulation *etc.* (Leekam *et al.*, 2007; Tomcheck and Dunn, 2007). In synaesthesia too, Banissy *et al.* (2009) found a relationship between the modality of synaesthetic experiences (e.g., sensations of colour) and sensory hypersensitivity in those same modalities (e.g., enhanced colour

perception). Both synaesthetes and individuals with ASC self-report increased sensory sensitivity across several sensory domains compared to controls (Ward *et al.*, 2017). Thus, atypical sensory experiences occur in both synaesthesia and autism, although whether this supports a direct causal explanation for any co-occurrence of these two conditions remains to be clarified.

Finally, there is some similarity across both conditions in their neural bases. Kemner *et al.* (1995) conducted an ERP study and found that people with autism showed what was described as 'synaesthetic-like' brain activity, with occipital activation (usually associated with visual processing) in response to auditory stimuli. Specifically, Kemner *et al.* identified a task effect that was unique to individuals with ASC (and not controls) involving significantly increased occipital activation across two auditory tasks. Jao Keehn *et al.* (2016) replicated this finding using fMRI, demonstrating increased activity in the visual cortex of participants with ASC during an auditory task compared to reduced activity in a control group under the same conditions. Neuroimaging thus demonstrates at least some functional similarities in the brains of synaesthetes and individuals with ASC. Structurally, both synaesthesia and ASC have been linked to altered neural connectivity, although this is also true of many conditions, including schizophrenia (McIntosh *et al.*, 2008). Differences in structural connectivity have been found in the brains of synaesthetes both globally (Zamm *et al.*, 2013) and in terms of local connectivity between adjacent brain regions (Bargary and Mitchell, 2008). For example, Rouw and Scholte (2007) showed that grapheme–colour synaesthetes had local clusters of greater anisotropic diffusion (associated with more coherent white matter) near colour-selective regions compared with matched controls. In ASC too, Casanova and Trippe (2009) suggest that some degree of hyper-connectivity in the brains of individuals with autism leads to the formation of short-range local connections in a similar way that local cross-activation in adjacent cortical areas may underlie synaesthetic experiences (Hubbard and Ramachandran, 2005).

Relevant to the current study is that associations between synaesthesia and ASC have also been suggested from quasi-epidemiological studies, comparing the two conditions directly. These studies were originally founded on case-reports of individuals showing both conditions (Baron-Cohen *et al.*, 2007; Bor *et al.*, 2007) although it is difficult to conclude from case studies whether the two conditions are linked causally or by chance alone. However, Baron-Cohen *et al.* (2013) tested 164 individuals with ASC along with 97 typical controls, asking them to self-report whether they had synaesthesia. Participants were asked to report not only grapheme–colour synaesthesia but a range of other variants (e.g., sound–colour, taste–colour, touch–colour, taste–shape, sound–taste). Across all these variants, synaesthesia was reported by 18.9% of individuals with ASC, and this was significantly higher than the 7.2% reported

in the control sample. Within this, grapheme–colour synaesthesia specifically was reported by 11.0% (18 out of 164) of individuals with ASC, compared to 3.1% (3 out of 97) of controls. Although no statistical comparison was made in that paper, we calculate here that this would be a significant difference across groups in the number of self-reported grapheme–colour synaesthetes [$\chi^2(1, 1) = 4.109$ with Yates' correction; $p \leqslant 0.05$). However, in that study there was no validation test to independently verify self-reports of synaesthesia, due to low participant uptake in a subsequent 'test of genuineness'. This is important because self-report alone can be unreliable in the diagnosis of synaesthesia (Simner *et al.*, 2006b).

However, these findings have been replicated in a second study (Neufeld *et al.*, 2013) that showed elevated rates of synaesthesia in people with ASC using both self-report and an objective test for synaesthesia. Neufeld *et al.* screened 29 individuals with Asperger Syndrome for grapheme–colour synaesthesia using a validated test of genuineness (described below). Although their sample was relatively small, they found the rate of synaesthesia was almost nine times higher in people with ASC (17.2%) than might be expected in the general population for which they used a baseline of 2.0%, taken from a study of the prevalence of grapheme–colour synaesthesia in the general population (Simner *et al.*, 2006b). From these studies we conclude that grapheme–colour synaesthesia occurs significantly more often in ASC compared to the general population. In the current study we ask whether grapheme–colour synaesthesia occurs more often in ASC, or whether it occurs particularly in a subset of individuals with ASC, namely, those who also have savant syndrome.

Savant syndrome is characterised by the presence of specific talents in individuals with a developmental condition such as autism (Howlin *et al.*, 2009; Treffert, 2009), where the talents exceed the individual's overall level of intellectual or developmental functioning. For example, an individual with autism might have a talent in drawing realistic portraits, despite having social communication or learning disability. Other savant skills are related to memory, mathematics, art or music. These skills have been described as 'islands of genius' since they exist in individuals with deficits in other domains (Treffert, 2009). Prodigious savant abilities are defined as those that occur in individuals who possess skills that are not only striking when compared to their own level of overall functioning, but also are outstanding in comparison to the general population.

Savant syndrome has been reported to occur in up to 37% of individuals with ASC (Howlin *et al.*, 2009) while as many as 50% of individuals with a savant skill are diagnosed with ASC (Chia, 2012). As well as being tied to ASC or related neurodevelopmental conditions, it has also been hypothesised that savant syndrome may be linked to synaesthesia (Baron-Cohen *et al.*, 2007; Simner *et al.*, 2009). In this body of research, we have suggested that the

combination of ASC and synaesthesia, co-occurring within a single individual, might provide the circumstances to give rise to savant syndrome.

Two recent studies have linked savant ability with synaesthesia. Baron-Cohen *et al.* (2007) reported a case study of DT, a man with synaesthesia, who also had Asperger Syndrome and savant syndrome. At the time of testing, DT could speak 10 languages, and also had remarkable mental calculation abilities (e.g., he could multiply six-digit numbers at lightning speed) and a prodigious memory for the mathematical constant π, which he had memorised to 22 514 decimal places. DT also reported experiencing multiple forms of synaesthesia including seeing numbers with distinct colours, textures, and abstract shapes. Baron-Cohen *et al.* (2007) suggested that his savant abilities may be the result of having both synaesthesia and ASC, and proposed that the co-occurrence of synaesthesia and ASC may increase the likelihood of developing savant syndrome in general. The logic behind this claim is that synaesthesia is known to confer certain cognitive advantages, such as in memory (Simner *et al.*, 2009; Rothen *et al.*, 2012) so this advantage may underpin the extraordinary memory of this savant case study. For example, when DT recalls the decimal places of π, each digit has an additional sensory dimension from his synaesthesia (a colour, shape, and texture) that might enhance memory through dual coding of the memory cue (Terhune *et al.*, 2013) or it might enable a superior mnemonic strategy to be used. In addition, ASC is associated with hyper-systemizing (Baron-Cohen *et al.*, 2003), that is, a strong interest in patterns and rule-based information. This is thought to underlie the unusually narrow interests, sometimes called obsessions. Simner *et al.* (2009) suggested that savant skills in autism may arise through the joint mechanisms of synaesthesia, leading to enhanced memory, and ASC leading to obsessive traits, resulting in over-rehearsal of this talent.

To test these suggestions we re-examined one type of savant syndrome and showed that this case likely rested on both synaesthesia and obsessive rehearsal (Simner *et al.*, 2009). Savant AJ (Parker *et al.*, 2006) has prodigious recall of autobiographical events, as well as sequence-space synaesthesia, in which time is seen projected into convoluted spatial arrays (according to our prima facie interpretation of her detailed case-history; see Simner *et al.*, 2009). Case reports suggested AJ also had obsessive traits similar to those seen in ASC and that this led to repetitive thoughts and rehearsal of events in her memory. We showed that the form of synaesthesia experienced by AJ facilitates autobiographical memory recall to above average levels. We proposed that AJ's savant-level of recall may have arisen from an obsessive over-rehearsal of this *a priori* synaesthetic advantage. Our hypothesis was supported by LePort *et al.* (2012) who showed that savants fit the profile predicted by this theory: namely, they have significantly high levels of obsessive-compulsive traits (using the *Leyton Obsessional Inventory Score-Short Form*; Mathews *et al.*,

2004) and show superior memory abilities in domains that mirror those of our synaesthetes (e.g., autobiographical memory recall). We highlight their finding because it is exactly as had been hypothesised by us previously (Baron-Cohen *et al.*, 2007; Simner *et al.*, 2009).

Our current study aims to further understand the association between synaesthesia, ASC, and savant syndrome by asking whether synaesthesia is related to ASC in general, as has been suggested by Baron-Cohen *et al.* (2013) and Neufeld *et al.* (2013), or whether this relationship is more specifically linked to the presence of savant skills. To answer this question, we recruited three groups of individuals: individuals diagnosed with ASC who also report having a prodigious savant skill (henceforth 'ASC-savants'); individuals diagnosed with ASC but without an accompanying savant skill (henceforth 'ASC-non-savants'); and 'controls' who have neither a diagnosis of ASC nor a reported savant skill. Our definition of a prodigious savant was any individual who has a diagnosis of ASC that co-occurs with a skill/ability/talent that is not only out of keeping with the participant's own level of overall functioning but also exceeds the level found in the general population.

We screened participants for synaesthesia using both self-report and an objective diagnostic test. We tested for grapheme–colour synaesthesia in particular to follow the methods of Neufeld *et al.* (2013), and because it is a well-understood variant of synaesthesia with a well-accepted diagnostic test. Previous studies (Baron-Cohen *et al.*, 2013; Neufeld *et al.*, 2013) suggest grapheme–colour synaesthesia may also be more prevalent in ASC, but our own hypothesis is slightly different. If savant skills can arise from the combination of synaesthesia and ASC, we predict that synaesthesia should be particularly common in ASC, but only in those ASC individuals who also report savant syndrome. Hence we predict equivalent rates of synaesthesia in controls and ASC non-savants, but elevated rates in ASC individuals who are also savants.

2. Method

2.1. Participants

A total of 103 participants (67 female; mean age 36.4, range 18–51, S.D. 9.7) took part in our study. They comprised 40 ASC individuals with savant skills ('ASC-savants': 22 female; mean age 35.45 years, range 20–49, S.D. 9.1), 34 ASC individuals without a savant skill ('ASC-non-savants': 21 female; mean age 37.0 years, range 18–51, S.D. 9.1), and 29 controls with neither ASC nor a savant skill (24 female; mean age 36.9, range 18–50, S.D. 11.4). Participants were matched group-wise on age. A one-way ANOVA showed no significant difference in age across the three groups: $F(2, 100) = 0.284$, $p = 0.8$.

Participants were recruited from two sources. Two of the 40 ASC-savants were recruited from The Savant Network that is a group of individuals with a self-reported savant skill who have expressed an interest in taking part in research studies at the University of Sussex. The remaining ASC-savants were recruited from the Cambridge Autism Research Database where they had self-declared having one or more savant skills (and we subsequently determined savant status by administering our own savant questionnaire; see Procedure below). The 34 ASC-non-savant individuals and 29 controls also came from the Cambridge Autism Research Database, which holds both ASC and non-ASC participants. Participants volunteered to take part in our study in response to an email advertisement that was sent to 4172 participants in these databases (553 ASC-savants, 930 ASC-non-savants, and 2689 typical adults without a diagnosis of ASC). We took care not to mention synaesthesia at the time of recruitment. Participants were classified as ASC *versus* control, and savant *versus* non-savant using both self-report and validation measures, and these measures are described in Sect. 3 below. The study was approved by the local University Ethics Committee.

In addition to the 103 participants, 20 further participants were recruited but later excluded from our study (12 ASC-savants, six ASC-non-savants, and two controls). Fourteen of these (10 ASC-savants, four ASC-non-savants) initially indicated ASC but failed to meet our criteria when probed further (see Sect. 3) and one control participant had taken the synaesthesia test previously. Finally, five participants (two ASC-savants, two ASC-non-savants, one control) did not provide sufficient information for us to match their identity to test scores in one of the tests (which matches participants *via* email address).

2.2. Materials and Procedure

Participants were sent a URL link *via* email, which sent them to a website where they were shown the information page and consent form. Participants then completed the following tests, in the order shown below, assessing whether they had ASC, whether they had a savant skill, and finally whether they had synaesthesia.

To determine ASC status, all participants responded to a self-report question which asked "Have you received a formal diagnosis of any of the following: Autism, Asperger Syndrome, Pervasive developmental disorder not otherwise specified; 'Other'?" Although we did not administer a diagnostic test ourselves, our question specifically stated that a formal diagnosis must have been given, and we used responses to classify participants according to their ASC status. Participants with ASC from the Cambridge Autism Research Database additionally are required to record that their ASC diagnosis was given by a psychiatrist, clinical psychologist, neurologist, or paediatrician, as well as the name of the recognised clinic where this took place.

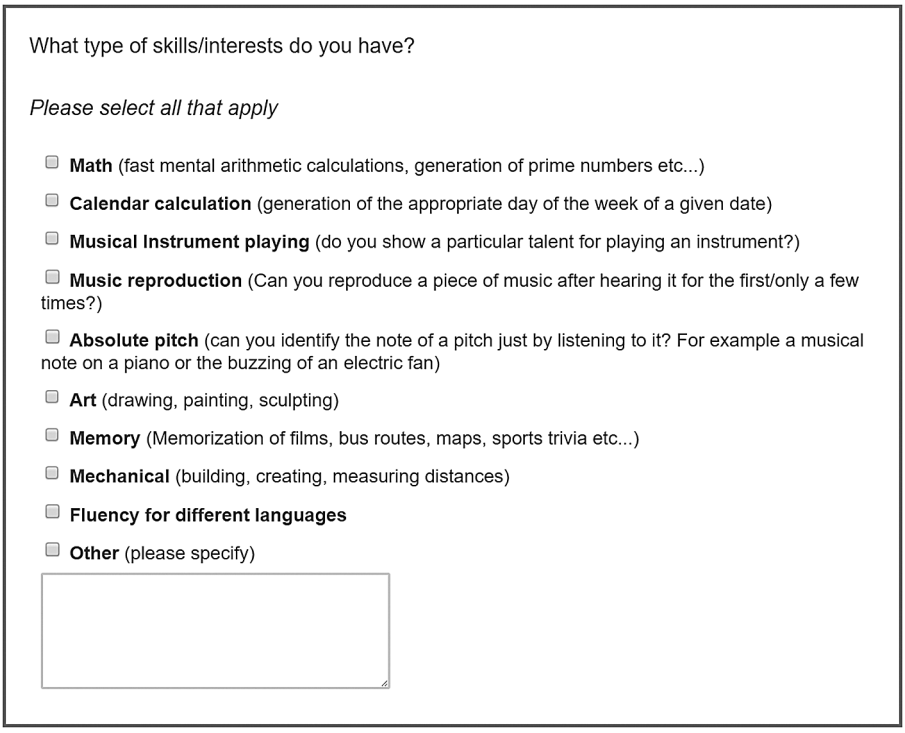

Figure 1. Savant skills presented in the self-report savant skills questionnaire.

Participants then completed a short questionnaire about savant skills, shown in Fig. 1. In this we provided a definition of savant syndrome and asked whether participants had any talents beyond those seen in the general population. If participants responded 'yes' they were given a list of nine categories of savant skills, with definitions, and could use check boxes to indicate as many as were relevant to them. They were also given space to specify other skills, and any other relevant information (e.g., how they acquired their skill). To our knowledge, there is no widely used standardized assessment for savant syndrome and so our questionnaire was created specifically for this study.

After the savant-skills questionnaire, participants followed a URL link to our assessment for grapheme–colour synaesthesia. For this we used what is considered the 'gold-standard' diagnostic test for synaesthesia (Asher *et al.*, 2006; Baron-Cohen *et al.*, 1987; Rich *et al.*, 2005). This is an assessment based on consistency: that inducer-concurrent pairings (e.g., the colours of individual letters) are highly consistent over time (Simner and Logie, 2008). The diagnostic test therefore assessed the consistency with which participants gave colour-choices for graphemes, and we used the same version of the test used by Neufeld *et al.* (2013; following Eagleman *et al.*, 2007). The diagnostic

test has two components. First, participants are asked whether they experience grapheme–colour synaesthesia, with the question "Do numbers or letters cause you to have a colour experience?" Participants respond by checking boxes for letters and/or digits. Those who check neither box are categorised as non-synaesthetes, and are guided to an exit screen. Those who respond in the affirmative for letters and/or digits are then given an objective test for grapheme colour synaesthesia (the 'consistency test'). In this test, participants are presented with each grapheme three times, in a randomised order. For each grapheme presented on-screen, participants selected their preferred colour association (e.g., A = red; B = purple...) from an on-screen colour palette, and graphemes are shown three times each, in a random order. In order to reduce the use of spatial memory techniques in remembering colour choices for each grapheme, a trial-by-trial randomisation of hue is employed on the colour picker. The mean distance in colour space between the three colours given for each grapheme was converted into a standardised consistency score, where a small standardised score reflects consistent colours (i.e., selections for the same grapheme were close in colour-space; see Eagleman *et al.*, 2007). The high level of consistency characteristic of genuine synaesthesia is indicated by a score less than 1 (Eagleman *et al.*, 2007; Neufeld *et al.*, 2013) and this was our diagnostic threshold. At the end of the test, participants received feedback on their performance in the task.

Participants were also tested for a range of other synaesthesias (e.g., weekday-colour, sound-colour) as well as for other types of individual differences (e.g., in mental imagery) and these data are reported elsewhere. The synaesthesia assessment took a maximum of 40 min to complete if participants reported synaesthesia. All 103 participants completed our initial ASC and savant questionnaires, and a total of 73 out of our 103 participants completed the synaesthesia task as described above (i.e., immediately after the ASC and savant questionnaires). Twenty-two of these 103 participants completed the synaesthesia test following a reminder by answering its initial question (i.e., "Do numbers or letters cause you to have a colour experience?") in an email before entering the main battery, and then completed the synaesthesia task in the same way as all other participants. Due to study drop-out, we were unable to obtain complete synaesthesia task scores from a total of eight out of 103 participants (see Sect. 3).

3. Results

3.1. Participant Status: Control, Versus ASC-Savant, Versus ASC-Non-Savant

The tests for ASC and savant syndrome confirmed that participants did indeed fall into three groups: ASC-savants, ASC-non-savants, and controls. All indi-

Table 1.
Cases of reported savant skill. NB. Some participants reported multiple savant skills

Skill type	Number of cases
Math	14
Calendar calculation	2
Musical instrument playing	9
Music reproduction	7
Absolute pitch	11
Art	14
Memory	23
Mechanical (building)	6
Fluency for different languages	11
Other	26

Note. Synaesthesia was not included in our list of savant skills.

viduals with ASC, but no controls, met the requirements for ASC status, which is that they self-reported having received a formal diagnosis of ASC ($n = 14$ 'autism'; $n = 59$ 'Asperger Syndrome'; $n = 1$ pervasive developmental disorder not otherwise specified). The categories of savant skills as reported by ASC-savants are shown in Table 1; no control participant reported a savant skill in any category.

3.2. Test for Synaesthesia

We investigated the prevalence of synaesthesia within each group following the standard protocol used by Neufeld *et al.* (2013), based on Eagleman *et al.* (2007). This protocol classifies participants as synaesthetes according to the following criteria: (1) self-reporting coloured letters and/or digits in the initial self-report questionnaire, *and* (2) achieving a colour-distance score below the 1.0 diagnostic threshold for synaesthesia. Participants who satisfied both criteria were classified as synaesthetes, and the remaining participants were classified as non-synaesthetes.

Of the 103 participants in our study, eight ended their participation before completing the synaesthesia test, but did nonetheless complete our ASC and savant questionnaires. These eight non-completing participants were approximately evenly divided across groups: ($n = 3$ ASC-savants, $n = 3$ ASC-non-savants, $n = 2$ controls). Below we present the percentage of synaesthetes per group according to how many subjects were recruited in total ($n = 40$, $n = 34$, and $n = 29$ respectively) based on the conservative assumption that the missing eight subjects were non-synaesthetes. Then we repeat our analyses using just those participants who were fully screened ($n = 37$, $n = 31$, and $n = 27$ respectively; see Note 1) given that we fully screened approximately the same

Table 2.
Types of grapheme colour synaesthesia, and types of savant skills (where applicable), reported by participants. Column 1 shows individual participants from each group. Columns 2 and 3 show the type of grapheme–colour synaesthesia diagnosed. Column 4 shows the type of savant skill reported

	Letter–colour	Number–colour	Talent reported
ASC-savant	x	x	music reproduction; art
ASC-savant	x	x	memory; languages
ASC-savant	x		instrument playing
ASC-savant		x	memory; art
ASC-non-savant	x		
Control		x	

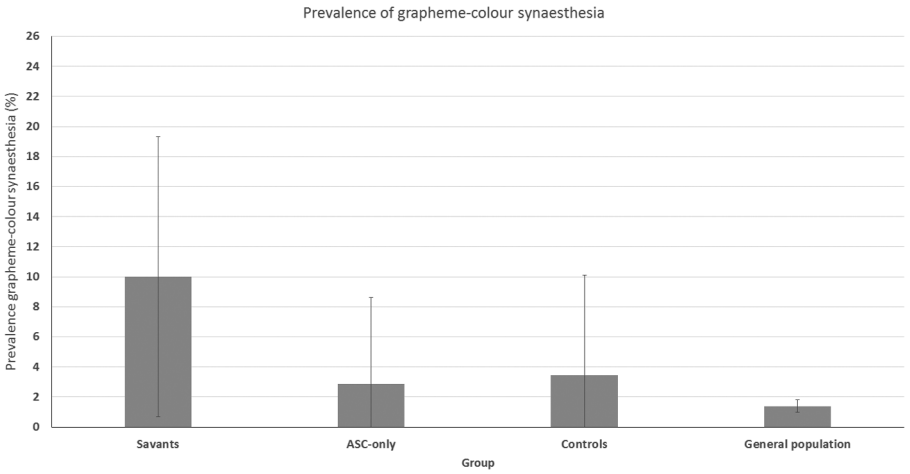

Figure 2. Prevalence of grapheme–colour synaesthesia (with 95% Wald confidence interval) in ASC-savants, ASC-non-savants, and Controls (columns 1–3 respectively). Column 4 shows the expected prevalence in the general population as reported by Simner and Carmichael (2015).

percentage across groups (i.e., 92.5%; 91.2% and 93.1% respectively). In both cases, the same pattern of results emerges.

In total, we found six grapheme–colour synaesthetes in our sample of 103 participants, of whom one was a control (one female), one was ASC-non-savant (one female) and four were ASC-savants (three female). Table 2 shows whether each synaesthete had coloured letters, coloured numbers, or both, and Fig. 2 shows the prevalence of grapheme–colour synaesthesia as a percentage of each group. Bars 1–3 show the ASC-savants, ASC-non-savants and Controls, respectively. Since our study is a direct comparison to Neufeld *et al.* (2013) we follow their approach of comparing each group pairwise to a robust

estimate of prevalence from the general population. This estimate is shown in bar 4 and is taken from Simner and Carmichael (2015) which represents the equivalent prevalence of grapheme–colour synaesthesia in the general population; i.e., the prevalence of people with synaesthetic colours for letters and/or numbers). We selected this particular control baseline because it represented a large, robust screening of the general population for grapheme–colour synaesthesia (Simner and Carmichael, 2015 screened $n = 3839$ people and found 54 grapheme–colour synaesthetes who had colours for letters and/or numbers, giving a prevalence of 1.4%). Moreover, this baseline has been shown to not differ from the baseline used by Neufeld *et al.* (see Simner and Carmichael, 2015) and it was generated *via* an identical diagnostic to the one used here.

The prevalence of synaesthesia in the ASC-savant group (10.0%) was over seven times higher than in the general population (1.4%) and this difference was significant [$\chi^2(1) = 14.721$, $p < 0.001$; chi square test with Yates continuity correction]. In contrast, there was no significant difference between baseline and ASC-non-savants [2.9%; $\chi^2(1) = 0.001$, $p = 0.972$] and no significant difference between baseline and Controls [3.5%; $\chi^2(1) = 0.022$, $p = 0.882$]. In summary, there were more synaesthetes than expected in the ASC-savant group, but not in our group of controls or ASC-non-savants.

4. Discussion

This study used the methods reported by Neufeld *et al.* (2013) to test whether grapheme–colour synaesthesia occurs significantly more often in people with ASC. Our rates of grapheme–colour synaesthesia were compared against a 1.4% baseline from the general population (Simner and Carmichael, 2015). In comparison we found no elevated rates in controls, nor people with ASC who do *not* report savant skills, but a significantly higher rate in ASC individuals *who do* report savant skills. Overall, our findings therefore suggest that synaesthesia is not linked to autism per se, but specifically to individuals with autism who report having savant skills. Below we discuss the implications of our findings, and the possible role of synaesthesia in the development of prodigious talent.

Our study was conducted in reference to two earlier experiments, by Baron-Cohen *et al.* (2013) and Neufeld *et al.* (2013). These studies had found a higher prevalence of synaesthesia in ASC but importantly had not controlled for the inclusion of savants in their ASC samples. When we do this here (i.e., remove the savants), we no longer find high rates of synaesthesia. In other words, the 2.9% synaesthetes found in our ASC-non-savant group falls far below Neufeld *et al.*'s finding of 17.2% [and is outside their 95% confidence interval (3.5%–31%)]. We therefore conclude that their higher prevalence rates

of synaesthesia in ASC likely arose from their inadvertent inclusion of savants. This in turn means we should be able to approximate Neufeld *et al.*'s results if we recombined both our (savant and non-savant) ASC groups together. And this is what we find: merging our samples (i.e., ASC-savants + ASC-non-savant groups) to approximate the participants in other studies gives an estimate of grapheme–colour synaesthesia (5 out of 74, or 6.8%) which falls within the 95% confidence range of Neufeld *et al.* Finally, our combined data also mirrors that of Baron-Cohen *et al.* (2013): when we re-calculate our prevalence of synaesthesia using their methods (i.e., self-report only; Baron-Cohen *et al.*, 2013) our prevalence from all ASC individuals combined (8 out of 74, or 10.81%) is strikingly similar to theirs (18 out of 164, or 10.98%). We therefore suggest that all three studies converge on the conclusion that synaesthesia is found at elevated rates in ASC populations (Baron-Cohen *et al.*, 2013; Neufeld *et al.*, 2013) but that this effect is likely driven by the savants. Nonetheless, all three studies have relatively small numbers and so future experiments replicating these findings are needed.

Future studies might also wish to assess savant skills using an objective test (e.g., for skills such as mental arithmetic) as this would have several advantages over the self-report method used here. For instance, a primary limitation of our current savant questionnaire is that different participants' interpretations of what constitutes a savant-skill may vary. Indeed, there is likely to be variation in the extent to which an individual's own skill level is perceived to be superior or inferior compared to the general population. In addition, other factors such as overestimating or underestimating one's own skill level, as well as personality traits such as modesty might further influence whether or not a participant classifies themselves as satisfying our criteria for having a savant skill. Using objective tests would help to standardize the classification of savant participants and we are now creating such a test battery in our lab.

The question remains as to why synaesthesia is observed more often in savant syndrome compared to other populations, and here we consider two alternative lines of argumentation. First, we have previously suggested (Baron-Cohen *et al.*, 2007; Simner *et al.*, 2009) that synaesthesia may lead to savant syndrome *via* "enriched-memory plus over-rehearsal". Specifically, synaesthesia is known to give marginally improved memory (e.g., for digits) because, for example, synaesthetic colours may enrich memory representations (e.g., if digits are encoded as both numbers and colours, they would have richer memory representations; Rothen *et al.*, 2012). Importantly, this improved memory could potentially be elevated to savant levels (e.g., memory for thousands of digits) if obsessively over-rehearsed — and crucially — obsessiveness is a trait that has been linked to ASC (Zandt *et al.*, 2006). Hence, when synaesthesia and ASC co-occur, the chances of developing a savant skill may be increased

(Baron-Cohen *et al.*, 2007; Simner *et al.*, 2009). If so, this would indeed be detected as higher levels of synaesthesia in savants, as found here.

An alternative line of argumentation linking synaesthesia and savant syndrome is what we might call 'systemizing and veridical mapping'. As stated earlier, systemizing is defined as the drive to identify patterns in rule-based information, and is a trait that is elevated in people with ASC (Baron-Cohen *et al.*, 2003). Veridical mapping is the related ability to match two different systems, by noting their shared consistent regularities (e.g., shared topographic relationships in two different charts). Veridical mapping and hypersystemizing may themselves emerge from other traits tied to autism, which are sensory hypersensitivity, excellent attention to detail and superior low-level perceptual abilities (Baron-Cohen *et al.*, 2009; Mottron *et al.*, 2006). Importantly, Mottron, Dawson and Soulières (2009; also Bouvet at al., 2014; Mottron *et al.*, 2013) have hypothesised that veridical mapping may motivate savant syndrome, and may independently motivate synaesthesia. For example, Mottron *et al.* (2013) suggest that the savant skill of hyperlexia (precocious reading ability) depends on veridical mapping across language systems: i.e., detecting patterns between the written system of graphemes and the spoken system of phonemes. Mottron *et al.* (2013) suggest that veridical mapping might also encourage grapheme–colour synaesthesia, in which relationships between graphemes (e.g., the low to high frequency of graphemes in English) are known to be mapped onto relationships between colours (the low to high frequency of colour-terms in English; see Simner *et al.*, 2005). In this way, Mottron *et al.* (2013) suggest that veridical mapping might be a causal factor in both savant syndrome and synaesthesia, albeit independently. If so, one consequence would therefore be elevated rates of synaesthesia in savant syndrome, as we find here.

It is important to note that 'enriched-memory plus over-rehearsal' and 'systemizing and veridical mapping' make different predictions about the role of synaesthesia in savant syndrome. The former predicts that savant skills are tied to the type of synaesthesia experienced (e.g., having coloured digits would lead to prodigious digit recall) while the latter does not. Our present data cannot speak to this question because we present data on only one type of synaesthesia. Hence a music savant may well show grapheme–colour synaesthesia, but further study would be needed to determine whether he/she also showed forms related to music. We therefore look to future studies that might match savant skills to synaesthesias that could shed direct light on this question.

In conclusion, our study found that grapheme–colour synaesthesia occurs significantly more often in savant syndrome compared to the general population, rather than being tied to ASC per se. Our results extend the findings of two previous studies (Baron-Cohen *et al.*, 2013; Neufeld *et al.*, 2013) that

both found synaesthesia to occur significantly more often in autism compared to controls. Our data further strengthens the link between synaesthesia and savant syndrome (Baron-Cohen *et al.*, 2007; Simner *et al.*, 2009) and the related theories proposing a mediating mechanism of veridical mapping (Bouvet at al., 2014).

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Note

1. If we repeat our analyses based only on the 95 participants who completed all aspects of our study (i.e., excluding those that did not complete the synaesthesia test) we find the same pattern of results. Four out of 37 participants with grapheme–colour synaesthesia in the ASC-savant group (10.8%) is significantly higher than the general population baseline [$\chi^2(1) = 16.373$, $p < 0.001$]. There is no significant difference to baseline for one synaesthete out of 31 ASC-non-savants [3.2%; $\chi^2(1) = 0.01$, $p = 0.920$] or for one synaesthete out of 27 controls [3.9%; $\chi^2(1) = 0.065$, $p = 0.799$]. Finally, the same pattern of results is seen if we use the population baseline of Neufeld *et al.* (2013; i.e., 2%, from Simner *et al.*, 2006b). Indeed, the pattern remains the same both when considering every participant who took part in this study [40 ASC-savants, $\chi^2(1) = 6.69$, $p = 0.01$; 34 ASC-non-savants, $\chi^2(1) = 0.00$, $p = 1.00$; 29 controls, $\chi^2(1) = 0.00$, $p = 1.00$] or just those who completed all aspects of our tests [37 ASC-savants, $\chi^2(1) = 6.43$, $p = 0.015$; 31 ASC-non-savants, $\chi^2(1) = 0.00$, $p = 1.00$; 29 controls, $\chi^2(1) = 0.00$, $p = 1.00$].

References

American Psychiatric Association (2013). *Diagnostic and Statistical Manual of Mental Disorders*, 5th edn. American Psychiatric Publishing, Arlington, VA, USA.

Asher, J. E., Aitken, M. R., Farooqi, N., Kurmani, S. and Baron-Cohen, S. (2006). Diagnosing and phenotyping visual synaesthesia: a preliminary evaluation of the revised test of genuineness (TOG-R), *Cortex* **42**, 137–146.

Asher, J. E., Lamb, J. A., Brocklebank, D., Cazier, J. B., Maestrini, E., Addis, L., Sen, M., Baron-Cohen, S. and Monaco, A. P. (2009). A whole-genome scan and fine-mapping linkage study of auditory-visual synesthesia reveals evidence of linkage to chromosomes 2q24, 5q33, 6p12, and 12p12, *Am. J. Hum. Genet.* **84**, 279–285.

Banissy, M. J., Walsh, V. and Ward, J. (2009). Enhanced sensory perception in synaesthesia, *Exp. Brain Res.* **196**, 565–571.

Bargary, G. and Mitchell, K. J. (2008). Synaesthesia and cortical connectivity, *Trends Neurosci.* **31**, 335–342.

Baron-Cohen, S., Wyke, M. A. and Binnie, C. (1987). Hearing words and seeing colours: an experimental investigation of a case of synaesthesia, *Perception.* **16**, 761–767.

Baron-Cohen, S., Richler, J., Bisarya, D., Gurunathan, N. and Wheelwright, S. (2003). The systemizing quotient: an investigation of adults with Asperger syndrome or high–functioning autism, and normal sex differences, *Phil. Trans. R. Soc. Lond. B Biol. Sci.* **358**(1430), 361–374.

Baron-Cohen, S., Bor, D., Billington, J., Asher, J., Wheelwright, S. and Ashwin, C. (2007). Savant memory in a man with colour form-number synaesthesia and asperger syndrome, *J. Consc. Stud.* **14**, 237–251.

Baron-Cohen, S., Ashwin, E., Ashwin, C., Tavassoli, T. and Chakrabarti, B. (2009). Talent in autism: hyper-systemizing, hyper-attention to detail and sensory hypersensitivity, *Phil. Trans. R. Soc. Lond. B Biol. Sci.* **364**(1522), 1377–1383.

Baron-Cohen, S., Johnson, D., Asher, J., Wheelwright, S., Fisher, S. E., Gregersen, P. K. and Allison, C. (2013). Is synaesthesia more common in autism? *Mol. Autism* **4**, 40. DOI:10.1186/2040-2392-4-40.

Bor, D., Billington, J. and Baron-Cohen, S. (2007). Savant memory for digits in a case of synaesthesia and Asperger syndrome is related to hyperactivity in the lateral prefrontal cortex, *Neurocase* **13**, 311–319.

Bouvet, L., Donnadieu, S., Valdois, S., Caron, C., Dawson, M. and Mottron, L. (2014). Veridical mapping in savant abilities, absolute pitch, and synesthesia: an autism case study, *Front. Psychol.* **5**, 106. DOI:10.3389/fpsyg.2014.00106.

Casanova, M. and Trippe, J. (2009). Radial cytoarchitecture and patterns of cortical connectivity in autism, *Phil. Trans. R. Soc. Lond. B Biol. Sci.* **364**(1522), 1433–1436.

Chia, N. K. H. (2012). Autism enigma: the need to include savant and crypto-savant in the current definition, *Acad. Res.* **2**, 234.

Cytowic, R. E. (1995). Synesthesia: phenomenology and neuropsychology, *Psyche* **2**, 2–10.

DePape, A. M. R., Hall, G. B., Tillmann, B. and Trainor, L. J. (2012). Auditory processing in high-functioning adolescents with autism spectrum disorder, *PLoS One* **7**, e44084. DOI:10.1371/journal.pone.0044084.

Dohn, A., Garza-Villarreal, E. A., Heaton, P. and Vuust, P. (2012). Do musicians with perfect pitch have more autism traits than musicians without perfect pitch? An empirical study, *PLoS One* **7**, e37961. DOI:10.1371/journal.pone.0037961.

Eagleman, D. M., Kagan, A. D., Nelson, S. S., Sagaram, D. and Sarma, A. K. (2007). A standardized test battery for the study of synesthesia, *J. Neurosci. Meth.* **159**, 139–145.

Gregersen, P. K., Kowalsky, E., Lee, A., Baron-Cohen, S., Fisher, S. E., Asher, J. E., Ballard, D., Freudenberg, J. and Li, W. (2013). Absolute pitch exhibits phenotypic and genetic overlap with synesthesia, *Hum. Mol. Genet.* **22**, 2097–2104.

Grossenbacher, P. G. and Lovelace, C. T. (2001). Mechanisms of synesthesia: cognitive and physiological constraints, *Trends Cogn. Sci.* **5**, 36–41.

Howlin, P., Goode, S., Hutton, J. and Rutter, M. (2009). Savant skills in autism: psychometric approaches and parental reports, *Phil. Trans. R. Soc. Lond. B Biol. Sci.* **364**(1522), 1359–1367.

Hubbard, E. M. and Ramachandran, V. S. (2005). Neurocognitive mechanisms of synesthesia, *Neuron* **48**, 509–520.

International Molecular Genetic Study of Autism Consortium (2001). A genomewide screen for autism: strong evidence for linkage to chromosomes 2q, 7q, and 16p, *Am. J. Hum. Genet.* **69**, 570–581.

Jao Keehn, R. J., Sanchez, S. S., Stewart, C. R., Zhao, W., Grenesko-Stevens, E. L., Keehn, B. and Müller, R. A. (2016). Impaired downregulation of visual cortex during auditory processing is associated with autism symptomatology in children and adolescents with autism spectrum disorder, *Autism Res.* **10**, 130–143.

Kemner, C., Verbaten, M. N., Cuperus, J. M., Camfferman, G. and Van Engeland, H. (1995). Auditory event-related brain potentials in autistic children and three different control groups, *Biol. Psychiat.* **38**, 150–165.

Leekam, S. R., Nieto, C., Libby, S. J., Wing, L. and Gould, J. (2007). Describing the sensory abnormalities of children and adults with autism, *J. Autism Dev. Disord.* **37**, 894–910.

LePort, A. K., Mattfeld, A. T., Dickinson-Anson, H., Fallon, J. H., Stark, C. E., Kruggel, F. and McGaugh, J. L. (2012). Behavioral and neuroanatomical investigation of highly superior autobiographical memory (HSAM), *Neurobiol. Learn. Mem.* **98**, 78–92.

Marco, E. J., Hinkley, L. B., Hill, S. S. and Nagarajan, S. S. (2011). Sensory processing in autism: a review of neurophysiologic findings, *Pediatr. Res.* **69**, 48R–54R.

Mathews, C. A., Jang, K. L., Hami, S. and Stein, M. B. (2004). The structure of obsessionality among young adults, *Depress. Anxiety* **20**, 77–85.

McIntosh, A. M., Maniega, S. M., Lymer, G. K. S., McKirdy, J., Hall, J., Sussmann, J. E., Bastin, M. E., Clayden, J. D., Johnstone, E. C. and Lawrie, S. M. (2008). White matter tractography in bipolar disorder and schizophrenia, *Biol. Psychiatry* **64**, 1088–1092.

Mottron, L., Dawson, M., Soulières, I., Hubert, B. and Burack, J. (2006). Enhanced perceptual functioning in autism: an update, and eight principles of autistic perception, *J. Autism Dev. Disord.* **36**, 27–43.

Mottron, L., Dawson, M. and Soulières, I. (2009). Enhanced perception in savant syndrome: patterns, structure and creativity, *Phil. Trans. R. Soc. Lond. B Biol. Sci.* **364**(1522), 1385–1391.

Mottron, L., Bouvet, L., Bonnel, A., Samson, F., Burack, J. A., Dawson, M. and Heaton, P. (2013). Veridical mapping in the development of exceptional autistic abilities, *Neurosci. Biobehav. Rev.* **37**, 209–228.

Neufeld, J., Roy, M., Zapf, A., Sinke, C., Emrich, H. M., Prox-Vagedes, V., Dillo, W. and Zedler, M. (2013). Is synesthesia more common in patients with Asperger syndrome? *Front. Hum. Neurosci.* **7**, 847. DOI:10.3389/fnhum.2013.00847.

Parker, E. S., Cahill, L. and McGaugh, J. L. (2006). A case of unusual autobiographical remembering, *Neurocase* **12**, 35–49.

Rich, A. N., Bradshaw, J. L. and Mattingley, J. B. (2005). A systematic, large-scale study of synaesthesia: implications for the role of early experience in lexical-colour associations, *Cognition* **98**, 53–84.

Rogers, S. J. and Ozonoff, S. (2005). Annotation: what do we know about sensory dysfunction in autism? A critical review of the empirical evidence, *J. Child Psychol. Psychiat.* **46**, 1255–1268.

Rothen, N., Meier, B. and Ward, J. (2012). Enhanced memory ability: insights from synaesthesia, *Neurosc. Biobehav. Rev.* **36**, 1952–1963.

Rouw, R. and Scholte, H. S. (2007). Increased structural connectivity in grapheme-color synesthesia, *Nat. Neurosci.* **10**, 792–797.

Simner, J. and Carmichael, D. A. (2015). Is synaesthesia a dominantly female trait? *Cogn. Neurosci.* **6**, 68–76.

Simner, J. and Logie, R. H. (2008). Synaesthetic consistency spans decades in a lexical–gustatory synaesthete, *Neurocase* **13**, 358–365.

Simner, J., Ward, J., Lanz, M., Jansari, A., Noonan, K., Glover, L. and Oakley, D. A. (2005). Non-random associations of graphemes to colours in synaesthetic and non-synaesthetic populations, *Cogn. Neuropsychol.* **22**, 1069–1085.

Simner, J., Glover, L. and Mowat, A. (2006a). Linguistic determinants of word colouring in grapheme–colour synaesthesia, *Cortex* **42**, 281–289.

Simner, J., Mulvenna, C., Sagiv, N., Tsakanikos, E., Witherby, S. A., Fraser, C., Scott, K. and Ward, J. (2006b). Synaesthesia: the prevalence of atypical cross-modal experiences, *Perception* **35**, 1024–1033.

Simner, J., Mayo, N. and Spiller, M. J. (2009). A foundation for savantism? Visuo-spatial synaesthetes present with cognitive benefits, *Cortex* **45**, 1246–1260.

Tavassoli, T., Hoekstra, R. A. and Baron-Cohen, S. (2014a). The Sensory Perception Quotient (SPQ): development and validation of a new sensory questionnaire for adults with and without autism, *Mol. Autism* **5**, 29. DOI:10.1186/2040-2392-5-29.

Tavassoli, T., Miller, L. J., Schoen, S. A., Nielsen, D. M. and Baron-Cohen, S. (2014b). Sensory over-responsivity in adults with autism spectrum conditions, *Autism* **18**, 428–432.

Terhune, D. B., Wudarczyk, O. A., Kochuparampil, P. and Kadosh, R. C. (2013). Enhanced dimension-specific visual working memory in grapheme–color synesthesia, *Cognition* **129**, 123–137.

Tomchek, S. D. and Dunn, W. (2007). Sensory processing in children with and without autism: a comparative study using the short sensory profile, *Am. J. Occup. Ther.* **61**, 190–200.

Treffert, D. A. (2009). The savant syndrome: an extraordinary condition. A synopsis: past, present, future, *Phil. Trans. R. Soc. Lond. B Biol. Sci.* **364**(1522), 1351–1357.

Ward, J., Hoadley, C., Hughes, J. E. A., Smith, P., Allison, C., Baron-Cohen, S. and Simner, J. (2017). Atypical sensory sensitivity as a shared feature between synaesthesia and autism, *Sci. Rep.* **7**, 41155. DOI:10.1038/srep41155.

Ward, J., Huckstep, B. and Tsakanikos, E. (2006). Sound-colour synaesthesia: to what extent does it use cross-modal mechanisms common to us all? *Cortex* **42**, 264–280.

Zamm, A., Schlaug, G., Eagleman, D. M. and Loui, P. (2013). Pathways to seeing music: enhanced structural connectivity in colored-music synesthesia, *Neuroimage* **74**, 359–366.

Zandt, F., Prior, M. and Kyrios, M. (2007). Repetitive behaviour in children with high functioning autism and obsessive compulsive disorder, *J. Autism Dev. Disord.* **37**, 251–259.