

Early detection of markers for synaesthesia in childhood populations

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We show that the neurological condition of synaesthesia—which causes fundamental differences in perception and cognition throughout a lifetime—is significantly represented within the childhood population, and that it manifests behavioural markers as young as age 6 years. Synaesthesia gives rise to a merging of cognitive and/or sensory functions (e.g. in grapheme-colour synaesthesia, reading letters triggers coloured visual photisms) and adult synaesthesia is characterized by a fixed pattern of paired associations for each synaesthete (e.g. if a is carmine red, it is always carmine red). We demonstrate that the onset of this systematicity can be detected in young grapheme-colour synaesthetes, but is an acquired trait with a protracted development. We show that grapheme-colour synaesthesia develops in a way that supersedes the cognitive growth of non-synaesthetic children (with both average and superior abilities) in a comparable paired association task. With methodology based on random sampling and behavioural tests of genuineness, we reveal the prevalence of grapheme-colour synaesthesia in children (over 170 000 grapheme-colour synaesthetes ages 0-17 in the UK, and over 930 000 in the US), the progression of the condition in longitudinal testing, and the developmental differences between synaesthetes and non-synaesthetes in matched tasks. We tested 615 children age 6-7 years from 21 primary schools in the UK. Each child was individually assessed with a behavioural test for grapheme-colour synaesthesia, which first detects differences between synaesthetes and non-synaesthetes, and then tracks the development of each group across 12 months (from ages 6/7 to 7/8 years). We show that the average UK primary school has 2-3 grapheme-colour synaesthetes at any time (and the average US primary school has five) and that synaesthetic associations (e.g. a = carmine red) develop from chaotic pairings into a system of fixed, consistent cogno-sensory responses over time. Our study represents the first assessment of synaesthesia in a randomly sampled childhood population demonstrating the real-time development of the condition. We discuss the complex profile of benefits and costs associated with synaesthesia, and our research calls for a dialogue between researchers, clinicians and educators to highlight the prevalence and characteristics of this unusual condition.

Keywords: synaesthesia; synesthesia; cross-modal; binding; grapheme-colour

Introduction

Synaesthesia is a neurological condition that gives rise to a merging of sensory and/or cognitive functions, and so for people with synaesthesia, everyday activities (e.g. reading, listening to music) trigger extra-ordinary experiences (e.g. colours, tastes). For lexical-gustatory synaesthetes, for example (Simner and Ward, 2006) reading, saying or thinking about words triggers perceptual sensations of flavour in the mouth, and for graphemecolour synaesthetes (Dixon et al., 2000; Rich et al., 2005), letters and numerals trigger sensations of colour (photisms). These atypical sensations arise spontaneously during development (i.e. without

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effort), and synaesthesia in adults is typified by the consistency of mapping between the triggering stimulus ('inducer') and the resultant experience ('concurrent'). Hence, for any given adult synaesthete, each inducer consistently triggers the same concurrent over time (e.g. if the letter a is carmine red, it is always carmine red). This consistency has been objectively demonstrated across months, years (Dixon et al., 2000; Ward and Simner, 2003, 2005; Rich et al., 2005; Simner and Ward, 2006; Simner et al., 2006; Smilek, et al., 2002a, b; Yaro and Ward, 2007) and even decades (Simner and Logie, 2008) and is considered to be the 'behavioural hallmark' of synaesthesia (Rich et al., 2005).

Studies of synaesthesia's familial transmission patterns (Smilek et al., 2002b; Ward and Simner, 2005) suggest a genetic mode of inheritance, and this inheritance is assumed to give rise to a predisposition for increased cerebral communication. Neuroimaging studies show that the sensory experiences reported by synaesthetes are accompanied by activation in the related sensory cortices (Nunn et al., 2002; Hubbard et al., 2005; Rouw and Scholte, 2007). Hence, synaesthetes reporting colours from letters or spoken words show increased activation in colour selective regions, in the human V4 (hV4) complex of the ventral-occipital cortex (Nunn et al., 2002; Hubbard et al., 2005). This activation arises when synaesthetes are exposed to linguistic, but not nonlinguistic stimuli and no similar activity is found in control participants. Synaesthetic experiences appear to have anatomical roots, and recent DTI investigations have shown increased structural connectivity in the brains of synaesthetes (Rouw and Scholte, 2007). Grapheme-colour synaesthetes demonstrated greater fractional anisotropy suggesting increased white matter tracts compared with controls in the right fusiform gyrus, close to regions involved in word and colour processing. Synaesthetes also showed increased connectivity in the left intraparietal sulcus and frontal cortex, and these are regions known to be involved in feature binding. Such findings are consistent with the notion of synaesthesia as an extreme or atypical binding phenomenon, and fit with neuroimaging and transcranial magnetic stimulation studies showing that the parietal cortex is essential for the synaesthetic binding of colour (Weiss et al., 2005; Esternan et al., 2006; see Hubbard, 2007).

There has been speculation on the question of lateralization in grapheme-colour synaesthesia, given diverging evidence from recent imaging work. One dominant model has suggested that, for synaesthetes, reading causes additional activity to pass from regions involved in grapheme processing (the 'visual word form area'; VWFA, McCandliss et al., 2003) to neighbouring regions in left colour processing areas (Hubbard and Ramachandran, 2005), and support has come from fMRI studies showing either left-lateralized or bilateral activation in hV4 (Nunn et al., 2002; Hubbard et al., 2005; Rouw and Scholte, 2007). In contrast, Rouw and Scholte (2007) found evidence of increased connectivity and fMRI activity in the right temporal cortex. Hubbard (2007) points to similar lateralization discrepancies for the role of parietal cortices in synaesthetic binding, and suggests that the issue of laterality should be approached with caution, given the differing techniques and small sample sizes that have thus far been brought to bear on the matter.

Synaesthesia has a complex profile of benefits and costs, which manifest differently depending on the variant. Lexical-gustatory synaesthetes for example (who experience tastes from words) report difficulties maintaining attention when reading (Ward and Simner, 2003), while grapheme-colour synaesthetes show superior colour (Yaro and Ward, 2007) and digit memory (Smilek et al., 2002a). At the same time, these latter demonstrate significant difficulties retaining letters and numerals presented in colours that conflict with their synaesthetic sensations (e.g. a red 5, if 5 is synaesthetically green; e.g. Smilek et al., 2002a) as well as a sense of malaise when viewing such mismatched configurations (Callejas et al., 2007). Equally, while grapheme-colour synaesthesia may endow superior spelling proficiency (Linn et al., 2008), other variants (visuo-spatial synaesthesia, in which numbers are projected into spatial arrays) are accompanied by indications of difficulties in arithmetic, both in self-report, and with detectable speed impairments in mental calculation (Ward J, et al., Manuscript in preparation).

Although studies on synaesthesia have proliferated during the past decade (for statistics on this growth see Simner, 2007), comparatively little attention has been given to developmental questions. Adult studies have speculated on the possible roots of synaesthesia (Ward and Simner, 2005) and one emergent view has been that extra connectivity may arise from a failure in synaptic pruning during the first few months of life (Maurer, 1997). However, an early view of synaesthetic determinism does not marry easily with the broad range of synaesthesia, the overwhelming majority of which (~88%, Simner et al., 2006) are triggered by learned linguistic units, or involve other cognitive constructs that are acquired during mid or even late childhood (Simner and Hubbard, 2006). However, in the last 10 years, a variety of data has suggested that human brain development, unlike that of nonhuman primates, is protracted and heterochronous (occurring at different times in different brain regions). For example, Huttenlocher and Dabholkar (1997) found that while synaptic density in the visual cortex falls off ~500 days after conception, density in the primary auditory cortex starts to decrease only after age 3.5 years, while frontal cortex density decreases more slowly from 3.5 until at least 11 years. Such data demonstrate that human brain development is a complex, prolonged process and that it may yet allow for delayed or failed pruning that could interact with learning at relatively late stages in development (Simner and Hubbard, 2006).

The involvement of learned units suggests that grapheme-colour synaesthesia must have been acquired at some stage after the acquisition of the inducer. For grapheme-colour synaesthetes, for example, adult-like connectivity (consistently linking, say, the colour red with the letter a) can only emerge once the specific grapheme triggers have been acquired, and this process of alphabet acquisition begins during early years of formal education. This places the moment of 'synaesthetic acquisition' some time in early childhood for grapheme-colour synaesthetes, rather than from the moment of birth (which is a logical possibility at least for other types of synaesthetes; e.g. sound-colour synaesthetes). Hence there is dissociation between adult recollections of unchanging, ever-present sensations (Dixon et al., 2004) and a more likely scenario in which synaesthesia emerges, probably

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incrementally, on a trajectory that shadows the acquisition of the inducer. Notwithstanding speculation from adult studies, there has been an almost entire absence of any direct testing of these developmental hypotheses. The few studies showing synaesthesia in children (Simner et al., 2006; Green and Goswami, 2008) have documented cases who are already in, or close to their teens and who already show adult-like synaesthesia. Moreover, a number of these cases would have come from self-referred samples, within families where the synaesthesia is known and discussed, and where the child's synaesthesia may have been shaped to some degree by this feedback. No study has ever examined the presence of this neurological condition in a randomly sampled young population, and no study has tracked its development over time. As a result, there has been no information about the prevalence of the condition among children, nor any longitudinal comparison of the development of synaesthetes and non-synaesthete. The current study aims to provide this data.

In our study, a large population of children aged 6-7 years were assessed for grapheme-colour synaesthesia (one of the most common variants; Simner et al., 2006), based on the assumption that synaesthetes' grapheme-colour associations will be more consistent than those of their peers (Dixon et al., 2000; Smilek et al., 2002a, b; Ward and Simner, 2003, 2005; Rich et al., 2005; Simner and Ward, 2006; Simner et al., 2006; Yaro and Ward, 2007). Synaesthetes were identified, conservatively, as those whose grapheme-colour associations were significantly more consistent in a surprise retest over 12 months compared with their peers' over just 10s, and our classifications were additionally supported by quantitative questionnaire data and developmental patterns over time. Questionnaires assessed the automaticity, consistency and certainty of our synaesthetes' colour sensations, and such questions have been shown to distinguish between synaesthetes and non-synaesthetes in adult studies (Simner et al., 2006). We also assessed the development of grapheme-colour synaesthesia over 12 months (from age 6/7 to 7/8 years) to determine how synaesthetic associations (e.g. a = red; b = purple) emerge over time. We compared this development to advances in cognitive memory performance for an equivalent task in a group of nonsynaesthetic children, with both average and superior memories for comparable stimuli.

Methods

Six hundred and fifteen children (male = 299; female = 316) aged 6–7 years (6=338; 7=277) were sampled from 21 primary schools in the UK (cities of Edinburgh, Scotland and Bath, England). All children spoke fluent English. An additional six children were tested but subsequently excluded: one reported himself to be colour blind, one withdrew consent midway through testing, and four failed to follow task instructions. All children completed a timed computerized test in Session 1, and a subset were retested 12 months later (Session 2). Children were tested individually in both sessions.

In Session 1, participants performed a computerized test in which they individually viewed each of 36 randomly ordered graphemes (*a*–*z*; 0–9) on screen, along with an electronic palette of 13 colours, whose arrangement was randomized on every trial. Our 13 colours (black, dark blue, brown, dark green, grey, pink, purple, orange, red, white,

light blue, light green and yellow) represent the irreducible colour terms of Berlin and Kay (1969), plus light/dark variants of blue and green [Although synaesthetes' colours are highly specific (e.g. Ward et al., 2005) and could be closely approximated with sufficient time and effort using an extensive palette comprising many thousands of colours (as we have done elsewhere; e.g. Simner et al., 2006), prior studies have shown that a presentation of our condensed palette allows for a successful assessment of synaesthesia that is practical for large numbers of participants, or for those with limited attention (Day, 2005; Simner et al., 2006). When faced with the core palette, synaesthetes are systematic in their choices, while non-synaesthetes are significantly more random (Simner et al., 2006)]. Participants were instructed to select (with a computer mouse) what they considered to be the 'best' colour for each grapheme. They were told there was no right or wrong answer but that they should avoid choosing the same colour repeatedly. Approximately 10s after completing all 36 graphemes, participants performed an immediate surprise retest, in which the order of graphemes and colours was re-randomized. (Specifically, our program presented all 36 graphemes in a random order, then paused for 10s, then began again, showing the same, but re-randomized, graphemes. During the pause, children were told that they should wait a few moments and then 'carry on as before'). We calculated each child's immediate consistency in Session 1 as the number of consistent colour choices for letters (out of 26) and numerals (out of 10) across this \sim 10s retest period. Participants performing significantly higher than the mean (whom we classed as 'potential synaesthetes') repeated the procedure in a surprise session one year later. Also tested in Session 2 were 40 'average-memory controls', orthogonally crossed for sex and age (6 versus 7 years) who were classified as 'average-memory' in that they had achieved the average score for their age in Session 1 (across letters and numerals combined). Our second test session provided two scores: immediate consistency in Session 2 (i.e. the number of consistent colours across 10 s in Session 2) and delayed consistency [i.e. the number of consistent colours across 12 months (see Results section)]. Synaesthetes were identified as those 'potential synaesthetes' who continued to outperform controls in immediate consistency in Session 2, as well as being significantly more consistent over 12 months (in delayed consistency) than their peer-group had been over 10s (in immediate consistency in Session 1). Hence, we 'stacked the deck' against our synaesthetes by holding them to a considerably higher standard than their peers in terms of the time across which their high consistency must be achieved (12 months versus 10s). 'Potential synaesthetes' who failed these criteria were classed as 'highmemory' non-synaesthetes, in that their superior scores in immediate consistency in Session 1 likely arose simply from a superior memory span for that task.

A three-part verbal questionnaire was submitted to synaesthetes and high memory participants at the end of Session 2. Participants were asked about the automaticity, consistency and certainty of their colour experiences, and these questions have been shown to distinguish between synaesthetes and non-synaesthetes in adult studies (Simner et al., 2006). In the verbal questionnaire, children gave one of six responses (never, almost never, not very often, sometimes, often, always) to each of the three questions below:

- (1) When playing the game, did you know for certain what the colours should be?
- (2) When you see or think about letters or numbers, do you automatically have a colour for them?
- (3) Do your colours for letters and numbers stay the same?

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These questions represent a modification of an earlier six-part adult questionnaire (Simner *et al.*, 2006) which we minimally reworded for our child participants (e.g. referring to the experiment as 'the game') and from which we excluded three questions that would have required a linguistic competence beyond the abilities of our child participants (e.g. questions involving double negatives: 'There were never not enough colours to choose from'). Children's responses (*never*, *almost never*, *not very often*, *sometimes*, *often*, *always*) were scored 0–5.

Results

Prevalence

Children made grapheme-colour selections twice in each of our two test sessions and we label these: Selections 1a/b; Selections 2a/b. Our results are based on three different analyses of consistency: immediate consistency in Session 1 (Selections 1a versus 1b); immediate consistency in Session 2 (Selections 2a versus 2b); and delayed consistency (Selections 1a versus 2a; or Selections 1b versus 2b). Note that because children gave four colours in total for each grapheme, delayed consistency could, in theory, be calculated by any of four different comparisons (Selections 1a versus 2a; 1b versus 2b; 1a versus 2b; 1b versus 2a). We conservatively discounted matches across first and second presentations (e.g. Selections 1a versus 2b), and conservatively disallowed high consistency to be achieved by a mix of consistency from within first presentations and within second presentations (e.g. four consistently coloured graphemes in Selections 1a versus 2a, and three consistently coloured graphemes in Selections 1b versus 2b does not equal a score of seven). For further conservativeness, we discounted any matches across light and dark versions of the same colour (e.g. 'light green' versus 'dark green' was considered a mismatch).

We first assessed immediate consistency in Session 1 for letters (/26) and for numerals (/10), for all 615 children. Girls and boys performed equivalently to each other, both for letters [respective means = 2.6 and 2.4; SDs = 2.1 and 1.9; F(1,611) = 2.3, P > 0.05] and numerals [respective means = 1.0 and 0.9; SDs = 1.0 and 0.9; F(1,611) = 1.2, P > 0.05], and there was no interaction with age [F(1,611)<1 for letters; F(1,611)<1 for numerals]. However, 7-year olds out-performed 6-year olds [for both letters: respective means = 2.8 and 2.3; SDs = 2.3 and 1.7; ([(1,611) = 12.5, P < 0.001] and numerals: respective means = 1.1 and 0.9; SDs = 1.1 and 0.9; F(1,611) = 4.2, P < 0.05)]. Hence, our data was analysed separately for children age 6 versus 7 years, but was collapsed across the sexes. The selection criterion for our 'average-memory controls' was a consistency of three or four out of 36 graphemes in immediate consistency in Session 1, since the population mean was 3.2/36 and 3.9/36 for children age 6 and 7 years, respectively. There were 47 'potential synaesthetes', whose colours were significantly more consistent (in Z-scores at P < 0.05) than the mean for their entire agegroup, in their colours for letters (Ss = 20), for numerals (Ss = 21) or for both (Ss = 6). In consistency for all 36 graphemes, the mean

score for the entire population (n = 615) was 9.7% (3.5/36) while the mean for 'potential synaesthetes' was 24.2% (8.7/36).

Of these 47 'potential synaesthetes', nine continued to score significantly highly (in Z-scores at P < 0.05) in their immediate consistency in Session 2 compared to age-matched controls for letters (control means: age 6 = 3.4; age 7 = 3.0) and/or numerals (control means: age 6 = 1.5; age 7 = 1.0), as well as being significantly more consistent in delayed consistency over 12 months compared with age-matched peers over 10 s (immediate consistency in Session 1). For added conservativeness, we removed one participant (C.B.) who may have used a verbal learning strategy not reminiscent of genuine synaesthetic reports. Forty-three percent of her consistent colour choices arose from pairing each colour term to its initial grapheme or phoneme (e.g. r = red), compared to only 16.5% (SD = 11.1) for the remaining eight participants in this group (Z = 2.4, P < 0.01) and this verbal strategy is known to be employed by non-synaesthetes more than synaesthetes (Simner et al., 2006). Moreover, C.B. was the slowest participant in both test sessions, and she took four times longer to respond than the population mean in Session 1 (C.B.'s mean RT = 15.0 s; versus population mean = 3.5 s; SD 1.8; Z=6.4, P<0.001) and almost three times longer in Session 2 (C.B.'s mean RT = 10.7 s; versus population mean = 3.8 s; SD 1.7; Z=4.1, P<0.001). Both factors indicate a strategy or rehearsal of a type not found for genuine synaesthetes, and not found in our remaining synaesthete participants (see below). Hence we classed eight children as synaesthetes and the remaining 39 as high-memory non-synaesthetes. Our n = 8 group comprised three synaesthetes for letters only (females age six and seven; male age six), two for numerals only (females age six) and three for both (female age six; males age six and seven). Figure 1 shows the colours chosen by example synaesthete JC across 12 months, and by an average age-matched control over 10 s.

Across 12 months, synaesthetes were approximately four times more consistent (42.0%) in their mean of letters and numerals combined than their peers had been over only 10 s (9.7%). For synaesthetes (n = 8), the mean score in delayed consistency over 12 months was 11.3/26 for (n=6) letter-colour synaesthetes (versus 2.5/26 consistent for all 615 children over 10s) and 3.8/10 for (n=5) digit-colour synaesthetes (versus 1.0/10 for all 615 children over 10 s). The superior performance of synaesthetes was all the more remarkable given that they did not incur a time penalty in the retest compared to high-memory children (whom they significantly outperformed; mean delayed consistencies 42.0 and 14.2%, respectively). Mean RTs in Session 2 were 4.8s for synaesthetes and 4.2 s for high-memory participants (Bonferroni test P>0.05). A three-way group difference emerged, however [F(2,84) = 329.9, P < 0.05], because average-memory controls took significantly less time to respond (mean RT = 3.3 s) than high-memory non-synaesthetes (Bonferroni test P = 0.05) and were also near-significantly faster than synaesthetes (Bonferroni test P < 0.06). This is reflected in the mean delayed consistency of average-memory controls, which was only 10.4%.

Questionnaire scores (out of five) from synaesthetes and highmemory non-synaesthetes also lend supports for our classifications. These indicate that synaesthetes' colours were selected with greater certainty (respective means: 2.8 and 2.5), are generated more automatically (respective means: 3.1 and 1.6) and are Childhood synaesthesia Brain 2009: 132; 57–64 | 61

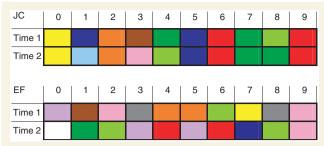


Fig. 1 Colour choices in the first presentation of numerals 0–9 in Sessions 1 and 2, for synaesthete JC (over 12 months) and age-matched control EF (over 10 s).

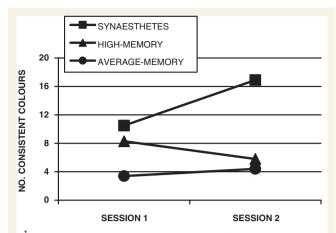
felt to be more consistent over time (respective means: 3.0 and 1.3), and similar findings have been shown to distinguish between synaesthetes and non-synaesthetes in adult studies (Simner *et al.*, 2006). A mixed ANOVA, crossing group (synaesthetes versus high-memory participants) and question (questions 1–3) shows these differences to be significant: there was a main effect of group [F(1,45)=5.3, P<0.05], no main effect of question [F(2,90)>1] and no interaction [F(2,90)=1.8, P>0.05].

Given the size and female/male composition of our sample population, we find the prevalence of synaesthesia in children ages 6–7 years to be 1.3%, with a female: male ratio of 1.6:1. Our small sample size of synaesthetes means the female skew cannot be verified statistically, although our trend fits with two of three adult investigations that also established prevalence via consistency in non-self-referred samples (i.e. 1.1:1, Simner et al., 2006; 1.7:1, Ward and Simner, 2005; but c.f. 0.9:1, Simner et al., 2006).

Development

We examined the number of new consistent associations acquired by grapheme-colour synaesthetes across 12 months, and compared this with improvements found from normal development in children with average and superior memories for comparable stimuli. Synaesthetes are assumed to have acquired a synaesthetic association (e.g. a = red) if this association is stable enough to remain consistent in an immediate retest. Hence, by comparing a synaesthete's immediate retest consistency in Session 1 (Selection 1a versus 1b) with her immediate retest consistency in Session 2 (Selection 2a versus 2b), we are able to determine how many stable associations were acquired over the 12 months that elapsed between the two sessions. Figure 2 shows (n=8) synaesthetes' immediate consistencies for 36 graphemes in each of the two test sessions, as well as the performance of high-memory non-synaesthetes (n=39), and average-memory non-synaesthetes (n=40).

Statistical tests allow us to compare the developmental trajectories of each group. First, we examined the development of consistent colours over time, by comparing mean scores for *immediate consistency in Session 1* with those in Session 2, as shown in Fig. 2. A 2×3 mixed design ANOVA, crossing group (synaesthetes, high-memory group, average memory group) and test session (Sessions 1 and 2) showed main effects of group [F(2,84) = 56.0, P < 0.001] and session [F(1,84) = 12.9, P < 0.01] as well as an interaction [F(2,84) = 28.9,



¹ Although synaesthetes' colours are highly specific (e.g., Ward *et al.*, 2005) and could be closely approximated with sufficient time and effort using an extensive palette comprising many thousands of colours (as we have done elsewhere; e.g. Simner *et al.*, 2006), prior studies have shown that a presentation of our condensed palette allows for a successful assessment of synaesthesia that is practical for large numbers of participants, or for those with limited attention (Day, 2005; Simner *et al.*, 2006). When faced with the core palette, synaesthetes are systematic in their choices, while non-synaesthetes are significantly more random (Simner *et al.*, 2006).

Fig. 2 Mean number of consistent colours selected by synaesthetes, high-memory non-synaesthetes and averagememory non-synaesthetes, in the immediate retest of 36 graphemes in Sessions 1 and 2.

P < 0.001]. Planned comparisons (with the Bonferroni test) showed that synaesthetes' immediate consistency improved across the two test sessions (from 29% to 47%) at a sharper rate than the average memory group (who improved from 9% to 12%; P < 0.001). Repeated measure t-tests show that while both synaesthetes and average-memory controls improved their immediate consistency across test sessions [t(7) = 4.1, P < 0.01; t(39) = 2.6, P < 0.02], the high-memory group actually fell slightly [from ~23% to 16%; t(38) = 4.3, P < 0.001]. Planned comparisons with the Bonferroni test showed that the numerical advantage for high-memory participants was non-significantly different (P > 0.05) to average-memory participants in Session 2. Finally, a comparison of synaesthetes' and high memory participants' immediate consistencies in Session 1 showed that-although we were unaware of this at the timesynaesthetes were already significantly out-performing the high memory group in our first test session (see Fig. 2; 29% versus 23% respectively; Bonferroni test P < 0.05). Both these groups were necessarily more consistent than average-memory controls, since this reflects the criterion on which synaesthetes and high-memory participants were originally selected (i.e. that they were superior performers in Session 1).

In summary, synaesthetes were already significantly outperforming both their average- and high-memory peers in Session 1, and they improved at a significantly sharper rate than either group. Both synaesthetes and average-memory controls significantly improved their immediate consistency across the test sessions, while the high-memory group fell slightly: in Session 2 they were numerically, but not significantly different from the average-memory group. We assume therefore that a subset of children achieved high-memory status in Session 1 by using a mnemonic strategy they failed to apply in Session 2.

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Conclusions

Our study represents the first assessment of synaesthesia in a randomly sampled child population, illustrating the prevalence of grapheme-colour synaesthesia and its development in comparison to age-matched non-synaesthetes. A perfect (or near-perfect) consistency of synaesthetic mappings over time has been judged the 'behavioural hallmark' of adult synaesthesia (Rich *et al.*, 2005), but our data show that this is a trait that develops over time. In our study, young child synaesthetes were required to outperform their peers in what would be a hugely challenging task for non-synaesthetes (i.e. surprise recall over 12 months, compared to 10s from peers) and we also sought corroborating evidence of synaesthesia from quantitative questionnaire scores, and diverging developmental patterns over time. This allowed us to identify child synaesthetes, even where the adult hallmark of 100% consistency was not yet present.

Our prevalence of grapheme-colour synaesthesia at 1.3% implicates over 170 000 children age 0-17 in the UK alone, and over 930 000 in the USA and suggests that the average primary school in England and Scotland (n = 168 pupils) contains 2.2 graphemecolour synaesthetes at any time, while the average-sized US primary school (n = 396 pupils) contains 5.1 (National Statistics: All people Part 1; General Register Office for Scotland: Table 1; Northern Ireland Statistics and Research Agency: Table S001; US Census Bureau: Table DP-1; Schools and Pupils in England: January 2006; Pupils in Scotland 2006; IES National Center for Education Statistics. Table 5). This is an estimate for graphemecolour synaesthesia only, while the combined figure for all variants of the condition is likely to be much higher (see Day, 2005 for the 50+ variants so far identified). Moreover, our prevalence for grapheme-colour synaesthesia is a necessary underestimate, because our methodology relies on the assumption that synaesthetes are consistent in their grapheme-colours, while our study has shown that this consistency is not yet fully achieved in young children. Adult synaesthetic associations are consistent at ~90-100% (Dixon et al., 2000; Smilek et al., 2002a, b; Rich et al., 2005; Simner and Ward, 2006; Simner et al., 2006; Yaro and Ward, 2007) and adult prevalence of grapheme-colour synaesthesia has been established at 2% from similar methodology (Simner et al., 2006). Hence, our childhood prevalence figure should be taken as a reliable lower cut-off, and indeed, there is reason to believe that synaesthesia in younger age groups may in fact be more common than in adults (Flournoy, 1893; Werner, 1957). In anecdotal accounts, adults have reported synaesthesia in childhood that died out, while the reverse pattern (i.e. developmental synaesthesia spontaneously appearing in adulthood) is not reported. This same pattern is hinted at by the current study, where our ratio of synaesthetes ages 6 versus 7 years was 2.5:1, although this cannot be verified statistically from our small sample size.

Most adult synaesthetes report that they have had synaesthesia for as long as they can remember, but our study demonstrates that grapheme-colour synaesthesia emerges along a developmental path. Our research has shown this development in action, since synaesthetes acquired on average 6.4 new grapheme-colour

associations during the course of our 12-month longitudinal testing. Synaesthetes had, on average, 10.5 stable grapheme-colour associations age 6/7 years, but 16.9 stable associations in the same test age 7/8 years. The trajectory of this acquisition was more rapid than expected from memory developments in nonsynaesthetic children for comparable stimuli, and these gains suggest that grapheme-colour synaesthesia may rely on, or endow, exceptional mechanisms in this domain and that these may be implicated in other assets associated with the condition. It is not known whether the developmental pattern shown by our synaesthetes (i.e. 6.4 new coloured graphemes per year) represents a linear acquisition, or whether greater gains are made in later years. A linear acquisition would predict that synaesthetes will have acquired all 36 grapheme-colour associations only by ~10/11 years, and our lab is currently tracking the development of this group to follow their transition into adult-like consistency. This will also allow us to establish independent measures in abilities for synaesthetes and controls, since the current study classified non-synaesthetes as 'high' or 'average memory' only with respect to how they performed in a task of matching colours to graphemes. Such future research will be important for determining exactly what implications synaesthesia might have for children's general cognitive performance in schools. Our ongoing research is also examining the order with which grapheme-colour associations are acquired: one intriguing recent study of nonsynaesthetes suggests that colours may first be assigned to letters that have naturally occurring shapes (e.g. o and x; Spector and Maurer, 2008) while studies on adult synaesthetes suggest that early mappings may also favour graphemes that are frequently occurring within language (Simner et al., 2005).

Our data show that grapheme-colour synaesthetes were already significantly outperforming their 'high-memory' peers in Session 1, suggesting that the roots of the synaesthetic experience are already emerging by age 6 years. This coincides with developmental stages that are relatively recent to the acquisition of alphabet and number sequences. The average 3-year-old child can recite sequenced numbers to at least six (Wynn, 1990, 1992), although meaningful understanding lags considerably behind this rote learning. By the end of the fourth year, most children have acquired the meanings of the smallest counting words, and learn to map higher numbers onto numerocities over the following year (Wynn, 1990, 1992). In alphabet acquisition, normally developing children know an average of six letters by age 3;9 years (Gallagher et al., 2000) and it takes until 5;7 years before the average middle class English-speaking child can name 18 or 19 of the 26 letter sequences from their uppercase and lowercase forms (Treiman et al., 1998). Hence the youngest children in our study (in the 6-year group) will have acquired the complete alphabetic sequence only very recent to their testing in Session 1. Nonetheless, a subset of those units were already bound to consistent cross-modal synaesthetic sensations for our child synaesthetes, and it is possible that some graphemes are accompanied by a concurrent mapping of a colour at the very initial stages. For other graphemes, colour may be stimulated only after some delay (perhaps once familiarity with the grapheme is more developed). The fact that our synaesthetes aged 8 years did not have a full compliment of consistent synaesthetic colours, even though Childhood synaesthesia Brain 2009: 132; 57–64 63

their alphabet would have been known in full, suggests some temporal gap between a complete knowledge of inducers, and the consistent neurological mapping of inducers to their sensory cross-modal concurrents.

Since grapheme-colour synaesthesia is intimately tied to knowledge of culturally acquired symbols, it could not have existed as such in infancy (but may have been present in other forms; see Duffy 2001). Although neuroimaging data suggests that perisylvian language areas are already well-organized for the perception of language even in pre-verbal infants (Dehaene-Lambertz et al., 2006a, b), it is only later, once specific linguistic items have been acquired (i.e. letters and numerals), that the types of representations required to drive grapheme-colour synaesthesia can be established. A small number of imaging studies have examined the development of reading skills in young children, and suggest a reorganization of the ventral visual areas in response to the pressures of learning to read (see McCandliss et al., 2003). An area of the left fusiform gyrus (the VWFA) becomes particularly specialized for reading functions (for reviews see McCandliss et al., 2003; Dehaene and Cohen, 2007) across a range of language backgrounds (e.g. Bolger et al., 2005) and this specialization appears to arise from developments in reading proficiency, rather than maturation per se (Shaywitz et al., 2002). Since the VWFA is adjacent to colour selective regions implicated in synaesthetic colour perception (Nunn et al., 2002; Hubbard et al., 2005; Rouw and Scholte, 2007), it is the development of the VWFA that would make a plausible candidate for observation during synaesthesia's 'growth'. Interestingly, recent work (Balsamo et al., 2006) indicates that this specialization for reading in the VWFA may be pre-dated by an early sensitivity to auditory input. If this pre-existing responsiveness to auditory language plays a role in the specializaton of the VWFA for reading in later life, there may be some contiguity in the development of coloured graphemes from coloured speech. That literacy acquisition is parasitic on the system for spoken language has been seen in other regions, also (see van Atteveldt et al., 2004 for evidence of the integration of speech sounds and letters in the superior temporal gyrus, for example) but connections between spoken representations and letter forms are particularly linked to synaptic growth in the VWFA. Such regions may therefore provide interesting areas of study, although any direct observation of synaesthesia's neural development would require the difficult task of very early identification of randomly sampled child synaesthetes, as well as improved imaging techniques in early childhood (Dehaene and Cohen, 2007).

In summary, we have presented a study of developmental synaesthesia based on a randomly sampled population of child grapheme-colour synaesthetes and age-matched controls assessed in longitudinal testing. We have provided the first direct evidence that this variant of synaesthesia evolves over time following environmental exposure to learned linguistic units, notwithstanding its likely genetic roots. We have shown, too, that this synaesthesia develops along an exceptional trajectory for grapheme-colour pairings, and that adult states showing consistent inducer-concurrent mappings are achieved only through stages of incremental acquisition.

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