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The prevalence of synaesthesia depends on early language learning

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ABSTRACT

According to one theory, synaesthesia develops, or is preserved, because it helps children learn. If so, it should be more common among adults who faced greater childhood learning challenges. In the largest survey of synaesthesia to date, the incidence of synaesthesia was compared among native speakers of languages with transparent (easier) and opaque (more difficult) orthographies. Contrary to our prediction, native speakers of Czech (transparent) were more likely to be synaesthetes than native speakers of English (opaque). However, exploratory analyses suggested that this was because more Czechs learned non-native second languages, which was strongly associated with synaesthesia, consistent with the learning hypothesis. Furthermore, the incidence of synaesthesia among speakers of opaque languages was double that among speakers of transparent languages other than Czech, also consistent with the learning hypothesis. These findings contribute to an emerging understanding of synaesthetic development as a complex and lengthy process with multiple causal influences.

1. Introduction

1.1. Learning and synaesthesia in the early years of research

During the first wave of scientific interest in synaesthesia, many researchers argued for a strong relationship between learning and synaesthesia (cf. Calkins, 1893, 1895; Flournoy, 1893; Galton, 1883; Jewanski, Simner, Day, & Ward, 2011; Marks, 1975). Mary Calkins, for example, argued that the unusual conscious experiences of adult synaesthetes develop in childhood "largely due to attention and to cultivation" of associations that the child has found "useful and pleasant" (Calkins, 1893). For example, when discussing number-form synaesthesia, in which numbers are experienced in a complex spatial arrangement in peripersonal space, Calkins argued that "…it is to the highest degree probable that most of these forms originate in the self-helping, topographical imagination of children introduced to the intricacies of number and word series" (Calkins, 1895). Calkins allowed that childhood synaesthesia might develop for other reasons (such as unusual neu-

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rophysiology), but suggested that even if this were the case, its preservation into adulthood depended on its being cultivated for learning (Calkins, 1893). Here we present a large-scale study that seeks to test the claim that childhood learning plays a causal role in the development or preservation of synaesthesia, by correlating the presence of synaesthesia in adults with their childhood language learning experiences.

1.2. A modern turn to genetics and rejection of learning

After more than fifty years of neglect (with a few notable exceptions such as Marks, 1975), synaesthesia re-emerged as a subject of serious scientific inquiry towards the end of the 20th Century (beginning with Cytowic, 1989a). At this time straightforward genetic accounts of its development gained prominence for a number of compelling reasons. The very strong tendency of synaesthesia to run in families, which had been reported for well over a century (Galton, 1883), was reconfirmed (Barnett et al., 2008; Baron-Cohen, Burt, Smith-Laittan, Harrison, & Bolton, 1996; Baron-Cohen, Harrison, Goldstein, & Wyke, 1993), and female:male ratios of 3:1 or higher were reported (Barnett et al., 2008; Baron-Cohen et al., 1993, 1996; Cytowic, 1989a). Furthermore, researchers at the time estimated the prevalence of synaesthesia to be 0.05% or lower (Baron-Cohen et al., 1996; Cytowic, 1989a, 1997; Ramachandran & Hubbard, 2001a). This overall pattern was consistent with synaesthesia being caused by a relatively rare x-chromosome-linked dominant mutation that is lethal to a substantial proportion of male embryos, similar to genetic diseases such as Rett syndrome (Bailey & Johnson, 1997).

While support for a genetic account was mounting, support for learned synaesthesia was dwindling. Many of the researchers responsible for the modern resurgence of interest in synaesthesia found it implausible that synaesthetic associations could be learned (cf. Cytowic, 1989b; Maurer, Gibson, & Spector, 2012; Ramachandran & Hubbard, 2003). Experiments using associative learning paradigms failed to produce any evidence of synaesthetic experiences (Elias, Saucier, Hardie, & Sarty, 2003; Meier & Rothen, 2009). Furthermore, synaesthetes themselves often describe their synaesthesia as being part of their earliest memories, and as being automatic, involuntary, and stable throughout their lives, characteristics that do not seem entirely compatible with a strong role for learning. Finally, researchers were at pains to establish that synaesthetic experiences (known as *concurrents*) were *genuinely perceptual*, and argued against critics who said that synaesthetic associations were learned (e.g. Ramachandran & Hubbard, 2001a, 2001b). These researchers and critics appeared united in assuming that people could not *learn* to have non-veridical perceptual experiences such as seeing the letter "A" as red. Researchers acknowledged that most stimuli that trigger synaesthesia (known as *inducers*), such as letters, were learned cultural artefacts. But aside from this prerequisite, synaesthesia was understood as fundamentally unrelated to learning.

1.3. The return of learning?

Recent research suggests a more nuanced view. While inheritance certainly plays a role in the development of synaesthesia, there is no single "synaesthesia gene": two genetic studies have implicated different loci (Asher et al., 2009; Tomson et al., 2011), and there is incomplete concordance in monozygotic twins (Bosley & Eagleman, 2015). Larger-scale surveys found a weak sex bias or none at all (Rouw & Scholte, 2016; Sagiv, Simner, Collins, Butterworth, & Ward, 2006; Simner & Carmichael, 2015; Simner et al., 2006; Ward & Simner, 2005), suggesting that previous studies had not detected a genuine sex difference, but merely found that women were more likely to respond positively to advertisements for synaesthesia studies. Furthermore, they found an overall incidence for synaesthesia of 4.4% (Simner et al., 2006), and an incidence of grapheme-colour synaesthesia of 1–1.4% (Carmichael, Down, Shillcock, Eagleman, & Simner, 2015; Simner & Carmichael, 2015; Simner et al., 2006). This is far less than very early reports (e.g. Calkins, 1895), but is orders of magnitude larger than hereditary illnesses such as Rett syndrome.

The evidence in favour of a learned aspect of synaesthetic development has also strengthened. Every known behavioral marker of synaesthesia can be produced in previously non-synaesthetic adults given associative training (Bor, Rothen, Schwartzman, Clayton, & Seth, 2014), trained participants report experiences similar to those reported by "natural" synaesthetes (Bor et al., 2014; Howells, 1944; MacLeod & Dunbar, 1988), and it is now established that many grapheme-colour synaesthetes learned their letter colours from childhood toys (Witthoft & Winawer, 2006; Witthoft, Winawer, & Eagleman, 2015). Furthermore, whatever adults may remember of their childhood synaesthesia, grapheme-colour synaesthetic associations are not stable in early life, and indeed their development continues for many years, at least into late childhood (Simner & Bain, 2013; Simner, Harrold, Creed, Monro, & Foulkes, 2009).

Thus it is not surprising that the idea that learning of some kind plays an important role in synaesthetic development has returned to prominence (cf. Asano & Yokosawa, 2013; Blazej & Cohen-Goldberg, 2015; Bor et al., 2014; Jürgens & Nikolic, 2012; Mroczko-Wasowicz & Nikolic, 2014; Price & Pearson, 2013; Simner, 2007; Simner & Bain, 2013; Simner et al., 2009; Ward & Simner, 2003; Ward, Simner, & Auyeung, 2005; Watson, Akins, & Enns, 2012; Watson, Akins, Spiker, Crawford, & Enns, 2014; Watson, Blair, Kozik, Akins, & Enns, 2012; Witthoft & Winawer, 2006, 2013; Witthoft et al., 2015; Yon & Press, 2014). The appropriate question is not "does learning influence synaesthetic development?", but rather "to what extent and in what manner does this influence occur?".

1.4. A developmental learning hypothesis of synaesthesia

One answer suggested by Watson et al. (2014) is that synaesthesia develops, at least in part, because concurrents are useful for gaining mastery over the inducer domain. For example, the internally-generated letter colours of grapheme-colour synaesthesia might be exploited to master the multitude of tasks involved in becoming literate (cf. Asano & Yokosawa, 2013). Genuinely coloured letters can aid children in these tasks: e.g. differentiating hard-to-discriminate letters (Jones, 1965), recognizing words in unfamiliar scripts (Otto, 1968), learning new spellings (Jones, 1968), and identifying crowded letters (Põder, 2007). Could synaesthetic letter colours play similar roles? There is no direct evidence concerning this hypothesis, but we do know that synaesthetic concurrents can aid memory (for reviews, see Rothen, Meier, & Ward, 2012; Watson et al., 2014), can be used in difficult category learning tasks (Watson, Blair, et al., 2012), and can assist with the implicit learning of artificial grammars (Rothen et al., 2013).

Consider also the developmental timelines of colour perception, literacy, and synaesthesia: children typically learn to recognize and categorize colours from ages 4–7 (Bornstein, 1985); shortly after this they begin a many-year process of developing and refining reading and writing skills; and grapheme-colour synaesthetic associations develop over this same time period, beginning prior to age 6 and continuing past age 11 (Simner & Bain, 2013; Simner et al., 2009). We suggest that during this period, some children use their newly-acquired ability to categorize colour as an unorthodox aid in mastering the domain of letters and words. This association of colours with letters helps them both to perceive and understand a new orthography.

Our learning theory claims that synaesthetic associations are useful aids for many different learning challenges, and that this is why the inducers that trigger synaesthetic experiences are almost always members of culturally-defined category structures learned with some difficulty in mid-to-late childhood, such as letters, time units, and aspects of music (Day, 2005; Rich, Bradshaw, & Mattingley, 2005; Simner et al., 2006). Conversely, the fact that inducers are usually members of learned categories suggests that synaesthesia rarely starts with "innate" inducers, or that associations with these inducers do not survive into adulthood.

The learning theory is consistent with the growing realization that multisensory encoding of stimuli enables superior recognition, even if subsequent presentations are unisensory (cf. Murray & Thelen, 2013), and with the use of mnemonic techniques such as the *method of loci* to enable extraordinary feats of memory (Maguire, Valentine, Wilding, & Kapur, 2003). However we stress that synaesthetic concurrents have the potential to assist with far more than recall. In synaesthesia, each member of an inducer category is placed into a multi-dimensional perceptual/conceptual space (such as colour), which provides multiple ways to map these inducers, and the relationships between them. Thus the structure of this concurrent space, which the child is already familiar with, can be used to represent multiple aspects of the inducer domain, which the child is struggling to learn. For example, the synaesthetic colours of letters can become shorthand for orthographic or phonological properties, in different ways for different languages (cf. Asano & Yokosawa, 2011, 2012; Barnett, Feeney, Gormley, & Newell, 2009; Rouw, Case, Gosavi, & Ramachandran, 2014; Simner, Hung, & Shillcock, 2011; Simner et al., 2005; van Leeuwen, Dingemanse, Todil, Agameya, & Majid, 2016; Watson, Akins, et al., 2012). In brief synaesthetic concurrents encode first- and second-order properties of their inducers (for an extensive review of this issue, see Watson et al., 2014). Thus we agree with Marks' characterization of synaesthesia as an *adjunct to verbal cognition*, a "shorthand" that is "both less abstract and more dense in informational content", and that is used to "summarize important cognitive distinctions in a convenient and economical way" (Marks, 1975).

This theory is by no means inconsistent with genetic or neurological influences on synaesthesia. Such factors may well predispose the development of synaesthesia, or even be necessary (although they are certainly not *sufficient*) for this development. We only posit that the utility of synaesthetic concurrents for mastering certain learning tasks is a major reason for their development, irrespective of any other factors.

1.5. Orthographic transparency as an influence on synaesthetic development?

The learning theory is far from definitively established. It is no longer sensible to deny that synaesthesia develops, at least in part, via associative learning (cf. Yon & Press, 2014), but there is as yet no direct evidence that it develops *for* learning. Such evidence might be found in cases where the difficulty of a childhood learning task differs between groups. The development of literacy is one of these cases: writing systems that are *orthographically transparent, consistent*, or *shallow* (have a close to one-to-one mapping between graphemes and phonemes) are easier to learn than writing systems that are *orthographically opaque*, *inconsistent*, or *deep*. Children who are native speakers of languages with transparent writing systems can read both words and non-words more accurately and quickly, have better letter knowledge, and develop basic literacy faster than native speakers of languages with opaque systems (there is a large literature on this, see, e.g. Caravolas, 2004; Carrillo, Alegría, & Marín, 2013; Ellis et al., 2004; Katz & Frost, 1992; Viise, Richards, & Pandis, 2011). For instance, native Czech-speaking children show superior performance to native English-speaking children on tests of reading non-words, spelling them, and isolating phonemes within them, despite the Czechs having received substantially less instruction in reading and writing, and this is understood to be due to the highly transparent Czech orthography and the highly opaque English one (Caravolas, 2004; Caravolas & Bruck, 1993). Thus if grapheme-colour synaesthesia develops as a strategic aid to literacy, native speakers of languages with transparent writing systems such as Czech ought to have a lower incidence of synaesthesia than native speakers of languages with opaque writing systems such as English, because mastering a transparent orthography is easier and so unorthodox strategies are less needed. Our study seeks to test exactly this expectation.

1.6. How to determine the prevalence of synaesthesia

Previous estimates of prevalence have been wildly variable. As noted in 1.2, modern researchers initially estimated synaesthesia's prevalence at less than 0.05% (Baron-Cohen et al., 1996; Cytowic, 1989a, 1997; Ramachandran & Hubbard, 2001a; Rich et al., 2005). These studies used a Test of Genuineness for synaesthesia, which confirms that reported synaesthetic associations are much more consistent across time than those of non-synaesthetes (Asher, Aitken, Farooqi, Kurmani, & Baron-Cohen, 2006; Baron-Cohen et al., 1996). However these estimates are far too conservative, as methodological limitations meant that the target populations of these studies were greatly over-estimated. When advertisements were placed in newspapers, for example, the population could only be estimated based on the total newspaper circulation, which is clearly much larger than the number of individuals who both noticed the advertisement and would have been willing to reply (Simner & Carmichael, 2015; Simner et al., 2006).

In contrast, earlier surveys either canvassed all members of a particular population or sampled randomly within it, finding prevalences of various forms of synaesthesia of 5% or higher (Calkins, 1893, 1895; Domino, 1989; Flournoy, 1893; Galton, 1880, 1883; Rose, 1909; Shindell, 1983; Ulich, 1957). However these studies did not use rigorous Tests of Genuineness, and since false positive responses to questions about synaesthesia are extremely common (Simner, 2012), these estimates have been criticized as too liberal (Simner & Carmichael, 2015; Simner et al., 2006).

More recent studies have avoided both conservative and liberal biases by using random sampling of a large number of individuals within a particular population, supplemented by rigorous consistency tests. These have found the prevalence of any type of synaesthesia to be approximately 4% (Simner et al., 2006), and the prevalence of grapheme-colour synaesthesia in particular to be 1–2% in most populations (Carmichael et al., 2015; Rothen & Meier, 2010b; Simner & Bain, 2013; Simner et al., 2006, 2009), estimates that lie comfortably between the conservative and liberal estimates of previous studies. Interestingly, one of these studies finds the prevalence of grapheme-colour synaesthesia to be 7% among fine arts students (Rothen & Meier, 2010b), suggesting that group differences such as those our study looks for can indeed be found.

Most recently, a survey asked a representative sample of the entire population of the Netherlands about their synaesthetic experiences (Rouw & Scholte, 2016). Various constraints meant that the study could not use consistency tests. Instead, any report of a form of synaesthesia was followed up with a request for five examples of specific synaesthetic experiences, and these examples were rated as satisfactory or unsatisfactory by one of the researchers according to various criteria. The overall prevalence of synaesthesia was estimated to be 24%, which may be so high because standard consistency tests are ruling out genuine synaesthetes. Many individuals fail these tests, but nevertheless report conscious synaesthetic experiences that appear genuine (Simner, 2012), and many individuals who report synaesthetic experiences also report that these experiences are not always consistent (Niccolai, 2012). Thus reliance on consistency tests may lead to overly conservative estimates. In light of the results of the present study, other possible explanations of this discrepancy are given in 4.8 below.

1.7. The present study

Our study seeks to verify if synaesthesia is more common among native speakers of languages with more opaque writing systems. A survey on synaesthetic tendencies and early language experience was given to university students at Charles University and the University of Economics (hereafter CU) in Prague, Czech Republic and Simon Fraser University (hereafter SFU) in Burnaby, Canada. Respondents who reported synaesthetic experiences were invited to take online consistency tests to verify their synaesthesia (Eagleman, Kagan, Nelson, Sagaram, & Sarma, 2007), which have been verified to produce similar results as other tests of consistency that require much longer time periods (Carmichael et al., 2015).

The initial hypothesis was that differences in early language learning would be correlated with the presence of synaesthesia, specifically that native speakers of transparent languages would have lower rates of confirmed synaesthesia than native speakers of opaque languages. As the main languages in our sample were Czech and English, we expected lower rates of synaesthesia among native Czech than native English speakers. However we anticipated there would also be many native speakers of other languages, particularly in the SFU sample, thus allowing for further testing of the transparency hypothesis. We also hypothesized that other early learning experiences, such as learning a second language, might influence rates of synaesthesia, and so conducted exploratory analyses of such effects.

A large sample size was needed to generate sufficient power to test these hypotheses (N = 11,404), which was much greater than the largest previous surveys of synaesthetic tendencies (Carmichael et al., 2015; Simner & Carmichael, 2015; Simner et al., 2006). Thus a benefit of the present study is that its estimates of the rates of different types of synaesthesia are substantially more precise than previous estimates.

Finally, an additional benefit is that this study allows for a detailed investigation of the putative sex bias in synaesthesia, in particular of the claim that women are more likely than men to comply with experimenters' requests to participate in synaesthesia research (Simner & Carmichael, 2015; Simner et al., 2006; Ward & Simner, 2005).

2. Material and methods

2.1. Phase I: Paper survey

The paper survey (available in the online Supplementary Materials) asked participants to indicate their gender, handedness, languages spoken and the age of acquisition of these languages; if they had any of six common forms of synaesthetic experiences (grapheme-colour, weekday/month-colour, sound-colour, word-taste, number-form and grapheme-personality); if they thought they might have another form of synaesthesia that was not one of the six given options; if they remembered having used letters or numbers as characters in childhood stories; and if they were early readers or had particular difficulties with reading or writing. We also asked participants for their email address in order to invite them to participate in Phase 2. The survey was in English at SFU and in Czech at CU.

Surveys were handed out over the course of two years to students in undergraduate university classes, primarily from Arts and Social Sciences faculties. The methodology was the same at both universities: professors introduced the survey team, who gave a brief explanation of the research project while handing out the survey. Approximately 10 min after the surveys had been handed out, they were collected.

Assuming the rate of grapheme-colour synaesthesia among English speakers to be about 1% (as per Simner et al., 2006), a sample size of 4671 participants per group would be required to have acceptable Type I and Type II error rates (0.05 and 0.20, respectively), if the true rate in Czechs was about half that among English speakers, or 0.5%. Thus we tried to enroll at least 5000 people in each study, although the large number of non-native English speakers at SFU meant we had to enroll substantially more students (6405), and were still able to obtain only 4001 native English speakers in the time allotted to data collection (2 years). 233 participants who completed the survey in two classes (revealed by comparing email addresses) had their second survey removed from the data.

2.2. Phase II: Synesthesia battery

All survey participants who provided an email address and reported experiencing coloured letters, numbers, weekdays, months, or sounds, were contacted and invited to register for the online Synesthesia Battery (Eagleman et al., 2007), which includes consistency tests for many types of synaesthesia. Those who reported other forms of synaesthesia were not contacted, as we did not have easily administrable tests of consistency. As incentive for registering, each Battery participant was entered into a lottery for 5000 Czech Crowns or 500 Canadian dollars (approximately 300 and 500 USD, respectively) with a greater than 1% chance of winning. While the value of the reward was lower in the Czech Republic, the cost of living is also substantially lower, making the difference in incentive impossible to quantify. 352 CU and 309 SFU participants registered for the Battery (thus we ran 4 lotteries in each country).

The Synesthesia Battery website presents participants with a checklist of synaesthesia types. If they select a form of synaesthesia for which the Battery includes a consistency test, then they can take that test. In each test, participants are presented with each member of the inducer class (letters, weekdays, etc) in random order, and asked to select the colour that best matches their association with the inducer (full details are given in Eagleman et al., 2007). The entire test is repeated three times, and the pairwise similarity between the three colours assigned to each inducer is calculated as three distances in colour space. These three distances are summed together, and the average sum of these three distances across all inducers is that participant's consistency score for the test. We used a consistency threshold in which any score of less than 135 CIELuv units was taken as indicative of genuine synaesthesia, as suggested by a recent evaluation of the Battery (Rothen, Seth, Witzel, & Ward, 2013).

Table 1 gives a summary of participant numbers at different stages of the experiment.

Table 1
Summary of participation in each stage of the study. For both the SFU and CU groups, total participants and native English/Czech speakers are reported at each stage, as both raw numbers and percentages of the total.

	# SFU/# SFU Native English	% Total SFU /% SFU Native English	# CU/# CU Native Czech	% Total CU/% Total CU Native Czech
Returned paper survey	6405/4003	100.0/100.0	4999/4790	100.0/100.0
Reported synaesthesia	3023/1713	47.2/42.8	3262/3106	65.3/64.8
Reported synaesthesia & provided email	1820/1049	28.4/26.2	1861/1779	37.2/37.1
Registered for battery	309/227	4.8/5.7	352/346	7.0/7.2
Completed at least one test	213/153	3.3/3.8	290/286	5.8/6.0
Confirmed synaesthetic	143/106	2.2/2.7	215/211	4.3/4.4

3. Results

3.1. Sample sizes in Phase I and II

In total, 4999 CU students and 6405 SFU students returned completed surveys. Unfortunately, a precise count of the number of surveys handed out was not kept (which would have allowed an exact count of the number of individuals who chose not to participate), but non-participation was rare, and experimenters report that well over 95% of surveys were completed and returned in both universities (note that, as described in 2.1, the survey team was introduced at the start of the class by the professor and time was set aside to complete the survey, thus a high degree of compliance is not surprising). Thus slightly over 5000 (CU) and 6400 (SFU) surveys were handed out. This is likely to produce slight over-estimates of the prevalence of synaesthesia, as individuals with synaesthesia would presumably be more interested in participating in the study, however it is important to note that the vast majority of surveys were returned, and as discussed in 4.5, there are a number of influences in the other direction.

68.9% of CU and 61.0% of SFU survey respondents were women. 37.2% of CU and 28.4% of SFU respondents both provided an email address and reported experiencing one of the forms of synaesthesia that could be verified by the Synesthesia Battery. All these individuals were emailed and invited to participate in the online phase of the study, and just over 1/5 of them complied with this invitation: 7.0% of CU and 4.8% of SFU respondents.

3.2. Linguistic characteristics of the samples

The two groups had very distinct linguistic profiles. Students at CU were almost all native Czech or Slovak speakers (95.8% reported learning Czech or Slovak prior to age 2) who learned multiple other languages (mean number of languages reported: 3.5; all but 5 participants reported a second language) that were primarily European (56 distinct languages reported, 20 non-European languages), but were not brought up as native multilinguals (only 2% reported two or more languages before age 2). (Czech and Slovak are largely mutually intelligible languages that use almost identical alphabets.) The SFU sample was more heterogeneous, with many non-native English speakers (only 62.5% reported learning English before age 2), fewer languages spoken per participant (mean number of languages reported: 2.3, 22% did not report a second language), but a far wider range of languages (152 distinct languages reported, 117 of them non-European), and far more native multilinguals (21% reported two or more languages before age 2).

3.3. Higher rates of reported synaesthesia among Czechs

Each form of synaesthesia described on the survey was endorsed by 5.9–35.4% of native Czech/Slovak or English speakers (Fig. 1). All varieties of synaesthesia were endorsed by a greater proportion of CU than SFU respondents, and this was sig-

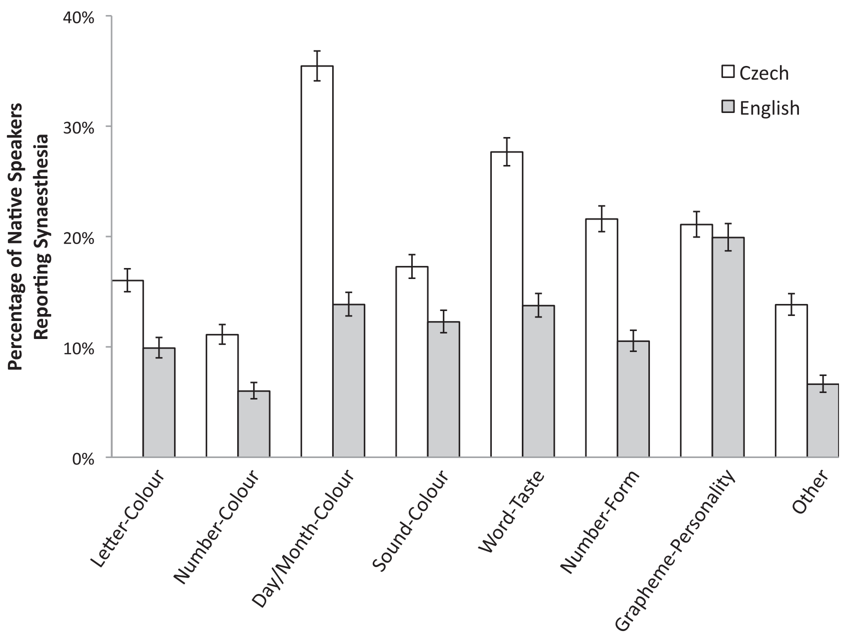

Fig. 1. Rates of reported synaesthesia among native Czech and English speakers. Error bars indicate 95% Wilson's C.I.s.

nificant (non-overlapping 95% Wilson's C.I.'s) in all cases except grapheme-personality synaesthesia. In total, 65.3% of CU and 47.2% of SFU respondents reported some form of synaesthesia.

The large majority of responses to the open-ended question were consistent with those reported by previous studies (e.g. Day, 2005), generally describing associations between particular learned categories (e.g. school subjects, seasons, tastes, bodily actions, car models, letters, music) and sensory experiences (e.g. colours, sounds, tastes, smells, shapes, sizes, textures, and temperatures), personifications of inanimate objects, or arrangements of the members of various category structures in peri-personal space.

Fisher's exact tests showed that reports of all forms of synaesthesia were positively associated with reports of all other forms in both samples (ϕs from 0.08 to 0.37, all *ps* < 0.001 after multiplying by 56 for Bonferroni correction). Tables S1 and S2 in the Supplementary Material present all these associations.

3.4. Higher rates of confirmed synaesthesia among Czechs

Proportions of synaesthesia confirmed by the Synesthesia Battery in Phase 2 ranged from 0.02% to 2.8% of native Czech/Slovak or English speakers (Fig. 2). As with rates of reported synaesthesia, absolute proportions were higher among native Czech/Slovak speakers than among native English speakers for all varieties of synaesthesia tested on the Battery, and these differences were significant in all cases except chord-colour and scale-colour synaesthesia. Clearly, our hypothesis of lower rates of grapheme-colour synaesthesia among Czechs was not supported. In total, 4.4% of Czech/Slovak and 2.7% of English participants were confirmed as having at least one form of synaesthesia.

Strikingly, a large number of synaesthetes were confirmed as having a form of synaesthesia that they denied having on the initial paper survey (Fig. 2). For instance, 22.0% (Czech/Slovak) and 38.0% (English) of confirmed letter-colour synaesthetes selected the response "No, I do not have experiences like this", when asked about associations between letters and colours on the survey (a similar result is reported by Simner et al., 2006). Since only participants who reported synaesthetic experiences were asked to register for the Synesthesia Battery, this means that all of these participants reported having at least one other type of synaesthetic experience. As noted in 2.2, the Battery begins with a checklist of synaesthesia types, and participants only take the tests for the types they have selected. This means that some of our confirmed synaesthetes gave answers to the Battery checklist that contradicted their responses on the initial paper survey.

Almost all forms of confirmed synaesthesia were positively associated with all other forms of confirmed synaesthesia in both samples (ϕs from 0.12 to 0.63, all *ps* < 0.001 after multiplying by 42 for Bonferroni correction). The only exceptions were that weekday-colour synaesthesia was not significantly associated with scale-colour synaesthesia among native English speakers ($p > 0.250$ after correction), and that chord-colour synaesthesia was not significantly associated with anything among native English speakers (all *ps* > 0.250 after correction) and only with month-colour and instrument-colour synaesthesia among native Czech/Slovak speakers ($\phi = 0.14$ and 0.25, $p < 0.001$ and $p = 0.003$, respectively; all other *ps* > 0.250). Given the tiny proportion of confirmed chord-colour and scale-colour synaesthesia in both samples (see Fig. 2), non-

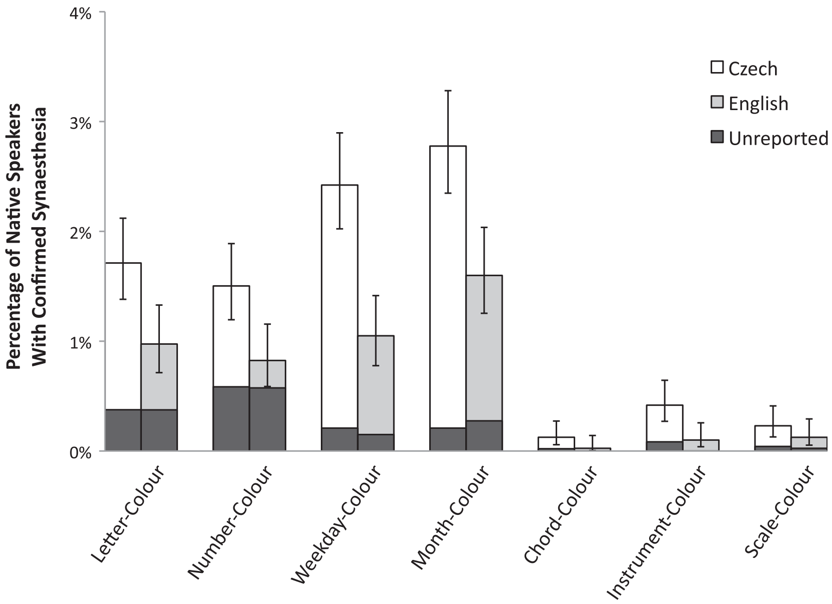

Fig. 2. Rates of confirmed synaesthesia among native Czech and English speakers. Light grey indicates subjects who initially reported not having a form of synaesthesia, but chose to take the consistency test and passed it. Error bars indicate 95% Wilson's C.I.s.

statistically-significant effects could well mask genuine associations. Tables S3 and S4 in the Supplementary Material present all these associations.

3.5. Higher rates of synaesthesia among Czechs are due to a negative association between early multilingualism and synaesthesia

The higher rates of synaesthesia among native Czech/Slovak than native English speakers ran counter to our initial hypothesis, and so we explored the data further to try and determine the cause of this difference. This exploratory analysis found that the between-sample difference could be explained almost entirely as stemming from a relationship between multilingualism and synaesthesia, in which *native multilinguals* (those who report speaking two languages prior to age two) are much less likely to develop synaesthesia than *non-native multilinguals* (those who report learning a second language later in life).

As noted in 3.2, the SFU sample contains a large number of these native multilinguals, while the CU sample does not (1230 and 76, respectively). Both groups, however, contain many non-native multilinguals. Fig. 3 presents rates of synaesthesia (any type) among all 11,404 participants at both CU and SFU, split according to their reported age of second language acquisition. It shows that the rate among non-native multilinguals (3.5%) is more than twice that among native multilinguals (1.4%), and the 95% confidence intervals of these estimates do not overlap.

Results are qualitatively the same when looking at the SFU sample alone. The CU sample has only 76 native multilinguals and 7 individuals who reported never having learned a second language, so rates of synaesthesia for these individuals (6.6% and 0%, respectively) cannot be usefully compared to the SFU rates.

When non-native multilinguals whose first language is Czech or English are compared between the CU and SFU samples, estimated prevalences are very similar, with confidence intervals that overlap each other's means (4.4% vs 3.9%). Thus the large number of native multilinguals in the SFU sample appears to be the main reason why overall rates of synaesthesia are lower among native English speakers than among native Czech/Slovak speakers. This is an important and unexpected finding, but even after controlling for second-language acquisition in this way, we do not find support for the hypothesis that CU would have a lower prevalence of synaesthesia. However the role of orthographic transparency can be further examined in our data, which we do in the following section.

3.6. Orthographic transparency is negatively associated with synaesthesia in the SFU sample

There were many languages other than Czech/Slovak and English reported in the survey, particularly in the SFU sample (3.2). Rates of synaesthesia among native speakers of these languages support the hypothesis that in general, transparent languages have lower rates of synaesthesia than opaque ones.

There were 152 distinct languages reported in our survey, the large majority of which have never been formally classified in terms of their orthographic transparency. Thus in order to test the effect of transparency among the large number of native languages in our sample, new classifications had to be generated. All languages reported in the survey were independently classified according to their orthographic transparency by two authors (MRW and KAA). This was accomplished by consulting standardized tables of grapheme and phoneme correspondences (found primarily at www.omniglot.com and www.wikipedia.org) and counting the total number of forward and backward exceptions (number of phonemes greater than 1

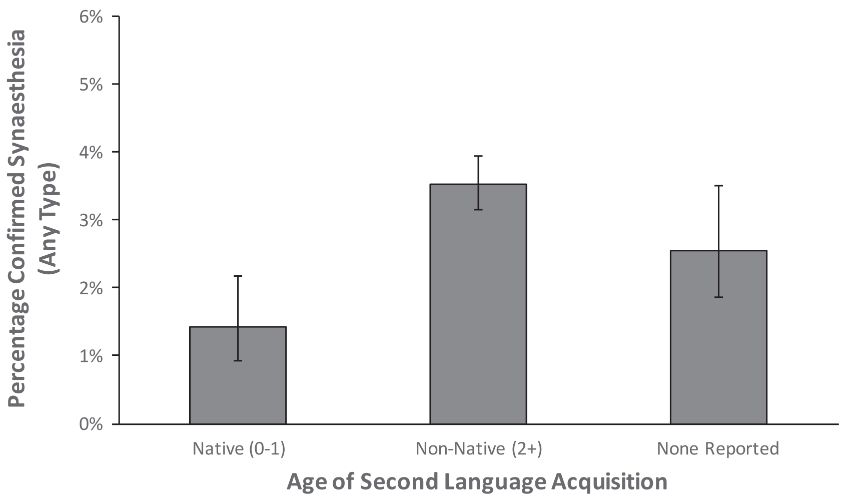

Fig. 3. Rates of synaesthesia (any type) according to age of second language acquisition among all participants in both samples, regardless of native language. Note that most of those who did not report a second language will actually have had some second-language training in elementary school. Error bars indicate 95% Wilson C.I.s.

associated with each grapheme, and number of graphemes greater than 1 associated with each phoneme, respectively). When information was available about rule-governed exceptions, such as one grapheme producing voiced and unvoiced variants of a consonant, or long and short versions of a vowel, or a bigraph being treated as a single letter (such as CH in Czech), these were not counted. Languages were classified as opaque (more than 5 forward and 5 backward exceptions), fully transparent (5 or fewer forward and 5 or fewer backward exceptions), forward/backward transparent only (5 or fewer forward exceptions but more than 5 backward exceptions, or *vice versa*), or unclassifiable (33 languages in all, generally due to not having a standard script). Classifications made by each author were compared and any discrepancies were resolved.

Our transparency classifications were broadly consistent with previous rankings of multiple languages according to their transparency (e.g. Borgwaldt, Hellwig, & Groot, 2005; Duncan et al., 2013; Seymour, Aro, & Erskine, 2003; Ziegler et al., 2010). The most complex and precise method of performing these rankings uses a measure of "entropy" that weights each exception by its prevalence in a corpus (Borgwaldt et al., 2005; Ziegler et al., 2010). This was simply not feasible in this case, but our classification of the languages used in these studies was consistent with their relative entropy scores. That is, all languages we classified as opaque had higher entropy scores than languages we classified as forward, backward, or fully transparent, and all languages we classified as forward or backward transparent had higher entropy scores than languages we classified as fully transparent. There were some discrepancies between our classifications and those of studies that used more *adhoc* methods of classification, for example we classified Dutch as transparent and German as opaque, but one study reported Dutch as more opaque than German (Seymour et al., 2003). Gratifyingly, however, in those cases where the same languages were classified by the more precise entropy measure, the entropy scores lined up with our classification (e.g. Dutch has an entropy approximately half that of German, meaning Dutch is more transparent, in Borgwaldt et al., 2005). Thus our determinations of orthographic transparency are consistent with the best data, when these data are available.

We also classified each language as alphabetic, abugida (where each character represents a consonant/vowel combination), abjad (where each character represents a consonant, and vowels are generally left unmarked), or morphographic (in this sample, only speakers of Chinese languages and Japanese). Languages were divided according to their scripts because there are differences in the ways these scripts are learned and processed (e.g. Koyama, Hansen, & Stein, 2008), which we reasoned might be important for synaesthetic development if, as we hypothesize, this is partly driven by learning to read and write. However we did not have a specific hypothesis here, and so our examination of script-specific effects on synaesthetic prevalence was purely exploratory.

In order to examine the orthographic transparency hypothesis separately from the effects of multilingualism, we focused solely on native monolingual speakers of each language. After grouping all native monolingual speakers with the same transparency or script type, any group with less than 100 members was eliminated from further analyses. Unfortunately this meant that the role of transparency and script could not be examined within the CU sample, which contained only 12 native monolingual speakers of a language that was not fully transparent, and only 1 native monolingual speaker of a language with a non-alphabetic script. In the SFU sample, only 63 native monolinguals spoke a language that was forward transparent only, only 9 spoke a language that was backwards transparent only, and only 39 spoke a language with an abjad script, so these were not analyzed further. (There were no confirmed synaesthetes found among any of the excluded groups.)

Chinese languages presented a particular challenge for this analysis. While many survey participants reported speaking "Cantonese", "Mandarin", or other specific Chinese languages and dialects (in total, 14 different terms were used), many others simply reported "Chinese". Without more information on their background, it is unfortunately impossible to determine which Chinese language, and how many of these languages, they were referring to. All university students who grew up in China would speak Mandarin, however for many of them it would not have been their native language. Furthermore, SFU has many Canadian-born students of Chinese ancestry, and a large number of these would not speak Mandarin at all, but Cantonese or possibly yet another language. Thus we simply grouped all reported speakers of any Chinese language or dialect under a single "Chinese" label, recognizing that there are substantial differences between some of these languages, and also that an unknown number of these individuals are actually speakers of more than one language.

As shown in Fig. 4, there was a higher rate of synaesthesia (any type) among native monolingual speakers of opaque languages than of transparent languages (2.8% vs 1.2%, Fisher's Exact $p = 0.018$), and a higher rate among speakers of alphabetic than abugida languages (3.0% vs 0.8%, $p = 0.045$), within the SFU sample. The rate among speakers of alphabetic languages was also higher than among morphographic languages, but this did not pass a 0.05 threshold (3.0% vs 2.0%, $p = 0.057$). No other comparisons approached significance (all ps > 0.250).

Fig. 5 shows the proportions of confirmed synaesthetes (any type) among native monolingual speakers of all languages with more than 20 native monolingual speakers in either the SFU or CU samples. Rates of synaesthesia vary quite widely with native language, ranging from 0% to 6.5%. Statistical comparisons are of limited use here, given that there are well under 100 native monolingual speakers of most of these languages, but certain patterns are worth highlighting. First, of the 15 languages with more than 20 native monolingual speakers in the SFU sample, 7 had no synaesthetes at all, and only one of these is an opaque language (Urdu), while of the 8 languages with at least one synaesthete, only three are transparent (Russian, Korean, and Punjabi), and two of these have less than 1.1% synaesthetes (Korean and Punjabi). Furthermore, languages typically spoken in areas with close cultural and genetic ties can have very different rates of synaesthesia that line up with their transparency: we find 3.6% French (opaque) but no Spanish (transparent) synaesthetes, and 6.5% Japanese (opaque) but only 1.0% Korean (transparent) and no Vietnamese (transparent) synaesthetes.

The only transparent languages with more than 1.1% synaesthetes are Czech/Slovak and Russian, and the fact that Russian has a relatively high rate of synaesthesia in both the CU and SFU samples (3.5% and 5.9%, respectively) suggests that this is

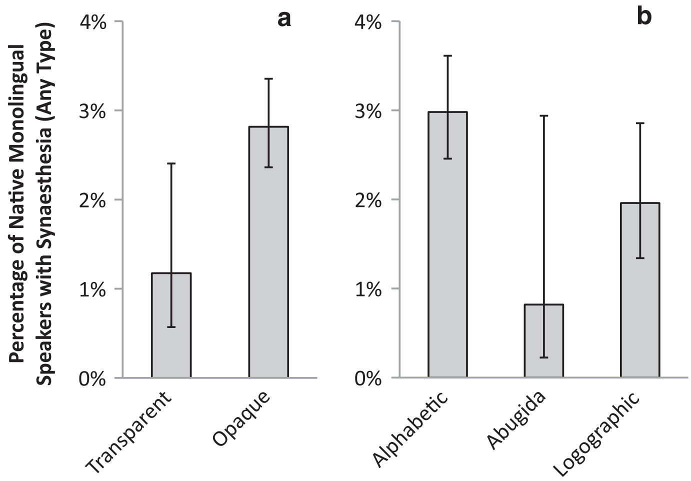

Fig. 4. Rates of synaesthesia (any type) in the SFU sample only, among native monolingual speakers of languages varying in (a) orthographic transparency and (b) script type. Error bars indicate 95% Wilson's C.I.s.

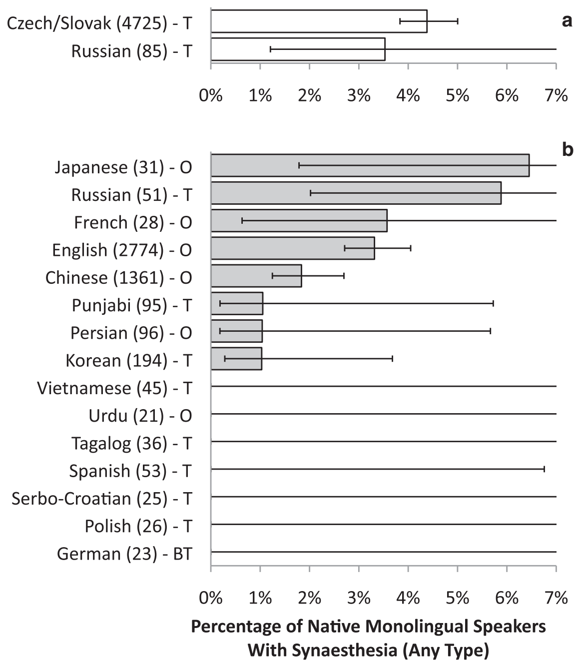

Fig. 5. Rates of synaesthesia (any type) among native monolingual speakers of any language with more than 20 native monolingual speakers in the CU sample (a) and the SFU sample (b). Numbers in parentheses indicate the number of native monolingual speakers in each sample, and the letter code indicates the orthographic transparency of the language – O: opaque, T: Transparent, BT: Backward Transparent. Error bars indicate 95% Wilson's C.I.s.

not simply due to chance. As both are Slavic languages, rates of synaesthesia were compared among all native monolingual speakers of Slavic and non-Slavic languages in the SFU sample, but there was no apparent difference between these groups (2.6% vs 2.4%, respectively).

3.7. A female bias for synaesthesia due to differences in compliance

As in previous studies (Barnett et al., 2008; Baron-Cohen et al., 1993, 1996), we found a female bias for synaesthesia: women were somewhat more likely to report having synaesthesia than men, and substantially more likely to be confirmed as having it. However the design of our study allowed us to directly examine whether this female bias was due to a genuine difference in the population, or to sex-linked differences in compliance with the experimental protocol, consistent with the explanation given by Simner and colleagues (Simner & Carmichael, 2015; Simner et al., 2006; Ward & Simner, 2005). We found strong evidence for this compliance hypothesis.

We quantified sex biases for each type of synaesthesia in our study using its female:male *relative rate* (RR – the rate among women dived by the rate among men). An RR of 1 indicates no sex bias, numbers above 1 indicate a female bias, and numbers below 1 indicate a male bias. Of the 8 types of synaesthesia on the paper survey, all were reported by more women than men (female:male RRs ranging from 1.04 to 1.50) except for the "Other" category in both samples (CU: 0.86, SFU: 0.95) and letter-colour synaesthesia in the SFU sample only (0.99). This female bias increased for confirmed rates: there were more women than men confirmed as synaesthetes for all 7 types of synaesthesia we tested on the Battery (RRs ranging from 1.06 to 4.51), except for chord-colour (0, as the 2 confirmed chord-colour synaesthetes were both male) and scale-colour synaesthesia (0.91) in the SFU sample. All these rates are given in Table S5 in the Supplementary Material available online. The female:male RR for reporting of any type of synaesthesia was 1.18 among native Czech/Slovak speakers in the CU sample, and 1.10 among native English speakers in the SFU sample, while for confirmed synaesthesia the RRs were 2.72 and 1.77, respectively.

In order to be confirmed as a synaesthete, subjects had to (1) report synaesthesia on the paper survey, (2) register for the Battery in response to our emailed invitation, (3) actually complete the Battery, and finally (4) achieve a score below the consistency threshold. To investigate sex differences in compliance, we calculated female:male RRs at each of these 4 stages. Specifically, we calculated female:male RRs (1) of reporting synaesthesia among those who were given the survey, (2) of registering for the Battery among those who reported synaesthesia on the survey, (3) of completing the Battery among those who registered for it, and finally (4) of scoring below the threshold among those who completed the Battery. Since the female bias was present in both samples and across virtually all forms of synaesthesia, all 11,664 participants were used in order to maximize power (results were qualitatively similar when run with either sample alone), and a single measure was used to indicate if a participant reported or was confirmed as having any one of the seven varieties of synaesthesia tested on the Battery.

Fig. 6 shows the results. There was a small female:male bias (RR = 1.23) for reporting synaesthesia, then a larger bias (RR = 1.76) for registering for the Battery after reporting synaesthesia. No significant bias existed for either completing a Battery test after registering for the Battery (RR = 1.11) or being confirmed as synaesthetic after completing a test (RR = 1.05). This means that the observed female:male bias in confirmed synaesthesia was almost entirely due to compliance: men were simply less likely to comply with our request to register for the Battery.

As noted in 3.1, the proportion of women was higher in the CU (68.9%) than the SFU sample (61.0%). This, combined with the female bias for confirmed synaesthesia, accounts for some of the higher prevalence of synaesthesia in the CU sample, but not very much. We estimate, using the data reported here, that if the CU sample had the same proportions of women and men as the SFU sample, but maintained the same proportions of female and male synaesthetes, then the overall rate of confirmed synaesthesia would be 4.2%, down only 0.2% from the actual CU rate of 4.4%. This is still well above the SFU rate of 2.7% ($\chi^2 = 15.15$, Fisher's exact $p < 0.001$).

3.8. Other survey questions

Responses to the other survey questions were not associated with any form of confirmed synaesthesia, but there were positive (but weak) associations between several forms of reported synaesthesia and reports of reading difficulties and having imagined narratives in childhood in which letters or numbers were characters. Further Analyses in the Supplementary Material explores these results in depth.

4. Discussion

4.1. Summary of results

Rates of synaesthesia vary widely with early language experience. This survey found that synaesthesia is almost twice as common among native Czech/Slovak speakers as English-speakers (3.3, 3.4). However this was due to the large number of native multilinguals in the SFU sample, who have lower rates of synaesthesia than non-native multilinguals. When only non-native multilinguals are compared, there is no difference in prevalence between the two groups (3.5). After controlling for second language by looking only at non-native multilinguals, we do find the expected effect of transparency: the incidence of synaesthesia is higher among native monolingual speakers of opaque languages than native monolingual speakers of transparent languages, with the notable exception of Czech/Slovak (3.6). We also show that the apparent female prevalence bias in this and other studies is almost entirely attributable to gender differences in compliance (3.7).

4.2. The prevalence of synaesthesia varies widely between groups

Among the most important implications of these results is that it is unhelpful to speak of an overall incidence of synaesthesia. Rather, incidences vary between groups according to linguistic and cultural factors. Orthographic transparency and age of second-language learning appear to be two such factors, but there are likely others.

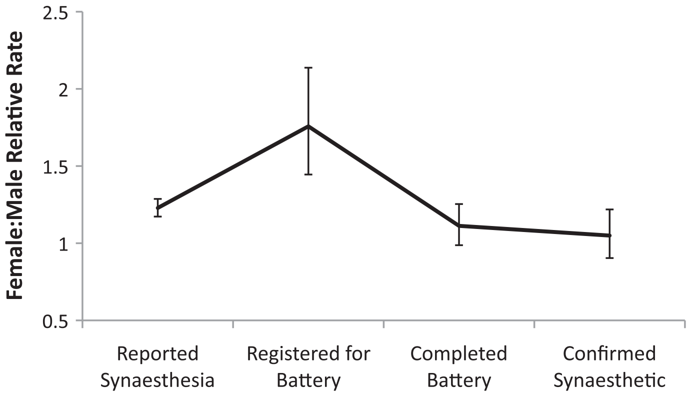

Fig. 6. Relative rates of female to male participants (averaged across all participants from both the CU and SFU samples) at various stages of the study: those who initially reported synaesthesia, those who registered for the Synesthesia Battery after reporting synaesthesia, those who completed at least one Battery test after registering, and those who were confirmed synaesthetic after completing a test. 1 indicates an equal proportion of females and males. Error bars indicate 95% C.I.'s.

For example, early interest or training in the fine arts might affect the prevalence of synaesthesia, consistent with the findings that synaesthesia is more common among fine arts students (Rothen & Meier, 2010b), and that synaesthetes have a higher rate of participation and interest in the arts than the rest of the population (Rich et al., 2005; Ward, Thompson-Lake, Ely, & Kaminski, 2008). Of course it is also possible that the causal connection to the arts runs in the opposite direction (people become interested in art because they have synaesthesia), or that a third factor promotes the development of both synaesthesia and artistic tendencies. Such a third factor might be a personality profile that encourages the development of both artistic inclinations and the use of synaesthesia for childhood learning challenges. Consistent with this, adult synaesthetes tend to have greater degrees of openness to experience and tendency to fantasize (Banissy et al., 2013; Rouw & Scholte, 2016), enhanced visual imagery (Barnett & Newell, 2008; Meier & Rothen, 2013; Price, 2009; Rizza & Price, 2012; Spiller & Jansari, 2008; Spiller, Jonas, Simner, & Jansari, 2015; but cf. Simner, 2012), stronger creativity (at least some types of creativity, cf. Ward et al., 2008), better memory for certain stimuli (Rothen & Meier, 2010a; Simner, Mayo, & Spiller, 2009; Ward, Hovard, Jones, & Rothen, 2013; Yaro & Ward, 2007), and a higher overall intelligence (Rouw & Scholte, 2016), all of which might influence the likelihood of being interested in art, and of using unusual perceptual/cognitive strategies to solve learning challenges. However once again we have a chicken-and-egg problem over the direction of causal influence. Synaesthesia or artistic training could influence the development of this personality profile (as argued by Banissy et al., 2013), and there could also be multi-directional influences. This illustrates how difficult it will be to determine a full range of influences on the development of synaesthesia, particularly if, as we suggest, some of these influences are only found in mid-to-late childhood.

4.3. Late second-language-learning as a trigger for synaesthesia?

We have argued that higher rates of synaesthesia among speakers of opaque languages (with the notable exceptions of Czech/Slovak and Russian) are due to the increased difficulty of becoming literate in these languages. A similar explanation may account for the finding of higher rates of synaesthesia among non-native multilinguals. Learning to read and write in two (or more) languages is easier for those who are native speakers of both languages, and so they might have less need for unusual learning strategies than those who learn a second language later in life.

Consider a monolingual child learning to speak, read, and write their second language, and a native multilingual learning to read and write a language they grew up speaking. From birth, the native speaker has honed a set of *metalinguistic* skills that are strong predictors of success in learning to read: *phoneme awareness*, the ability to individuate phonemes in spoken words (cf. Bar-Kochva & Breznitz, 2014; Caravolas et al., 2012; Ghanem & Kearns, 2014); *morphological awareness*, the ability to recognize and use parts of words (cf. Bar-Kochva & Breznitz, 2014; Ghanem & Kearns, 2014; Wei et al., 2014); and *prosodic sensitivity*, the ability to recognize and use pitch, duration, and intensity in spoken language (cf. Calet, Gutiérrez-Palma, Simpson, González-Trujillo, & Defior, 2014; Whalley & Hansen, 2006). Thus the monolingual second-language learner is at a clear disadvantage compared to the native speaker.

What is the result if both the bilingual and monolingual are learning a novel language which neither can yet speak? There is substantial evidence that monolinguals learning their second language are often at a disadvantage compared to native bilinguals learning their third (or trilinguals learning their fourth, etc) (reviewed in Cenoz, 2013). One reason for this may be that native multilinguals have better metalinguistic capabilities than monolinguals, even for languages that they do not yet speak (Jessner, 2014; Rauch, Naumann, & Jude, 2012; Thomas, 1988). Another is that native multilinguals have a variety of general cognitive advantages that are not specifically linguistic in nature, but that give obvious benefits in learning to speak, read, and write new languages. These advantages have mainly been found in executive function (particularly in cases involving resolving conflicts between different rules or sources of information), selective attention, and working memory (for recent reviews, see Adesope, Lavin, Thompson, & Ungerleider, 2010; Bialystok, 2015; Kovács, 2015). They are consistent across cultures and languages (Bialystok & Viswanathan, 2009; Yang, Yang, & Lust, 2011), and are found throughout the lifespan, originating in infancy (cf. Brito, Sebastián-Gallés, & Barr, 2015; Estes & Hay, 2015; Gervain & Werker, 2013), and persisting into old age (Bialystok, Craik, & Luk, 2012; Gold, 2015).

Thus in general, non-native second-language learners face learning challenges that native multilinguals do not face, or should find substantially easier. The learning challenge hypothesis would therefore predict lower rates of synaesthesia among native than among non-native multilinguals, exactly as we observe.

4.4. Native bilingualism as a guard against synaesthesia?

An alternative possibility is that native bilingualism discourages the development of synaesthesia because the native bilingual is faced with conflicting correspondences between phonemes and graphemes from a very early age, and this may make it difficult to "pin down" a referent for synaesthetic colours. Against this possibility, we note that a speaker of a language as opaque as English has arguably as great a conflict between phonemes and graphemes as any bilingual, since every letter is associated with multiple English phonemes. Japanese provides a particularly interesting example here, since every literate Japanese person maps the same set of sounds onto 4 different scripts (Hiragana, Katakana, Kanji, and Romaji), which is essentially the inverse of the problem faced by many native bilinguals, who map multiple sounds onto one script. As Fig. 5 shows, the estimated prevalence of synaesthesia among native Japanese speakers is actually the highest in our entire study, suggesting that phoneme-grapheme conflicts do not provide any impediment to the development of synaesthesia, indeed quite the reverse, consistent with our learning hypothesis.

4.5. Confirmed rates as lower bounds on synaesthesia's true prevalence

4.4% (CU) and 2.6% (SFU) of our survey respondents passed the Synesthesia Battery's confirmation threshold for at least one form of synaesthesia (3.4), but these rates are certainly underestimates. First, most of those who reported synaesthesia on the paper survey chose not to comply with our request to take the Synesthesia Battery (3.1). Our strong female bias for confirmed synaesthetes is particularly notable in this regard, both since we find disproportionate male attrition (3.7) and since the recent consensus is that no notable sex bias actually exists (Rouw & Scholte, 2016; Simner & Carmichael, 2015; Simner et al., 2006). Even if we assume that there is no sex bias, and raise the number of confirmed male synaesthetes accordingly, these sex-adjusted rates would probably still be too low. It seems very likely that some women (and somewhat more men) who would have passed the Battery chose not to register for it, despite the motivation provided by the lottery.

Participants who initially reported synaesthesia on the paper survey but did not complete the Battery were emailed and asked for clarification. Many of those who responded reported that they had misunderstood the survey questions or simply stated that they were not actually synaesthetes (Simner, 2012 reports a similar result). However most of these individuals did not respond, and it seems reasonable to assume that there were some genuine synaesthetes in this group who were simply not interested in our research.

Furthermore, a surprisingly large number of participants were confirmed as having a type of synaesthesia that they denied having in the paper survey (3.4, cf. also Simner et al., 2006). These participants were invited to take the Battery only because they had responded positively to a survey question about at least one form of synaesthesia, yet after registering they decided to take a test for another form, and were found to have consistent associations. This raises the possibility that there are "unconscious synaesthetes" who are unaware of having any forms of synaesthesia at all, and would even deny this when asked, but who would nevertheless pass one or more of the Battery tests. Any of these individuals would not have been discovered using our methods. (We discuss these individuals further in 4.6.)

Finally, as noted in 1.6, a strict test of genuineness such as the Synesthesia Battery likely rules out many genuine synaesthetes who have less consistent inducer-concurrent associations (Rouw & Scholte, 2016; Simner, 2012). Unfortunately, it does not seem possible to identify these individuals on the basis of their responses to the paper survey since, as we have just noted, our survey questions produced many false positives, consistent with previous results (Simner, 2012). Perhaps with a more detailed and stringent set of survey questions, including a requirement to give examples, we might have been better able to rule these out (Rouw & Scholte, 2016), but this would likely have conflicted with the need for a very rapid survey to avoid excessive class interruptions.

For all these reasons, the rates of reported and confirmed synaesthesia given in Figs. 1 and 2, respectively, should be interpreted as upper and lower bounds on the true rates.

4.6. The puzzle of "unconscious synaesthesia"

The two most common operational criteria for synaesthesia are self-report and the consistency of reported concurrents, which our data (and that of Simner et al., 2006) demonstrate to be dissociable. Furthermore, neither criterion may be necessary at all. Several researchers argue that consistency is not a defining feature of synaesthesia (Niccolai, 2012; Rouw & Scholte, 2016; Simner, 2012), and if "unconscious synaesthetes" exist, then self-report is not one either.

Synaesthetes who report no conscious synaesthetic experiences, but have consistent inducer-concurrent experiences (and possibly other markers of synaesthesia) are hard to understand, but even the more limited ones we find in our study are quite puzzling. However the large number of these individuals, and their distribution across all types of synaesthesia, suggests that this is a robust finding, and one in need of explanation.

It seems that many individuals *fail to notice that the majority of the letters in the alphabet induce highly specific and consistent colours* (and so on for the other types of synaesthesia we test). This is especially puzzling given that the survey began with a description of synaesthesia, each question was illustrated with concrete examples, and each of these individuals responded positively to another question about synaesthesia. How could someone miss this? Perhaps they have a latent tendency to associate particular shapes with colours (cf. Albertazzi et al., 2013) that allows them to reliably choose similar colours for particular letter shapes. Alternatively, perhaps they have specific letter-colour associations, but these play little or no role in their day-to-day phenomenology, and so it is only after several rounds of questions that they become fully aware of them. Whatever the explanation, the evidence suggests that synaesthesia phenomenology can be extremely subtle.

4.7. Explaining rates of synaesthesia among native Czech/Slovak and Chinese speakers

It is difficult to pinpoint the precise effects of orthographic transparency given the current data. If the present data set only contained native Czech/Slovak and English speakers, it would be very strong evidence *against* our hypothesis that transparent languages have lower rates of synaesthesia (3.3 and 3.4). Alternatively, if the present data set came entirely from the SFU sample, it would be very strong evidence *for* this hypothesis (3.6). It is clear that orthographic transparency by itself cannot explain both (1) higher rates of synaesthesia among native Czech/Slovak speakers (or equal rates if we control for age of second-language learning), and (2) lower rates of synaesthesia among native speakers of other transparent languages (with the exception of Russian). As noted in 3.7, there was a higher proportion of women in the CU sample, but this explains virtually none of the between-sample difference. Some factor in addition to gender, orthographic transparency and multilingualism must be at play here.

We consider two possible causes of higher rates among Czech, Slovaks, and Russians, that are not meant to be mutually exclusive. First, there might be a simple cultural difference in reporting biases, such that Czechs, Slovaks and Russians of both sexes may be particularly willing to report unusual experiences such as synaesthesia, a possibility consistent with the anecdotal experiences of a number of this paper's authors. Second, and more controversially, former Soviet cultures have had a long-standing explicit reverence of synaesthesia as a creative force in music and art, popularized by such luminaries as Kandinsky, Rimsky-Korsakov, and Scriabin (cf. Galeyev & Vanechkina, 2001; Ione & Tyler, 2003; Peacock, 1985), and it is possible that this encourages the development or preservation of synaesthesia. This reverence could presumably only influence those with extensive enough artistic training to have encountered it, and we have no information about the artistic backgrounds of our participants, but it may nevertheless account for some of the differences we observe. It has been denied that this "artistic" synaesthesia is the same as the perceptual variety studied in this article (Galeyev, 2001, 2005a, 2005b; Galeyev & Vanechkina, 2001), but this is largely on the ground that it is "associative" or "mental", and as we have argued extensively in the Introduction, this is no longer a tenable position.

Similarly, the somewhat lower rate of confirmed synaesthesia among the primarily Chinese speakers of morphographic languages (Fig. 4) may stem from a cultural reluctance to report unusual experiences. This lower rate might also result from an unidentified number of Chinese native multilinguals who grew up speaking multiple Chinese languages or dialects, but only reported speaking "Chinese" (3.6). Thus rates among true Chinese native monolinguals might be somewhat higher than we report, but our data cannot confirm this. It might also be that there is some other factor related to the processing of morphographic scripts that explains this result.

4.8. Comparisons with three previous prevalence estimates

At present, the most widely-accepted estimates of synaesthetic prevalence are those of Simner and colleagues, taken primarily from undergraduates at Edinburgh University (Carmichael et al., 2015; Simner & Carmichael, 2015; Simner et al., 2006). These studies did not report the linguistic backgrounds of their participants, but we can infer that they had a substantially higher rate of native English speakers than our SFU sample, and far fewer native multilinguals. In 2011, 68.4% of residents of Vancouver, the main city close to SFU, spoke primarily English at home (Statistics Canada, 2012), in comparison to 93% of Scottish residents (National Records of Scotland, 2013). Edinburgh University presumably has a higher proportion of non-English speakers than the rest of Edinburgh, and Edinburgh will have a higher proportion than the rest of Scotland, but it nevertheless seems very unlikely that participants in these studies would have anything close to as low a rate of native English speakers as SFU (62% in our study, 3.2). These studies used both self-report and consistency tests to confirm synaesthesia, and so we should expect their results to be reasonably close to our confirmed results for native English speakers (Fig. 2). This is exactly what we find: e.g. 1.0% grapheme-colour synaesthetes in the present study, in comparison to 1.0–1.4% in these studies (Carmichael et al., 2015; Simner & Carmichael, 2015; Simner et al., 2006).

Most recently, Rouw and Scholte (2016) found higher rates of synaesthesia in the Dutch population. Differences in methodology and analysis are important to note here. First, they grouped synaesthesia into the five broad clusters suggested by Novich, Cheng, and Eagleman (2011). Of these, only three are relevant to the present study. The vast majority of our confirmed synaesthetes (Fig. 2) belong in the *coloured sequence* cluster (which would include all of our letter-, number-, weekday-, and month-colour synaesthetes). The remainder of our confirmed synaesthetes all belong in the *coloured music* cluster (chord-, instrument-, and scale-colour synaesthetes)), while those who report number forms (Fig. 1) belong in the *spatial sequences* cluster. Second, as previously discussed (1.6), they used a novel method of confirming synaesthesia, which both allows synaesthetes with somewhat inconsistent inducer-concurrent associations to be confirmed as synaesthetic, and rules out many of those who falsely report synaesthesia. If we had used the same technique, we would expect to find rates that were higher than our confirmed rates (because more inconsistent synaesthetes would be accepted) but lower than our reported rates (because more false reports would be rejected). This is what we find for each of the three clusters we can compare, whether looking at rates from the CU or SFU samples.

Rouw and Scholte (2016) did not report the linguistic backgrounds of their participants, but their participants were selected to be representative of the Dutch population, which is largely composed of ethnically Dutch individuals who speak Dutch natively (Central Intelligence Agency, 2016) and at least two other languages (European Union, 2012). Dutch is a fairly transparent language (3.6), and while we do not have precise data on the age of second language learning in this sample, it seems most probable that it has relatively few native multilinguals, making the linguistic profile much closer to our CU than our SFU sample. With our results in mind, then, the transparency of Dutch should be a negative influence on the prevalence of synaesthesia, while non-native second language learning should be a positive one. Unfortunately the differences in methodology and analysis just discussed make it difficult, if not impossible, to meaningfully compare rates between our studies, beyond what we have already done above.

Finally, we turn to one of the first large-scale studies of synaesthesia (Calkins, 1893, 1895), which is thought of as having an overly liberal bias because its confirmation of synaesthesia was solely on the basis of self-report, and its prevalence estimates were far higher than most modern studies (Simner & Carmichael, 2015; Simner et al., 2006). We would argue, however, that the results merit closer study. Participants were given a lengthy and highly detailed questionnaire (far more detailed than any modern one we have seen), and required to give specific examples of any synaesthetic experiences. Formal consistency tests were not used, and in most cases reports were taken at face value, but almost 200 participants were asked to provide a second set of responses to the questionnaire several months to a year after their first responses. All but one of these participants gave very similar responses to both questionnaires, and thus further verification of responses was deemed unnecessary, except in the case of grapheme-colour synaesthesia. While there is no formal quantification of the degree of similarity between each set of responses, this methodology is far more rigorous than it might appear, and is really closer in spirit to that of Rouw and Scholte (2016) with the addition of an informal consistency test administered to some participants.

Calkins (1895) suggests that the rates of synaesthesia she reports are underestimates, as they are calculated based on the total number of surveys given out, but she managed to greatly increase her response rate during the three years of the study, and the overall prevalence of synaesthesia in each of these three years increased accordingly (approximately quadrupling from 1893 to 1895). Unfortunately, she does not provide a detailed breakdown of the prevalence of each form of synaesthesia in each year, so we can only use her estimates from the data combined across all three years, accepting that these numbers should be substantially higher. She reports 4.5% letter-colour, 1.4% number-colour, and 6.4% music-colour, all of which are higher than the confirmed prevalences among native English speakers in our study (Fig. 2), but lower than reported prevalences (Fig. 1), just as with Rouw and Scholte's (2016) results. Her prevalence of number forms (24.1%) is far higher than that reported among native English speakers in our study, but similar to that reported by Czechs (Fig. 1), and also to other modern studies of number forms (Mann, Korzenko, Carriere, & Dixon, 2009; Sagiv et al., 2006).

There are a number of reasons that might explain why Calkins' (1895) prevalences are higher that our confirmed rates. Like Rouw and Scholte (2016), her methodology is probably substantially more tolerant of inconsistent synaesthetic associations. All participants were female, so any sex bias would be working to increase her rates. Virtually all these participants would have been native English speakers, but all would have had extensive training in other languages, as every candidate for entry into Wellesley College (where the survey was conducted) had to have mastered Latin, as well as some combination of Greek, French, or German (depending on the specific program applied to, cf. Wellesley College, 1892). According to the learning hypothesis, then, we would expect higher rates of synaesthesia, because of their opaque first language, their monolingual early upbringing, and their extensive non-native language acquisition. Furthermore, many would have been accomplished artists and musicians, both because this was a standard expectation of young middle- or upper-class women at the time, and because the college had Schools of Art and Music that allowed combined degrees with other subjects (Wellesley College, 1892). Given the stringent entrance requirements for Wellesley (Wellesley College, 1892), they would also have been unusually intelligent. Thus they matched the "synaesthetic personality profile" discussed in 4.2 (Banissy et al., 2013; Rich et al., 2005; Rothen & Meier, 2010b; Rouw & Scholte, 2016). Overall, it seems that there was something of a "perfect storm" of synaesthesia-promoting tendencies in this study.

We cannot provide a comprehensive review of all previous estimates of synaesthesia's prevalence, but we hope that the discussion of these three studies shows how estimates are likely to vary both due to the nature of the sampled group, and to the methodological and analytical choices made.

4.9. Some suggestions for future studies

A future study might probe for the presence of synaesthesia in multiple ways, by requiring *all* participants to answer simple questions about synaesthetic experiences (as in our initial survey, and Carmichael et al., 2015; Simner & Carmichael, 2015; Simner et al., 2006), to provide detailed examples of any reported synaesthesia (as in Calkins, 1895; Rouw & Scholte, 2016), and to participate in a rigorous consistency test for multiple varieties of synaesthesia (as in our study, and Simner et al., 2006), even if their initial responses indicated that they did *not* have any conscious synaesthetic experiences (unlike our study, and Simner et al., 2006). Ideally extensive demographic information on all participants would also be collected, including linguistic history. And while we are making such inconsiderate demands on future researchers' time and resources, we suggest that this should be a large-scale study that uses representative samples of multiple different linguistic groups, preferably with more similar cultures than Canada and the Czech Republic (based on Fig. 5, Spain and France, or Korea, Japan and China might provide appropriate comparisons).

Comparing the three prevalences obtained in such a study would quantify the differences in estimated prevalence that stem from methodological differences. It would also determine the ratio of "inconsistent" and "unconscious" synaesthetes to the more standardly-studied synaesthetes who both report synaesthesia and pass consistency tests. And it should provide stronger evidence for or against the learning hypothesis, further data on the effects of second-language learning, and if the populations were chosen carefully enough, one might even be able to find differential effects of second-language learning on rates of synaesthesia depending on the orthography of one's native and second languages (cf. Bialystock & McBride-Chang, 2005; Cenoz & Gorter, 2011).

Of course this would be an extraordinarily difficult, time-consuming, and complex study, or more likely series of studies, though an international team of researchers might be able to lower the investigative load with coordinated effort. Despite its difficulty, such a study would resolve many of the issues we have raised in this Discussion.

4.10. Why are effects found for all synaesthesia types?

The focus of our study was on the prevalence of grapheme-colour synaesthesia in particular, but we find differences between linguistic groups in rates of colour synaesthesia *of any type*, not simply grapheme-colour. Indeed, the co-occurence of all types of synaesthesia in our samples is so strong that it would be impossible to isolate differential effects without a vastly larger sample. There are at least two ways of accounting for the non-specificity of rate differences that are consistent with the learning hypothesis. First, it may be that once children have success at solving a particular learning challenge with an unusual method, they are more likely to use similar strategies for future challenges. So a child who learns to write an opaque orthography (or the orthography of a non-native second language) using colour associations might then use colours to overcome the challenge of understanding and using time units such as days or months. Another possibility is that other factors (possibly neurophysiological and/or genetic ones) influence the initial likelihood of developing colour associations at all, and hence of being able to make the associations with learned category structures that constitute the vast majority of adult synaesthesias. Of these associations, perhaps only the most useful or pleasant are systematized and maintained into adulthood (Calkins, 1893). This might be compatible with the "neonatal synaesthesia" hypothesis, according to which we are all born synaesthetic, but neural pruning removes the connections that underlie synaesthesia in most of us (Maurer, 1997; Maurer & Mondloch, 2006).

4.11. Learning challenges as one of many influences on synaesthetic development

The transparency and multilingualism effects we have reported are consistent with the hypothesis that synaesthesia is more likely to develop, or to be preserved, in children faced with particularly difficult learning challenges (Watson et al., 2014), although the surprisingly high prevalence of synaesthesia in Czech/Slovak provides an important challenge to this theory. This is not to deny that other factors play a critical role in the development of synaesthesia, including but not limited to one's genetic inheritance (Asher et al., 2009; Tomson et al., 2011) and neurophysiological profile (Maurer, 1997; Maurer & Mondloch, 2006; Ramachandran & Hubbard, 2001a). Rather, genetics, neurophysiology, language, and presumably other developmental factors all contribute to the phenomenology of adult synaesthesia that most researchers have studied to date. Determining how these factors interact over the course of childhood to produce these unusual conscious experiences is one of the primary challenges facing the field at this time.

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Appendix A. Supplementary material

Supplementary data associated with this article can be found, in the online version, at http://dx.doi.org/10.1016/j.concog.2016.12.004.

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