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Synesthesia in a congenitally blind individual

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ABSTRACT

Synesthesia represents an atypical merging of percepts, in which a given sensory experience (e.g., words, letters, music) triggers sensations in a different perceptual domain (e.g., color). According to recent estimates, the vast majority of the reported cases of synesthesia involve a visual experience. Purely non-visual synesthesia is extremely rare and to date there is no reported case of a congenitally blind synesthete. Moreover, it has been suggested that congenital blindness impairs the emergence of synesthesia-related phenomena such as multisensory integration and cross-modal correspondences between non-visual senses (e.g., sound-touch). Is visual experience necessary to develop synesthesia? Here we describe the case of a congenital blind man (CB) reporting a complex synesthetic experience, involving numbers, letters, months and days of the week. Each item is associated with a precise position in mental space and with a precise tactile texture. In one experiment we empirically verified the presence of number-texture and letter-texture synesthesia in CB, compared to non-synesthete controls, probing the consistency of item-texture associations across time and demonstrating that synesthesia can develop without vision. Our data fill an important void in the current knowledge on synesthesia and shed light on the mechanisms behind sensory crosstalk in the human mind.

1. Introduction

In synesthesia, a particular sensory experience is induced by an object, person or event without a real stimulation in that sensory domain (Ward, 2013). For example, synesthetes may see fluctuating shapes while listening to the sound of a saxophone, experience the letter “A” as written in bright red and perceive the smell of almonds as “pale blue” (Day, 2005). In some cases, synesthesia is acquired after brain damage (Ro et al., 2007), drug usage (Luke and Terhune, 2013) or sensory loss (Afra et al., 2009; Armel and Ramachandran, 1999). However, many synesthetes report having this kind of experiences since they can remember (Sinke et al., 2012), qualifying as cases of ‘developmental synesthesia’ (Ward, 2013).

Epidemiological data indicates that, among the five senses, vision seems to play a central role in synesthesia. According to the most recent estimates (Day, 2005; Niccolai et al., 2012a; Simner et al., 2006), between 80% and 97% of synesthetes report color-related synesthesia (e.g., graphene-color, music-color). On the contrary, synesthetic phenomena between non-visual senses (e.g., touch-smell, audition-touch) are reported by a small proportion of the interviewed synesthetes (ranging between 8% and 15%; see Day, 2005; Niccolai et al., 2012a) and, in the majority of the cases, these individuals also report synesthetic experiences that exhibit a visual component (either as inducer or concurrent, see Niccolai et al., 2012a; and the literature review reported in the supplementary materials).

The existent data therefore seems to suggest a privileged role of vision in the synesthetic phenotype (i.e., the modalities in which synesthesia manifest itself). However, it remains unclear what the role of vision might be in the development of synesthesia (i.e., the mechanisms by which synesthesia emerges). One possibility is that, although most of the synesthetic phenomena are visual, vision is not necessary for the development of synesthesia. For instance, the lack of functional vision from birth could have obvious consequences on the content of synesthetic associations (e.g., a congenitally blind person would never perceive as ‘pale blue’ the smell of almonds) but leave unaffected the possibility to develop synesthesia between the spared senses (e.g.,

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almonds may smell like the opening movement of Beethoven's 5th symphony).

A single case of a congenitally blind person with developmental synesthesia would support this hypothesis. Congenital blindness is a rare disease which affects about 0.03% of the population (Silverstein et al., 2013). Considering that the estimated incidence rate of synesthesia is around 3% (Brang and Ramachandran, 2011; Simner et al., 2006), the joint probability of a person having both conditions, if the two are independent, would be 0.0009%. Although this is a low incidence, it is not undetectable (Silverstein et al., 2013). Considering the current population of the United States (328.2 million), we should expect about 3000 cases of Americans congenitally blind synesthetes (See Silverstein et al., 2013, for a similar argument applied to the joint probability of blindness and schizophrenia). Moreover, if we consider also the cases of people who became blind within the 3rd year of age (i.e., early blind) the incidence would increase significantly, approximating the base-rate of disorder such as schizophrenia (0.07% (Silverstein et al., 2013);) and Asperger Syndrome (0.06% (Taub, 2013);) for which cases of synesthesia have been reported (Jakhar and Mehta, 2017; Neufeld et al., 2013). Under these circumstances, it is at least suspicious that we were not able to find in the literature a single case of a congenitally or early blind person with developmental synesthesia, despite the fact that both blindness and synesthesia have been the subject of several scientific papers. In the literature we could however find cases of synesthetes that became blind late in life and maintained their visual synesthesia or developed a new one after sensory loss (Niccolai et al., 2012b; Steven and Blakemore, 2004), and the interesting case of BP, a congenitally blind musician that reported multisensory hallucinations after LSD consumption (Dell'Erba et al., 2018).

The virtual absence of developmental synesthesia among congenitally blind people opens to a second possibility, namely that vision is necessary to develop synesthesia. Indeed, evidence supporting this hypothesis can be drawn from recent studies suggesting that early blindness impairs the development of cross-modal correspondences (CMC), a phenomena based onto the cross-talk between different sensory systems and often related to synesthesia (Bankieris and Simner, 2015; Lacey et al., 2016; Martino and Marks, 2001; Ramachandran and Hubbard, 2001; Ward et al., 2006). CMCs are non-arbitrary associations between seemingly unrelated sensory features. Two examples are the sound-shape association (e.g., the sounds/buba/and/kiki/are associated with round and spiky shapes, respectively; Kohler, 1947) and the pitch-size correspondence (high and low pitch is associated with small and big size, respectively; Spence, 2011). Some studies have shown highly-reduced or absent CMCs in congenitally blind people (Deroy et al., 2016; Fryer et al., 2014; Hamilton-Fletcher et al., 2018; Sourav et al., 2019), despite the fact that these CMCs involved experiential domains that could be directly experienced haptically or auditorily (e.g., shape-sound; pitch-height) and were robustly observed in blindfolded sighted control groups. One possible explanation of these results is that vision plays a pivotal role in setting up multisensory functions during ontogeny, acting as a sort of coordinator across the different senses (Hötting and Röder, 2009; King, 2009), a theoretical framework proposed to explain also some instances of reduced multisensory integration in congenitally blind people (Champoux et al., 2011; Hötting and Röder, 2009; Occelli et al., 2012). The hypothesized detrimental role of congenital lack of vision in establishing different forms of sensory cross-talk (multisensory integration, crossmodal correspondences) that share neural mechanisms with synesthesia (Ward et al., 2006), together with the absence in the scientific records of a congenitally blind synesthete, suggests that early blindness may impair the development of synesthesia and maybe prevent it *tout-court*.

Here, we provide evidence that, despite the aforementioned results and hypotheses, synesthesia can develop in the complete absence of vision by reporting, for the first time to our knowledge, the case of a congenitally blind person (CB) with graphene–texture, lexeme–texture and spatial sequence synesthesia. Our report of this case will be divided in two parts. First, we will describe in detail the synesthetic experience of CB, highlighting the structural similarities with previously reported cases of visual synesthesia (e.g., graphene-color, sequence-space synesthesia); Second, in order to empirically verify the reality of his synesthesia, we tested the consistency of CB graphene-texture and lexeme-texture associations, across two experimental sessions, adapting to the haptic domain an established objective procedure to test visual synesthesia (Eagleman et al., 2007).

2. Methods

2.1. Case description

CB is a 40-year-old male, born totally blind by sighted parents due to congenital retinopathy caused by maternal rubella during pregnancy. He is right-handed, has a PhD in Computer Science, is fluent in Braille reading and has no history of neurological or psychiatric disorders. CB is an Italian native speaker and he is also fluent in English. The interview and the experiment described in this paper were conducted in Italian and were approved by the local ethical committee at the university of Trento (protocol 2014–007).

CB reports a series of synesthetic experiences, involving numbers, letters, days, months, texture and space, that he has been experiencing since he can remember. CB's parents, as well as his twin sister, also born congenitally blind, have never experienced any form of synesthesia.

We will describe his synesthetic associations separately for clarity. Number-related synesthesia is overall the strongest one, however, spatial and haptic sensations related to letters, days and months are also quite consistent and immediate in CB's experience. It should be noted that it is always the semantic representation (i.e., numbers, letters) that induces tactile and proprioceptive (spatial) sensations in CB, never vice versa. Furthermore, CB reports to feel the tactile synesthetic sensation of the different textures mostly on his index fingers.

The following information were collected during an interview constructed on the basis of preliminary data provided by the participant in a first informal meeting, as well as on the relevant literature on synesthesia (Day, 2005; Ward, 2013).

2.1.1. Number-texture synesthesia

Whenever CB hears, writes, reads or thinks about a whole number (i.e., 1, 2, 3, etc.) he experiences it as having a shape (i.e., a small cube of different sizes depending on the number) and very specific texture properties (See Table 1). For example, number 1 and 2 are experienced as being as scratchy as cardboard, while number 3 is as soft as velvet. Number 4 and 5 are as smooth as plastic; so do numbers 6 and 7, but the type of plastic he perceives is different from number 4 and 5. Numbers

Table 1
A list of CB's number-texture associations.

Inducer	Synesthetic experience
1 and 2	Scratchy as cardboard
3	Velvet
4 and 5	Smooth like plastic
6 and 7	Smooth like plastic, but less than 4 and 5
8	Plastic but less smooth than number 5, but smoother than 6 and 7
9	Similar to 3 but less velvet like
10, 11,12	Metallic
13	Similar to 9 but less velvet like
14, 15	Similar to 4 and 5 but smoother
16, 17	Similar to 6 and 7
18	Similar to 8 but scratchier
19	Similar to 9
20	Similar to 2 but smoother
21–29	Similar to numbers ranging between 1 and 9
30	Similar to 3
31–39	Similar to numbers ranging between 1 and 9
Hundreds	Similar to numbers ranging between 1 and 9
1000	Scratchy as cardboard

10, 11 and 12 are experienced as being smooth and cold like metal. Numbers containing the same digit (e.g., 17 and 27) are experienced as being similar in texture. For instance, numbers 9 and 19 are both soft as velvet but differ in the quality of this softness, with 9 being softer than 19. This also translates into a sort of regularity among numbers, by which, for example, numbers between 21 and 29, or 31 and 39, present similar characteristics to numbers 1 and 9. This sort of regularity is typically encountered in other cases of visual synesthesia, such as color-number synesthesia (Ward, 2013).

A similar pattern occurs for hundreds too: number 100 and 200 are similar to number 1 and 2, and the numbers between 101 e 199 are similar to 1 and 99. This pattern repeats for the other hundreds (i.e., 200, 300, 400, etc) but also for the thousands (i.e., 1000, 2000, etc). Finally, negative numbers do not differ in texture from positive numbers, so that, for example, −1 is experienced as being scratchy as number 1.

2.1.2. Number-space synesthesia

CB describes the numbers (cubes) to be aligned in a 3D mental space, which is activated each time CB thinks, writes or hears a number. Numbers are not simply aligned along a single line but proceed along broken lines with a direction change every 10 numbers/cubes. The overall impression is that of a broken line that proceed in a sort of zig-zag path with oblique segments and abrupt directional changes. Negative numbers appear specular with respect to positive numbers.

This internal mental space comprises an entire scenery composed of mountains on the background and a sort of vacuum on the upper and lower spatial borders (See Fig. 1 for an attempt to represent, visually, CB's number space). When CB retrieves numerical knowledge, he finds himself to be immediately located in this mental space where he moves through the cubes/numbers navigating safely, he says, as he would in his own home, since the number space it is a very familiar environment. The modality of entrance in the space is simultaneous to the number presentation, CB finds himself directly focused on the number, but because of the velocity of this mechanism he reports that he could be unaware of his body displacement process. The numerical map is anchored allocentrically, meaning that numbers/cubes do not move as a consequence of CB movements. Thanks to its world-centered and three-dimensional nature, CB can explore the cubes from different perspectives (e.g., from one particular side, from above or, less often, from below). The use of an allocentric reference frame and the possibility to explore the synesthetic spatial environment from multiple perspective has been previously reported in visual sequence-space synesthesia (Ward, 2013).

All the cubes are equally spaced between each other and have the same dimension, except from the hundreds (100, 200, 300, etc.) and the number 1000, which are bigger than the other numbers. Cubes' dimensions stop changing after the number 1000. It is important to note that the texture, shape, size and spatial location of numbers are not dissociable for CB, in the sense that hearing or reading a number would

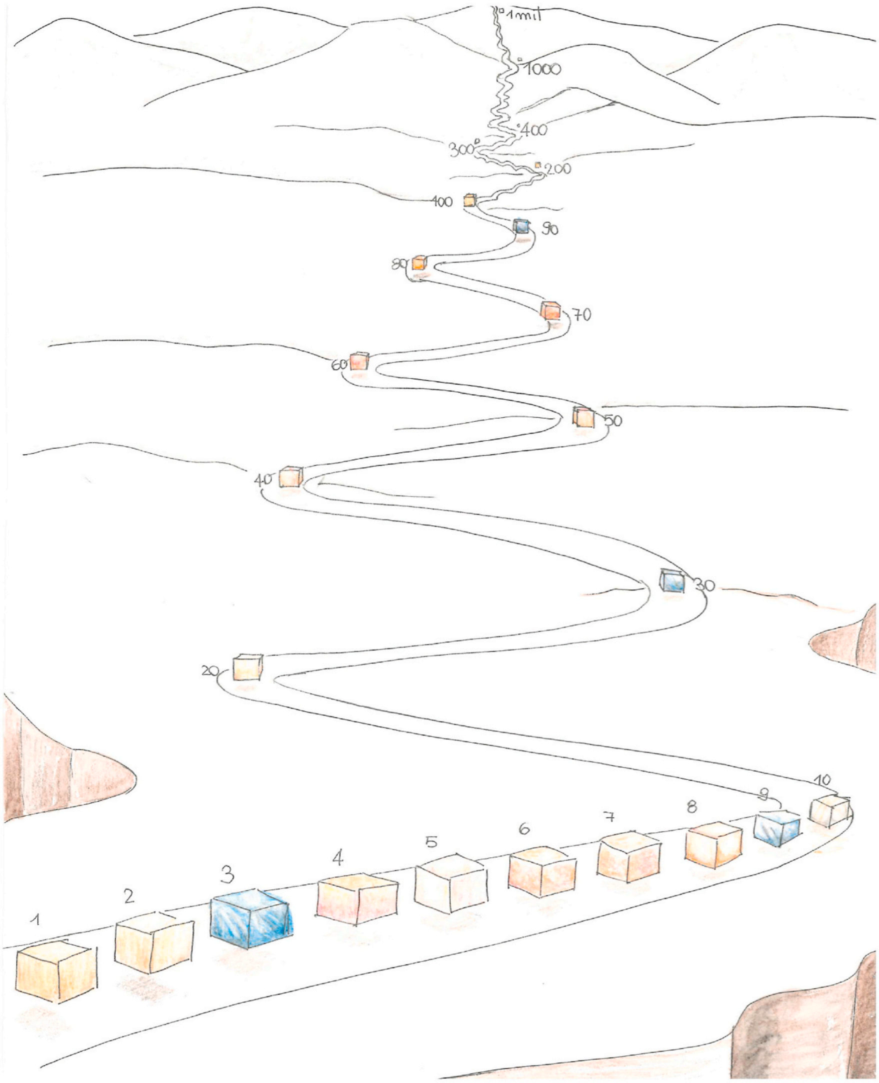


Fig. 1. Tentative rendering of the number-space described by CB's as it could appear in vision. Cube-textures are here indexed with different colors. In CB's number-space each single number is associated with one single cube along the path, and cubes are equally spaced. For graphical reasons here we are showing each single cube only for the first tent (1–10). The intent of this rendering is to show the overall structure of the mental space more than provide a detailed representation. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

never elicit in CB the tactile sensation only, without its spatial location, or its shape but not its size, etc. However, we shall remark that shape associations may not be considered synesthetic (contrary to texture and space associations) because they lack inter-item arbitrariness (i.e., all the numbers have the same shape), which is a common characteristic of synesthesia. This is the case also for the shape of letters, months and days of the week.

2.1.3. *Letter-texture synesthesia*

CB perceives letters as two-dimensional squares with a specific texture. For example, A is as soft as snow, B is similar to plexiglass and E is icy. K is metallic while L, M and N are all plastic-like but very different from the type of plastic he experiences when thinking about the letter R. T is as smooth as velvet but less smooth than Y (Table 2 reports all letters, each associated with a texture).

2.1.3.1. Letter-space synesthesia. All the squares have the same size and are spatially organized from A to Z onto a single diagonal left-to-right line that CB experiences in front of him.

2.1.4. *Days/months-texture synesthesia*

The seven days of the week are represented as gates all having same size, while the twelve months of the year are represented as cubes/boxes of very similar size. As for numbers and letters, days and months also possess a very specific texture: Monday is wooden-like and smooth, Tuesday is like rough plastic, Wednesday and Thursday are like smooth plastic, Friday is similar to Wednesday but smoother, Saturday is metallic and Sunday is velvet.

January and February are as scratchy as cardboard, March is metallic and April is plastic. September feels like a mop-like material, and November is rough like raw-wool (see Table 3 for a list of days/months-texture associations). Months do not have the same texture association as the numbers from 1 to 12, indeed, month and number are represented in two distinct spatial maps.

2.1.5. *Days/months-space synesthesia*

Days and months are displayed in a mental space that is separated from the one in which numbers are represented. Months are organized in a circle and floating in the vacuum. However, this vacuum is different from the one surroundings the number map and it has a different density. Within each month/box there is a standard numerical map that goes from 1 to 31 (or 30, 28), where each number/cube stands for a day of the month. These number/cubes within the month boxes have the same spatial and haptic characteristics of the numbers in the original number space. In fact, these number/days are never associated with a particular day of the week (e.g., Monday or Thursday).

Instead, hearing, reading or thinking about specific days brings CB to another space, in which the 7 days of the week are represented as gates along a straight road and have their own haptic properties (See Table 3).

Table 2
A list of CB's lexeme-texture associations.

Inducer	Synesthetic experience
A	Snow
B	Plastic like plexiglass
C	Plastic, similar to B but less smooth
D	Plastic, similar to B
E	Icy
F	Smooth plastic
G	Plastic, similar to C
H	Rubbery material
1	Plastic, similar to B and D
J	Similar to H
K	Metallic
L	Plastic but less smooth than C and F
M	Plastic but less smooth than C and F
N	Plastic but less smooth than C and F
O	Similar to E but scratchier
P	Similar to F
Q	Cardboard
R	Plastic but less smooth than M and N
S	Paper
T	Velvet
U	Metallic
V	Plastic
W	Plastic
X	Velvety paper
Y	Smooth velvet
Z	Plastic, similar to L, M and N

Table 3
A list of CB's month-texture associations.

Inducer	Synesthetic experience
Monday	Wooden-like and smooth like being freshly painted
Tuesday	Rough plastic
Wednesday	Smooth plastic
Thursday	Smooth plastic
Friday	Similar to Wednesday but smoother
Saturday	Metallic
Sunday	Velvet
January	Cardboard
February	Cardboard
March	Metallic
April	Plastic
May	Metallic, similar to March but smoother
June	Similar to April
July	Plastic similar to Thursday
August	Cardboard
September	Mop-like material
October	Smooth plastic
November	Raw-wool
December	Similar to November

3. Experiment

3.1. *Participants*

In this experiment we compared the performance of CB with that of 8 age- and education-matched sighted controls that never experience synesthesia (Mean age: 38.6 (5.14) 6 men and 2 women) and two non-synesthete congenitally blind volunteers (both females of 26 and 38 y. o.). All participants were Italian native-speakers and one of them was left-handed. Participants gave informed consent for the study and the experiment was ethically approved by the local ethical committee (protocol 2014–007). The size of the control group was based on previous control samples for single cases of synesthesia, as well as on the usually large effect size obtained when comparing true synesthetes and non-synesthetes in consistency tests (Ward, 2013).

3.2. *Behavioral testing*

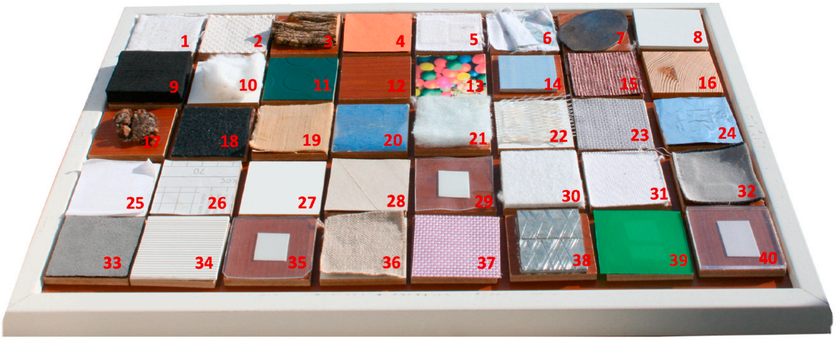
3.2.1. *Tactile board*

We designed a “tactile board” (45 × 40 cm) containing 40 rectangular wooden cards (8 × 5 cm). Each card was covered by a particular material with a precise texture (see Fig. 2). We collected different sample textures among material types mentioned by CB during the interview (e.g., fabric, plastic, metal). A list of the textures is reproduced in Fig. 2.

The different textures belong to one of 5 material categories: Paper, wood, metal, plastic or fabric. There was more than one texture for each material, with different characteristics (e.g., satin, tablecloth, velvet), in order to recreate the complexity and the perceptual fine-grain differentiation that characterizes true synesthetic experience (Ward, 2013).

3.2.2. *Experimental procedure*

The experiment aimed at measuring the participants' consistency in retrieving, over time, the matching between concepts and textures.



1. Cotton I (F)
2. Cotton II (F)
3. Wood I (W)
4. Cardboard 1 (Pa)
5. Cotton III (F)
6. Tablecloth (F)
7. Metal I (M)
8. Plastic I (P)
9. Rubber (P)
10. Cotton III (F)
11. Plastic II (P)
12. Laminated Wood (P)
13. Laminated Cardboard (P)
14. Metal II (M)
15. Velvet I (F)
16. Wood II (W)
17. Wood III (W)
18. Felt I (F)
19. Velvet II (F)
20. Wrapping paper (Pa)
21. Felt II (F)
22. Net (P)
23. Tablecloth (P)
24. Tinfoil (M)
25. Paper I (Pa)
26. Paper II (Pa)
27. Plastic III (P)
28. Cardboard III (Pa)
29. Plastic IV (P)
30. Felt III (F)
31. Cotton IV (F)
32. Satin (F)
33. Leather (F)
34. Plastic V (P)
35. Plastic VI (P)
36. Bag (F)
37. Plastic VII (P)
38. Metal III (M)
39. Plastic VIII (P)
40. Plastic IX (P)

Fig. 2. Tactile board. The letters in parenthesis indicate the material category to which each texture was assigned (F= Fabric; W= Wood; Pa= Paper; P=Plastic; M = Metal). Numbers are assigned arbitrarily.

Testing the consistency and precision of synesthetic associations over time is an established method to prove the perceptual nature of synesthetic experiences (Eagleman et al., 2007), and has been successfully used across different types of synesthesia such as graphene-color (D Brang et al., 2010) or lexeme-taste (Ward and Simner, 2003). If CB's synesthesia evokes true perceptual experience, we expected him to display highly-precise and reliable item-texture associations, such that he would be able to discriminate the precise tactile sensation elicited by a given concept (e.g., a number or a letter) across a variety of similar tactile sensations and doing it consistently across different experimental sessions.

Participants engaged in a concept-texture association task comprising of two identical sessions that were conducted 30 days apart (+-5) from each other. The position of the cards was randomly shuffled before each session. Participants were warned that they will undergo the same test after one month, and that their test-retest consistency will be measured. We predicted that CB would present a higher test-retest consistency compared to non-synesthetic controls.

Before each experimental session, participants were blindfolded and conducted into a quiet room. Instructions were then provided orally by the experimenter. At the beginning of the session participants had the possibility to explore the board with both hands, touching all the cards, for 3 min. We selected 69 items (24 numbers, the 12 months of the year, the 7 days of the week, the 26 letters from the alphabet) to be associated with a given texture during the experiment. These items were presented in a randomized order to the participants.

Each trial started with the experimenter uttering the item word (e.g., "April"); then the participant waited at least 5 s and thought about the tactile sensation elicited by that word, without touching the board. After this temporal window, the experimenter said "go", indicating that the participant could start exploring the board in order to find the texture that was closer to the tactile sensation elicited by the item word. Participants could take all the time they needed to find the right texture. Once the participant had found the texture, s/he was asked to indicate the chosen card to the experimenter, who could then record the response. Immediately after, the participant rated how much the texture chosen was close to the 1 s/he had in mind, from one to five (1 = Not similar at all; 5 = very similar). Each experimental session lasted between 60 and 80 min, and participants took 3 short breaks during each session.

3.2.2.1. Similarity ratings. In a final session of the experiment, performed on-line through the platform Zoom©, we collected from CB similarity ratings concerning the different tactile experiences elicited by different numbers and letters. In two separate sessions we collected pairwise similarity ratings related to numbers (digits from 1 to 10; 45 pairs) and letters (325 pairs). Each pair (e.g., letters "A" and "R") was uttered by the experimenter, and CB needed to evaluate the similarity of the tactile sensations elicited by each item. Similarity was rated from 1 to 7 (1 = very dissimilar, 7 = very similar). The aim of this experimental session is to compare the similarity of the synesthetic experience elicited by different numbers or letters, with the orthographic similarity between the same numbers and letters, in Braille. Do letters (or numbers) that elicit similar synesthetic textures also have similar Braille patterns? In order to compare different Braille patterns with each other we relied on Jaccard similarity, which is the ratio of the Intersection over the Union of two finite sets:

$$J(A,B) = \frac{|A \cap B|}{|A \cup B|}$$

In the case of two Braille letters, it will be the ratio of the number of dots presents in both letters over the total number of dots obtained superimposing the two letters.

4. Results

CB test-retest consistency in the number domain was of 75%. This means that CB associated the same item (e.g., 'one', 'seven', 'twelve') with the same exact texture across the two sessions 75% of the time (See Table 4). The same measure revealed an accuracy of 42% in the domain of Letters, 42% in Days of the week and 8% in Months. On the other hand, the control group showed the following test-retest consistency results: Numbers = 7%; Letters = 8%; Days of the week = 9%; Months = 18%.

In the domains of Numbers, Letters and Days of the week, none of the control subjects performed better than CB (see Table 4 and Fig. 3). A Fisher Exact test confirmed that the performance of CB was significantly higher than the performance of control subjects in the domain of

Table 4
Test-retest consistency across experimental sessions for CB and controls.

	Num	Let	Days	Months
CB	.75	.42	.43	.08
SC1	.12	.12	.14	.42
SC2	.12	.04	.00	.42
SC3	.17	.23	.14	.08
SC4	.04	.04	.14	.00
SC5	.00	.04	.00	.17
SC6	.00	.08	.00	.25
SC7	.12	.15	.29	.17
SC8	.12	.08	.14	.08
BC1	.00	.04	.00	.25
BC2	.04	.04	.00	.00
All Con	.07	.08	.09	.18

SC= Sighted Control; BC= Blind control.

Numbers (Odds-ratio = 35.83, $p < 0.001$) and Letters (Odds-ratio = 7.83, $p < 0.001$). The Fisher-exact test was significant also in the domain of Days of the week (Odds-ratio = 7.83, $p = 0.03$), although the p-value would not survive a Bonferroni correction for multiple comparisons (corrected-alpha = 0.0125). The same comparison, instead, did not reach significance in the domain of Months (Odds-ratio = 0.40, $p = 0.69$).

As a further step, we correlated the accuracy of CB and controls with the matching ratings provided by each participant for each item-texture association in the first session. Since it is possible that some items elicited in CB a particular texture sensation that was not adequately represented in our tactile-board, we predicted that CB accuracy would have been higher for the items that were associated, in session 1, with textures that CB considered highly-similar to the one elicited in his synesthetic experience. Matching ratings varied from 1 to 5, and we excluded from this analysis rating values that, for each participant, were used less than 5 times, since a low number of observations may lead to inflated (or deflated) accuracy (For instance, CB used the matching rating "1" only 1 time, leading to either 100% or 0% accuracy for that rating bin). The distribution of matching ratings for CB and for the control participants is shown in Fig. 4a and b. As expected, CB provided overall higher matching ratings than controls. Most importantly, as shown in Fig. 4c, CB's accuracy was greater for items that were rated as a good match (higher matching rating), whereas this trend was not present in the controls.

Finally, we conducted analysis on the similarity ratings between tactile sensations elicited by different numbers or letters. Since CB experiences these synesthetic textures on the index fingers, which are also used to read Braille letters and numbers, we investigated whether the similarity of Braille codes correlates with the similarity of synesthetic textures. Indeed, in the literature we can find some cases of letter-color synesthetes in which letters with a similar shape were associated with similar colors (Brang et al., 2011).

We constructed two similarity matrices (one for numbers and one for letters) based on the judgments provided by CB, and two similarity matrices based on the Jaccard similarity of Braille patterns. We correlated the matrices and tested the significance of the correlation using a permutation test with 10,000 permutations and shuffling at each iteration the labels of the two matrices. There was no significant correlation with Jaccard similarity in the number domain (Spearman's Rho = 0.23, $p = 0.12$; Fig. 5a), but a significant positive correlation emerged in the domain of letters (Spearman's Rho = 0.22, $p = 0.004$; Fig. 5b), showing that the more two Braille letters were orthographically like each other, the more the synesthetic texture elicited by those letters tend to be similar to each other. Importantly, the correlation between orthographical similarity remained significant even when controlling (through partial correlation) for the vicinity of letters in the alphabet sequence (Spearman's Rho = 0.15, $p = 0.04$; Fig. 5d), suggesting that letters with similar orthography trigger similar synesthetic experiences independently (at least in part) from their relative position in the

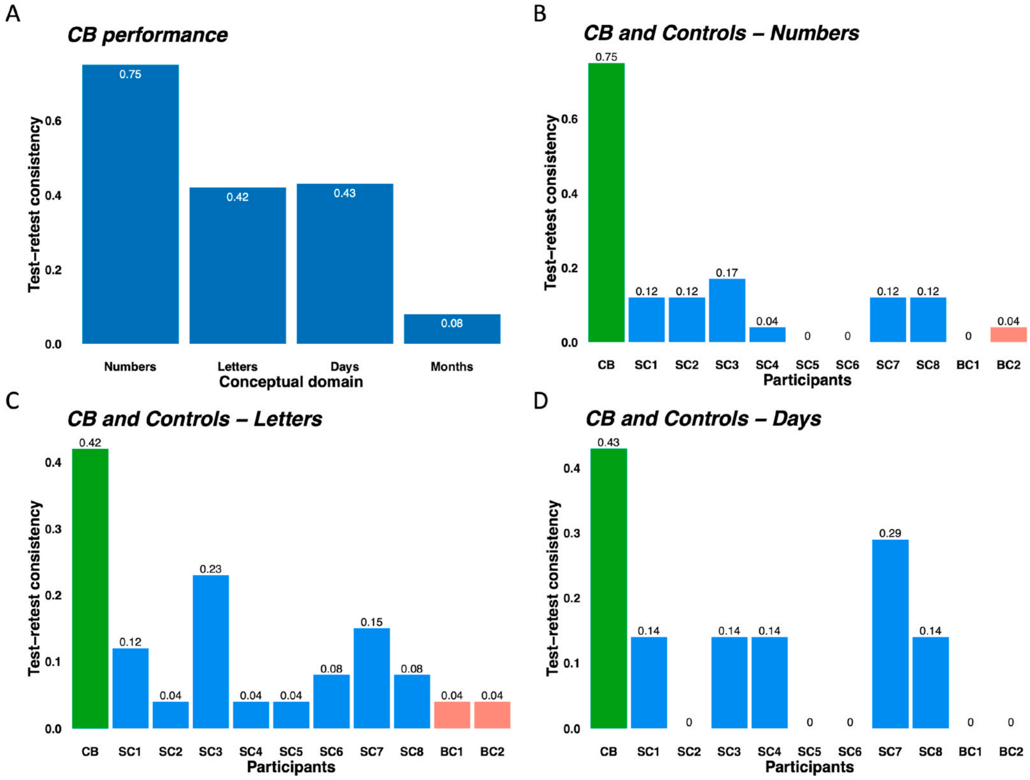


Fig. 3. CB's performance in terms of test-retest consistency within each conceptual domain (A). Comparison between CB's performance and those of sighted controls (SC1-8) and congenitally blind controls (BC1–2) in the domains of Numbers (B), Letters (C) and Days of the week (D).

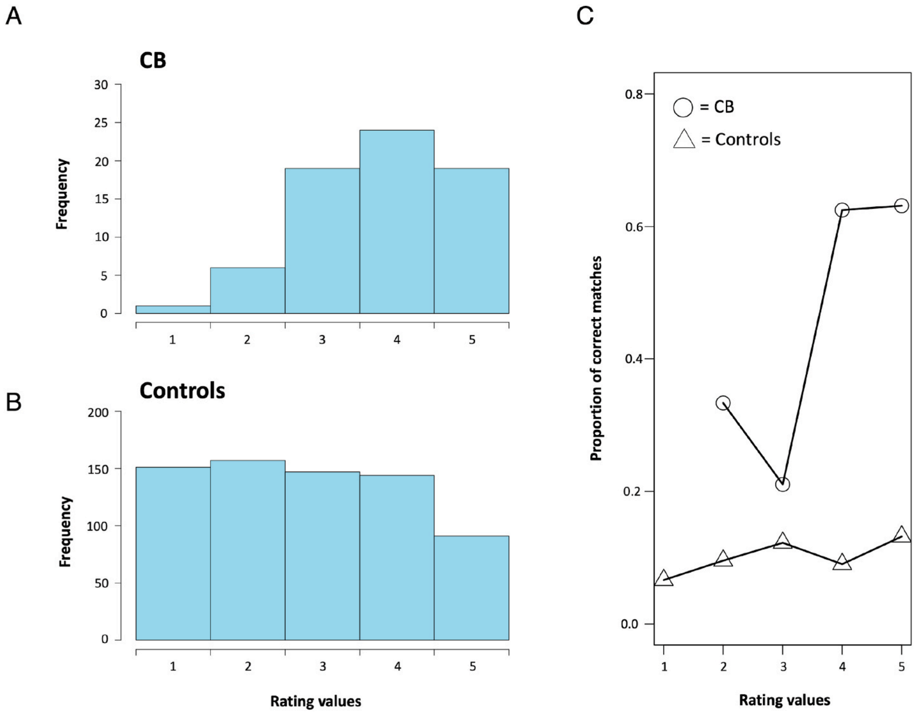


Fig. 4. CB (A) provided overall higher matching ratings compared to control participants (B). Importantly, the test-retest consistency (accuracy) was higher, in CB's data, for items that was rated as a good match (C). This trend was less pronounced in controls.

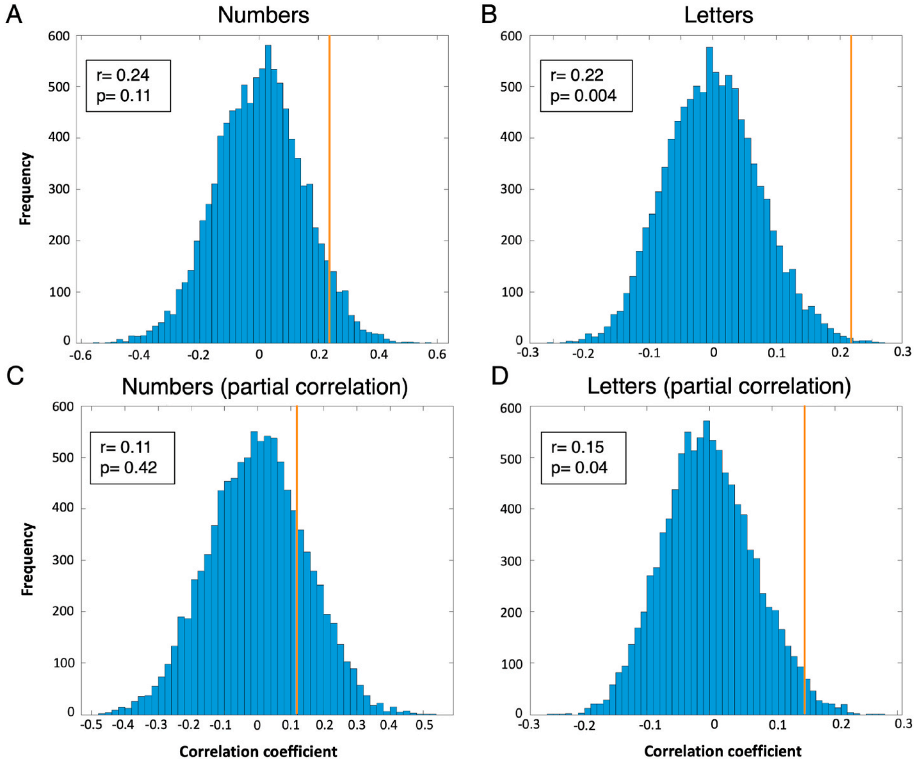


Fig. 5. Correlation (A, B) and partial correlation (C, D) between the orthographic similarity and the texture similarity of numbers (A, C) and letters (B, D).

alphabet. The same partial correlation analysis in the number domain gave a non-significant result (Spearman's Rho = 0.11, p = 0.42; Fig. 5c).

5. Discussion

We documented, for the first time to our knowledge, a case of developmental synesthesia in a congenitally blind person. CB reports a complex synesthetic phenotype that can be triggered by different types of inducers: numbers, letters, days and months. Each of these items is associated with a particular position in space and with a particular texture. CB synesthetic experience, although completely non-visual, carries similar characteristics with previously documented visual synesthesia. For instance, number-texture associations presents mathematical regularities (e.g., the texture of 3 is similar to the one of 13, 23 and 333), which are typical of number-color synesthesia (Ward, 2013), and CB's spatial map of numbers presents structural regularities (nested configurations), spatial anchoring (allocentric) and modalities of exploration (from different viewpoints) that are typical of visual forms of sequence-space synesthesia (Ward, 2013). Although we did not directly test CB's spatial synesthesia, it is interesting to notice that CB's case can pull apart the spatial and the visual component of sequence space synesthesia (which are usually confounded in sighted synesthetes; Gould et al., 2014) showing that, at least in some cases, spatial synesthesia can be non-visual.

To empirically assess the genuineness of CB' synesthesia we tested the stability of item-texture associations over time. Compared to non-synesthetic controls, CB showed a significantly higher test-retest consistency (over a period of one month) for numbers and letters. On the other hand, his performance in the domain days of the weak was not statistically different from the one of controls once correcting for multiple comparisons. Finally, CB performance in the domain of months was somewhat lower compared to the other conceptual domains and not significantly different from the one of control subjects.

Thus, based on our test, we could not firmly assess the reliability of CB's months-texture and days-texture synesthesia. However, the null result in these two conceptual domains should not be necessarily interpreted as evidence of absence of synesthesia. First, at least for days of the week, the low number of datapoints available (only seven) could have prevented us to statistically detect a real difference, even if none of the control subjects had a better performance than CB in this domain (see Fig. 3). Second, Lacey and colleagues recently showed that strength and consistency of synesthetic experiences are not necessarily correlated (Lacey et al., 2021), and that strong but inconsistent synesthesia may not be detected by tests that focus on consistency alone. Further experimentation is needed to assess the genuineness of CB's month-texture and day-texture synesthesia. On the contrary, the reliability of CB's item-texture synesthesia emerged strongly in the domains of numbers and letters. In the case of number-texture associations (CB's stronger type of synesthesia), CB associated each number with the same exact texture 75% of the time. A striking performance, if we consider that the available textures were sometimes extremely similar to each other (e.g., different types of plastic, wood or metal; See Fig. 2) and that the best of the control subjects scored a mere 17%.

CB's case fills the void left by the absence of synesthesia reports in congenitally blind people and rules out the hypothesis that vision is necessary to develop synesthesia. Although vision holds a central role in the synesthetic phenotype (Eagleman and Goodale, 2009), we now show that congenitally blind people can develop synesthesia among the spared senses.

Although congenital visual deprivation does not prevent, tout court, the development of synesthesia, is the incidence of synesthesia reduced among congenitally blind individuals? This is still an open question that only large-scale epidemiological studies can address. As mentioned in the introduction, the hypothesis that congenital blindness may reduce the probability to develop synesthesia is encouraged by recent studies on crossmodal correspondences, a phenomena often related with synesthesia (Bankieris and Simner, 2015; Martino and Marks, 2001; Ramachandran and Hubbard, 2001). For instance, two recent studies could not find reliable shape-sound associations (SSAs) in congenitally blind people tested with a tactile version of the Bouba-Kiki task (Fryer et al., 2014; Sourav et al., 2019). Whereas blindfolded sighted people associated "kiki" with spiky 3D shapes and "bouba" with smooth 3D shapes most of the times, congenitally blind people did not show a preferred association, despite the fact that shape can also be experienced haptically. Other studies failed to find shape-pitch (i.e., high pitch – spiky shape) and space-pitch associations (i.e., high pitch – upward movement) in congenitally blind individuals, whereas these associations emerged strongly in matched control subjects (Deroy et al., 2016; Hamilton-Fletcher et al., 2018). Interestingly, in one study, a group of individuals who regained their sight thanks to visual restoration after congenital cataracts, did not show consistent SSAs (in a bouba-kiki task), neither for haptically nor for visually presented shapes (Sourav et al., 2019), suggesting the existence of a critical period during which lack of vision prevent the formation of SSAs.

However, despite this evidence, concluding that congenital lack of vision hinders the development of crossmodal correspondences (and other forms of sensory crosstalk), appears to be premature. Recent studies have shown that reliable CMCs can be elicited also in early blind subjects (Barilari et al., 2018; Bottini et al., 2019), and suggested that differences between blind and sighted found in previous studies may be due, at least in part, to different graphene-shape associations across the two populations (some written letters like 'D' or 'B' have a round shape in Latin alphabet but not in Braille; Bottini et al., 2019). Moreover, although early blind people may show reduced or absent shape-pitch associations (Hamilton-Fletcher et al., 2018), they seem to develop other forms of CMCs that are not present in the sighted (Hamilton-Fletcher et al., 2018), such as pitch-texture associations (i.e., high pitch – smooth texture; low pitch – rough texture). This result, together with our current findings, suggest that early lack of vision does not prevent, *tout court*, the sensory cross-talk involved in CMCs, multisensory integration or synesthesia, but may simply re-modulate sensory interactions across different domains of experience.

In an additional set of analysis, we showed that letters that are orthographically similar (i.e., similar Braille patterns) also tend to have similar associated textures in CB's synesthetic experience. The association between orthographic and synesthetic similarity has been previously reported in graphene-color synesthetes (Brang et al., 2011) and interpreted as supporting the general hypothesis that synesthesia emerges due to excessive neural connections between cortical regions (Ramachandran and Hubbard, 2001) together with the more specific hypothesis that graphene-color synesthesia is due to cross-activation of graphene and color processing regions in the ventral visual stream (Brang et al., 2010; Brang et al., 2011). This result opens to the possibility that a similar process is taking place in CB's posterior temporal brain regions: Previous studies have shown that Braille characters are processed by the blind in the ventral occipital-temporal word form area, which is activated cross-modally by Braille letters shape (Reich et al., 2011; Siuda-Krzywicka et al., 2016). Potentially, some ventral occipital-temporal regions (not too far from graphene and color processing regions) that are selective for haptic texture representations (Eck et al., 2016; Podrebarac et al., 2014) may be cross-activated with the word-form area in creating synesthetic experiences in CB. Such a configuration will uncover a new pattern of connectivity underlying graphene-related synesthesia, since graphene-texture synesthesia involving only texture and not color has never been reported to our knowledge (one more reason why CB's case is a unique and valid addition to the literature), and previous neuroimaging investigations on color/texture synesthesia did not report activations in texture-sensitive areas, but only in color areas (Eagleman and Goodale, 2009). Another fascinating possibility is that typical color processing regions (i.e., in the V4-complex; Zeki and Marini, 1998) which are not processing colors in congenitally blind individuals (Bottini et al., 2020; Wang et al., 2020), have been co-opted for haptic texture processing in CB's brain thanks to cross-modal plasticity (Frasnelli et al., 2011).

Thus, the correlation between texture and orthographic similarity in CB's experience is consistent with a cross-activation account of developmental synesthesia. However, alternative explanations based on higher-level semantic associations (Chiou and Rich, 2014) cannot be completely ruled out. First, the correlation between orthography and synesthetic experience does not explain why a particular texture gets associated with a particular Braille pattern in the first place (Root et al., 2018). Moreover, according to CB′ subjective report, number synesthesia emerged when CB was about 4 years old, two-three years before he learned the Braille alphabet. This may explain why the effect of orthographic similarity is not significant in the number domains (although this result needs to be taken cautiously because of the low number of items tested: 10 numbers vs 26 letters). On the contrary, letter synesthesia emerged later, probably around 6 years old (CB personal communication), at least partially overlapping with the acquisition of Braille orthography. Nevertheless, as noted by Chiou and Rich (Chiou and Rich, 2014) the observed correlation with orthography can in principle be explained by a simple learning mechanism: A synesthete may learn a new graphene (or braille pattern) based on its similarity to a graphene learnt earlier, inheriting also its synesthetic associations. Further studies involving neuroimaging investigations may help disentangling between the possible neural mechanisms underlying CB's synesthesia and synesthetic experience in general.

Credit author statement

Roberto Bottini: Conceptualization, Methodology, Formal analysis, Investigation, Writing – original draft, Writing – review & editing, Supervision. **Elena Nava**: Methodology, Investigation, Writing – review & editing. **Isabella De Cuntis**: Methodology, Investigation, Writing – review & editing. **Stefania Benetti**: Methodology, Writing – review & editing. **Olivier Collignon**: Methodology, Writing – review & editing, Supervision, Funding acquisition.

Open practices statement

The experiment reported in the paper was not formally preregistered; De-identified data for the experiment along with the data analysis scripts are posted at [https://osf.io/a49dp/]; access to the data is fully open access. The materials used in these studies are widely available.

Declaration of competing interest

The authors have no competing interests to declare.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.neuropsychologia.2022.108226.

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