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# Audio-visual temporal perception in children with restored hearing

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ABSTRACT

It is not clear how audio-visual temporal perception develops in children with restored hearing. In this study we measured temporal discrimination thresholds with an audio-visual temporal bisection task in 9 deaf children with restored audition, and 22 typically hearing children. In typically hearing children, audition was more precise than vision, with no gain in multisensory conditions (as previously reported in Gori et al. (2012b)). However, deaf children with restored audition showed similar thresholds for audio and visual thresholds and some evidence of gain in audio-visual temporal multisensory conditions. Interestingly, we found a strong correlation between auditory weighting of multisensory signals and quality of language: patients who gave more weight to audition had better language skills. Similarly, auditory thresholds for the temporal bisection task were also a good predictor of language skills. This result supports the idea that the temporal auditory processing is associated with language development.

## 1. Introduction

Our five senses provide complementary information about the environment, which needs to be combined to yield a single percept of the world. Integration of the different modalities develops steadily during childhood through to scholastic years (Adams, 2016; Gori et al., 2008, 2012a, 2012c; Nardini et al., 2008; Petrini et al., 2015). In children, reliability-based multisensory integration seems to develop late, after 8–10 years of age: before then, one sense (such as haptic) dominates the other (such as vision) (Gori et al., 2008, 2012a, 2012c). The absence of one sensory modality, such as audition, during the first period of life, induces profound modifications of interactions with the environment (Hensch, 2005; Merabet and Pascual-Leone, 2010). Many studies have investigated the impact of deafness on brain development in humans and animals (for a review Bulter and Lomber (2013), Cardon et al. (2012), Friston (2009), Kral and Sharma (2012)). Auditory experience in the first period of life is fundamental for the maturation and organization associated with speech perception and production (see Kral (2007) for a review), and for the development of the remaining visual and somatosensory modalities (Bavelier and Hirshorn, 2010; Dye and Bavelier, 2010; Gori, 2015; Gori et al., 2010, 2014, 2012c). This is particularly true during the first 3 years (Bavelier et al., 2006; Gori et al., 2010, 2012c), when the differentiation within multi-sensory areas is established (Levanen et al., 1998). In the absence of one sensory signal, such as audition, compensatory mechanisms develop (Bavelier et al., 2006; Bavelier and Neville, 2002), enhancing the skills of the remaining senses, such as vision (Bavelier et al., 2006; Neville and Lawson, 1987), providing benefits in those aspects that are integrated with the auditory sense in typical subjects (Bavelier et al., 2006). On the other hand, some deficits have been observed in non-auditory modalities when the auditory input is absent suggesting that it is necessary for the development of these skills (see Dye and Bavelier, 2010 for an exstensive review on the two compelling "compensatory" and "deficitary" theory).

When audition is restored, such as with cochlear implants, audio processing needs to be established (Lee et al., 2001). Restoration of audition requires the brain to develop new functional interactions with the other modalities, such as vision, and provide a new resource for acquisition of language skills (Giraud and Lee, 2007; Giraud et al., 2001). Although, functional gain is optimized when implantation occurs in the first 3 years, the gain in processing capacity is maximal before the end of the sensitive period of plasticity, around the age of 7 years (Lee et al., 2005, 2007; Sharma et al., 2009).

Several researchers have investigated visual-auditory multisensory integration in children and adults with cochlear implants by studying speech perception (e.g. Bergeson et al., 2010; Doucet et al., 2006; Rouger

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et al., 2008; Schorr et al., 2005). For example, Schorr et al. (2005) investigated the McGurk effect (McGurk and MacDonald, 1976), in which typical observers show clear evidence of multisensory fusion. In 92% of children with implants visual perception dominated when visual and auditory speech conflicted, but, in those children who did not show visual dominance, bimodal fusion was strong and consistent. The authors found that auditory-visual fusion declined with age of implant (beyond 2.5 years), concluding that this is a sensitive period for bimodal integration in speech perception (Schorr et al., 2005). The McGurk effect is typically illustrated by perception of simple monosyllabics (ba/da etc), which requires little in the way of language processing. On the other hand, it is a speech task involving associations between meaningful sounds and lip movements. No studies to date have investigated the impact of auditory deprivation in simpler temporal multisensory situations. Since audition is fundamental for temporal processing, one might expect that auditory deprivation during development might compromise simple, semantically unrelated, sensory processes.

Here we investigate unisensory and multisensory temporal perception in the developing deaf child with restored hearing, using a visual-auditory temporal task. In particular, we compared temporal perception of deaf children with CI implanted before the age of 5 years with a group of typical children. We also correlated the individual temporal capabilities of children with restored hearing with their language skills. Firstly, we investigated how the new audio temporal information is processed and combined with the visual input after a period of auditory deprivation. Then, since auditory experience is fundamental for speech perception and production, we investigated how the restored temporal processing is linked to language skills in deaf children with hearing restored. To study integration, we used the Bayesian approach, which predicts that different sensory inputs are combined after weighting for reliability. The presence of multiple sensory signals (such as the visual and the auditory ones) provides a gain in the precision of the multimodal estimation. The Bayesian approach has been shown to be a powerful method to predict multisensory gain in many tasks (Alais and Burr, 2004; Clarke and Yuille, 1990; Ernst and Banks, 2002; Ghahramani et al., 1997; Landy et al., 2011). Our results suggest that deaf children with restored hearing have much less auditory dominance than typical children. Importantly, the deaf children with restored hearing who with higher auditory dominance and lower auditory bisection thresholds had better language skills. These results suggest that temporal auditory dominance can be important for the development of language skills and that simple semantically unrelated stimuli, such as the bisection task used here, can be used as language screening.

## 2. Methods

### 2.1. Participants

27 children without sensory disabilities (aged between 7 and 13 years) and 9 children with congenital or early-acquired sensori-neural hearing loss (aged between 7 and 13 years), with hearing supplemented first by auditory amplification prostheses, and then by cochlear implants (before the age of 5 years, see Table 1 for more details), performed the temporal bisection tasks illustrated in Fig. 1. Five typical children were removed from the analysis because they were defined as outliers (after performing a chi-squared test for detection of outliers, by using the package: Lukasz Komsta, 2011; Outliers: tests for outliers. R package version 0.14. https://CRAN.R-project.org/package=outliers). 22 children without sensory disabilities were included in the analysis. Typically developing children were recruited from elementary and intermediate schools in Genoa. Children with sensory disabilities were tested at the Stella Maris Scientific Institute in Pisa. The participants, and their parents, provided written informed consent in accordance with the Declaration of Helsinki. The study was approved by the ethics committees of the local health services *(Comitato Etico, ASL3 Genovese, Italy and Comitato Etico, IRCCS Fondazione Stella Maris 36/2010)*.

### 2.2. Stimuli and procedure

Three different instances of a particular stimulus type (visual, auditory or both) were presented sequentially for a total duration of 1000 ms. Observers were required to indicate by button-press whether the middle stimulus appeared closer in time to the first or the third stimulus (temporal bisection). In the visual task (Fig. 1C) the subject was presented with a sequence of three lights, red, yellow and green. The subject had to respond whether the second (yellow) light appeared closer in time to the first (red) or to the last (green) light. Similarly, in the auditory task (Fig. 1D) the subject had to respond if the second sound was presented closer in time to the first or to the third. In the bimodal task (Fig. 1E) the subject perceived a sequence of three lights associated with three sounds (like the uni-sensory stimuli). The visual and the auditory stimuli could be presented either at the same time, or "in conflict", with the auditory stimulus presented before or after the visual. The procedure was similar to that of Burr et al. (2009) and identical to that of Gori et al. (2012a): in the second stimulus the light was presented before the tone by $\Delta$ ms ($\Delta = 0$, or $+50$ ms), while in the first and the third stimulus the offset was inverted in sign, so the light was presented before the tone by $-\Delta$ ms. We varied the timing of the second stimulus (tone and flash together) to span the interval between the first and third stimuli. We used a child-friendly setup (Fig. 1A and B), which presented the sequence of three lights (red, green and yellow) and sounds (by the speakers behind the clown), or both. The visual stimuli were 1° diameter LEDs displayed for 75 ms. Auditory stimuli were 750 Hz tones played for 75 ms. Accurate timing of the visual and auditory stimuli was ensured by setting priority in the operating system to maximum during stimulus presentation to thereby avoid interrupts by other processes (checked by calibration with light sensor and microphone). Before collecting data, subjects were familiarized with the task with two training sessions of 10 trials each (one visual and one audio), where subjects indicated after each presentation of the three stimuli whether the second appeared earlier or later than the midpoint between the first and third stimuli (as in the main experiment). We provided feedback during these training sessions so observers could learn the task and minimize errors in their responses. No feedback was given after the training sessions. During the experiment proper, 5 different conditions were intermingled within each session: vision only, auditory only, and three two-cue conditions. The total session comprised 150 trials (30 for each condition).

The time of presentation of the probe was varied by independent QUEST routines (Watson and Pelli, 1983), which homed in on the point of subjective equality (PSE): the time offset for which the second stimulus on average appeared to bisect the first and third stimuli. The QUEST estimate was perturbed by adding a random number to ensure that the psychometric function was well sampled over its entire range, important when estimating both the PSE and slope. It also gave observers a few encouraging "easy" trials from time to time. Data for each condition were fitted by cumulative Gaussians, yielding PSE and threshold estimates from the mean and standard deviation of the best-fitting function (see Fig. 2 and 5). Standard errors for the bisection PSE and threshold estimates were obtained by bootstrapping (Efron, 1993). All conflict conditions were used to obtain the two-cue threshold estimates.

We used R (R Core Team, 2012) and "*nlme*" (Pinheiro et al., 2016) to perform a linear mixed effects analysis of the relationship between thresholds and PSEs, and group and condition. As fixed effects, we entered group, condition and their interaction into the model. As random effects, we had intercepts and slope for subjects, as well as by-subject random slopes for the effect of condition.

We managed heteroschedasticity by applying the function *lme* of the package *nlme* with the argument "*varIdent*" (Pinheiro, 2006) set in a way to allow a different variance for each group. p test value were obtained using Analysis of Deviance Table.

Post hoc comparisons were obtained by means of *t*-tests accounting

**Table 1**
clinical details of the children involved in the study. CI = Cochlear implantation PTA = Pure Tone Audiometry between 0,5-1-2 kHz measured in decibels (Db); The value is the average of the audio threshold at 500, 1000 and 2000 Hz for each child.

| Subj. | Age at test | Etiology | Age at first hearing aids fitting | Age at CI | CI Side | Unaided PTA | PTA with hearing aids (pre CI) | PTA with CI |
|---|---|---|---|---|---|---|---|---|
| S1 | 11 | Prenatal unknown | 9 m | 3 y, 3 m | left | 110 db | 80 | 40 |
| S2 | 12 | Genetical (connexin mutation) | 3 y | 5 y | right | 110 db | 55 | 35 |
| S3 | 7 | Prenatal Unnown | 16 m | 3.2 y | right | 100 db | 60 | 30 |
| S4 | 9 | Genetical (connexinmutation) | 18 m | 2 y 7 m | right | 105 db | 50 | 25 |
| S5 | 8 | Perinatal (acquired infection) | 9 m | 8 y, 6 m | left | 110 db | 50 | 35 |
| S6 | 9 | Prenatal (CMV infection) | 9 m | 2 y, 10 m | left | 120 db | 70 | 30 |
| S7 | 12 | Prenatal (inner ear malformation) | 24 m | 8 y | left | 110 db | 50 | 30 |
| S8 | 13 | Genetical (connexinmutation) | 12 m | 7 y, 4 m | left | 100 db | 60 | 30 |
| S9 | 13 | Genetical (Jervell and Lange syndrome) | 18 m | 3 y, 4 m | right | 100 db | 50 | 25 |

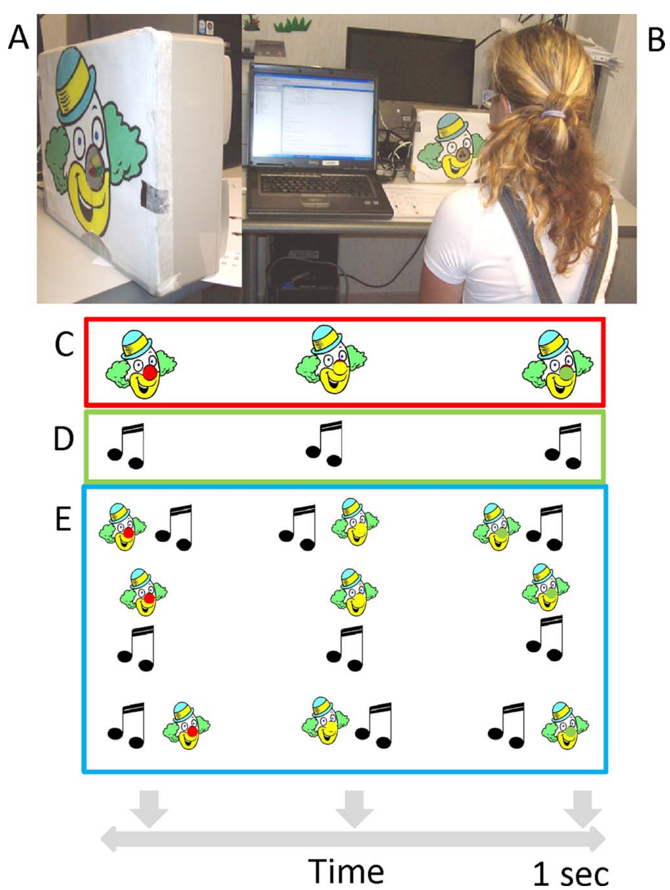


**Fig. 1. Setup.** A. Illustration of the setup with the three LEDs in front and two speakers behind. B. Child performing the task. C. Representation of the visual stimulus. The subject was presented with a sequence of three lights: the first red, the second yellow and the third green. The task of the subject was to respond whether the second light appears closer in time to the first or the last one. D. Representation of the auditory stimulus. The subject was presented with three sounds. The task of the subject was to respond if the second sound was presented closer in time to the first or the last one. E. Representation of the bimodal stimulus. The subject was presented with a sequence of three lights (as in C) together with three sounds (like D). The visual and the auditory stimuli were presented with a temporal offset of $+\Delta$ (= 0, ± 50 ms) in the second stimulus, -Δ in the first and third: see Section 2 for more details). (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article).

for different variances of the groups (Satterthwaite tests) and we corrected p values using the Bonferroni method (i.e by multiplying them by the number of comparisons). In the text we report corrected p value and we retain as significant those p values that were $< 0.05$.

### *2.3. Language tests*

In the children with sensory disabilities we performed four tests to evaluate their language skills.

1. Expressive vocabulary was tested by means of the One-Word Picture Vocabulary Test (OWPVT, Brizzolara, 1989) in which 104 pictures corresponding to high (52) and low (52) frequency words are presented. In each display, the child must name each of four simple, black-and-white pictures. Children who performed within 2 standard deviation of the mean were considered within the normal-border range (cut-off −2 standard deviations) for this test (OWPVT, Brizzolara, 1989) were assigned one point in the final language aggregate score calculation.
2. Receptive vocabulary was tested by means of the Peabody Picture Vocabulary Test PPVT-R (Revised 3rd Edition, Dunn and Dunn, 1997). This is an untimed, individually administered test with an oral presentation of 5 training items followed by 175 test items arranged in an order of increasing difficulty. Each item has four simple black and white illustrations arranged in a multiple-choice format. The task is to select the picture that best illustrates the meaning of an orally presented word-stimulus. Children who reached the threshold to be considered within the normal range (QL ≥85) for this test (as defined in Revised 3rd Edition, Dunn and Dunn, 1997) were assigned one point in the final language aggregate score calculation.
3. Expressive grammar was clinically evaluated by analyzing language samples collected during a narrative task. Morphological organization was evaluated in terms of omissions and errors in terms of a substitution of free or bound morphemes (percentage of substitutions of free morphemes and bound morphemes). The analysis of the syntactic organization of the narratives produced by the participants included a measure of utterance length and of syntactic complexity. A test of repetition of sentences with clitic pronouns, designed by Bottari et al. (1998) for the study of morphological development in Italian children was also administered. Clitic pronouns are monosyllabic unstressed free morphemes that are bound to inflected verbs or auxiliaries. The position of clitics is determined grammatically as they may either precede or follow the inflected form according to a

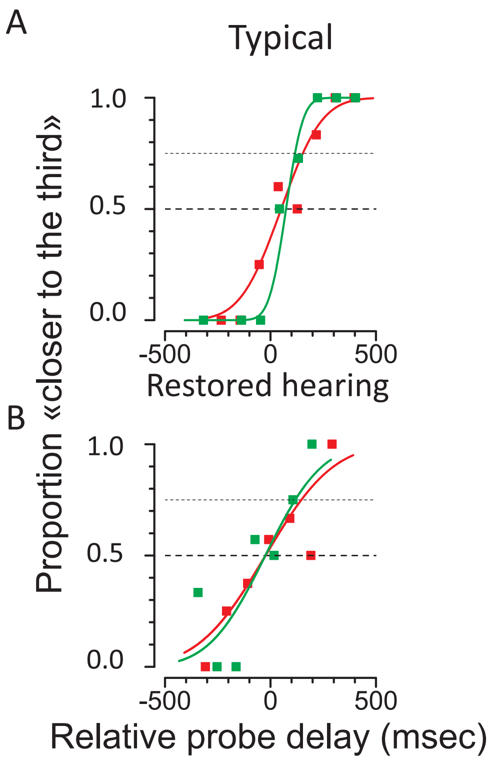


**Fig. 2. Unisensory visual and audio psychometric functions.** Example of visual and auditory psychometric functions for two children: proportion of responses closer to the third stimulus are plotted against the probe offset relative to stimulus midpoint. **A.** Typical child, aged 9 years. **B.** Deaf child with cochlear implant, aged 9 years. The red symbols and lines represent the visual condition and the green symbols and lines the auditory condition. The black dashed horizontal lines show the 50% performance point, intersecting with the curves at their PSE. The grey dashed horizontal lines show the 75% performance point, intersecting with the curves determining their JND: as the difference with the PSE. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article).

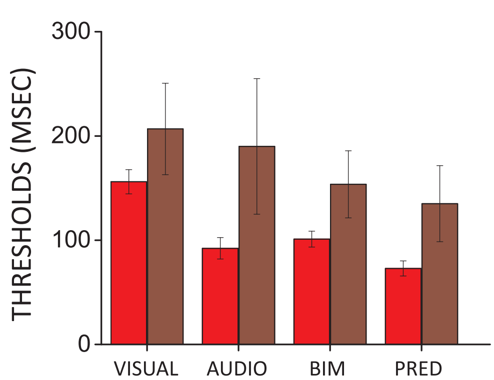


**Fig. 3. Average of unisensory visual and audio and bimodal thresholds.** Average thresholds: visual, audio, Audio-visual and Bayesian prediction. Lighter bars represent the typical group (N = 22), darker bars the deaf group (N = 9).

complex set of syntactic rules. Clitics are organized into a rich paradigm in which forms vary according to person, gender, number and case features. The test consists of 30 six-word sentences, containing a total of 24 clitics, 44 articles, and 10 prepositions. Ten different contexts for clitic insertion were selected to reflect the general properties of clitics in main clauses. For each of these ten contexts a sentence containing a full noun phrase instead of the clitic pronoun is also provided. Children who reached the threshold to be considered within the normal-border range (threshold within 2 standard deviations of the mean) for this test as defined in Bottari et al. (1998) were assigned one point in the final language aggregate score calculation.

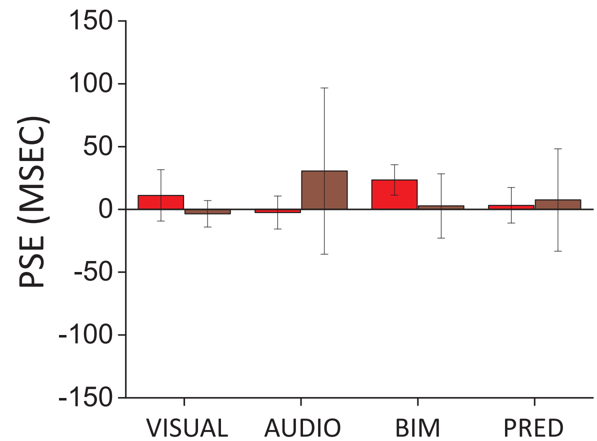


**Fig. 4. Average of unisensory visual and audio and bimodal PSEs.** Average PSEs: visual, audio, Audio-visual and Bayesian prediction. Lighter bars represent the typical group (N = 22), darker bars the deaf group (N = 9).

4. Sentence comprehension was also tested using a pictured multiple-choice comprehension grammar test for children (TCGB) (Chilosi, 2006) which examines the child's ability to understand orally presented 8 different type of grammatical structures (locative, nominal-verbal inflections, affirmative and negative active and passive sentences, relatives, datives); it includes 76 requests. Children who reached the threshold to be within the normal-border range for this test as defined in Chilosi (2006)) were assigned one point in the final language aggregate score calculation.

The sum of the scores on the four tests was used to calculate the language aggregate score used in the plots in Fig. 8: 0 – all tests failed; 1 – one test performed over threshold to be considered within the normal range; 2 – two tests performed over threshold to be considered within the normal range; 3 – three tests performed over threshold to be considered within the normal range; 4 – all tests performed over threshold to be considered within the normal range.

### 2.4. Bayesian predictions

The MLE calculations used in this study assume that the optimal bimodal estimate of PSE ($\hat{S}_{VA}$) is given by the weighted sum of the independent Audio and visual estimates ($\hat{S}_V$ and $\hat{S}_A$).

$$\hat{S}_{VA} = w_V \hat{S}_V + w_A \hat{S}_A \quad (1)$$

where weights $w_V$ and $w_A$ sum to unity and are inversely proportional to the variance ($\sigma^2$) of the underlying noise distribution, assessed from the standard deviation σ of the Gaussian fit of the psychometric functions for visual and audio judgments:

$$w_V = \sigma_A^2/(\sigma_A^2 + \sigma_V^2), \quad w_A = \sigma_V^2/(\sigma_A^2 + \sigma_V^2) \quad (2)$$

The MLE prediction for the visuo-audio bisection threshold ($\sigma_{VA}$) is given by:

$$\sigma_{VA}^2 = \frac{\sigma_V^2 \sigma_A^2}{\sigma_V^2 + \sigma_A^2} \le \min(\sigma_V^2, \sigma_A^2) \quad (3)$$

where $\sigma_V$ and $\sigma_A$ are the visual and audio unimodal thresholds. The improvement is greatest (factor of $\sqrt{2}$) when $\sigma_V = \sigma_A$.

To calculate the visual and audio weights from the PSEs, we substituted the actual times (relative to standard) into Eq. (1):

$$\hat{S}(\Delta) = (w_V \Delta - w_A \Delta) = (1 - 2w_A)\Delta \quad (4)$$

The slope of the function is given by the first derivative:

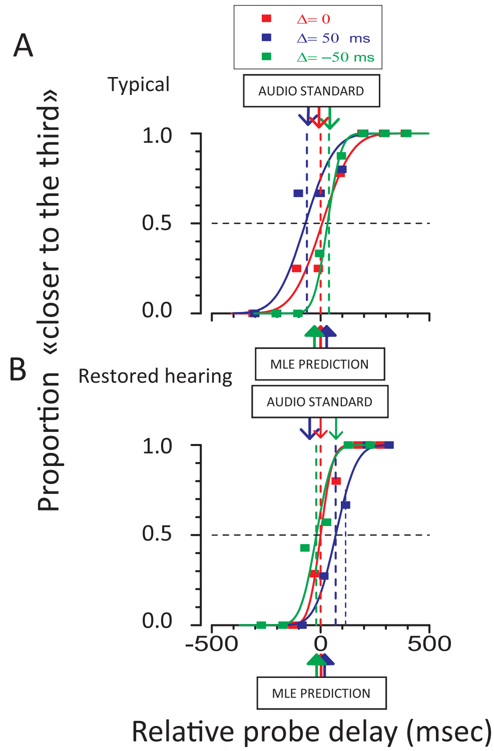


**Fig. 5. Example psychometric functions for two children, with various degrees of cross-modal conflict. A.** Typical child, aged 9 years. **B.** Deaf child with cochlear implant, aged 9 years. The lower colour-coded arrows show the MLE predictions, calculated from threshold measurements (Eq. (3)). The black dashed horizontal lines show the 50% performance point, intersecting with the curves at their PSE (shown by short vertical bars). The upper colour-coded arrows indicate the delay of the audio standard. The typical children generally were dominated by auditory information, while the deaf followed the Bayesian prediction integrating visual-auditory cues. The amount of conflict was 0 for the red symbols, +Δ ms (where plus means vision was later) for the blue symbols and –Δ ms for the green symbols. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article).

$$\hat{S}(\Delta)' = 1 - 2w_A \tag{5}$$

Rearranging:

$$w_A = (1 - \hat{S}(\Delta)')/2 \tag{6}$$

The slope $\hat{S}(\Delta)'$ was calculated by linear regression of PSEs for all values of Δ, separately for each child.

## 3. Results

Fig. 2 reports examples of psychometric curves for a typically developing child (A) and a congenitally deaf child with hearing restored (B), for visual and auditory discriminations. For all children and conditions, we plot the proportion of trials where the second stimulus was reported to be closer to the third as a function of the presentation time of the second stimulus (relative to mid-point). The curves were then fit by gaussian error functions, shown by the continuous curves. The standard deviation of the fitted curves is taken as an estimate of threshold. In the example shown, the typically developing child had a lower auditory than visual threshold, as the psychometric function for the auditory task is steeper than for the visual task; however, for the deaf child with hearing restored, the two psychometric functions were almost superimposed, reflecting similar thresholds.

We performed the analysis with two separate mixed models with an analysis of deviance for thresholds and PSEs. As fixed effects, we entered group, condition and their interaction into the model. As random effects, we had intercepts and slope for subjects, as well as by-subject random slopes for the effect of condition. Considering the thresholds (Fig. 3 shows the average visual and audio bisection thresholds for the two groups of subjects considered) a main effect for condition (chi-square (3) = 66.92, $p < 0.0001$), and a main effect for interaction group X condition (chi-square (3) = 7.83, p = 0.04) was found. A Bonferroni post hoc test revealed that in deaf children with restored hearing, auditory thresholds were high, about the same as visual thresholds (two-tailed, paired *t*-test t(14.04) = 0.23, p = 0.82, Fig. 3). Contrarily, in typical children auditory thresholds were lower than visual thresholds (two tailed, paired *t*-test t(41.42) = 4.11, p = 0.002, Fig. 3), implying higher auditory precision for temporal perception, as previously reported for both adults and children (Gori et al., 2012b; Burr et al., 2009). Audio thresholds of deaf children with restored hearing were statistically higher than those of the control group (compare light and dark red bars in Fig. 3, (two-tailed, paired *t*-test t(29) = 2.35, p = 0.02), but not vision thresholds (two-tailed, paired *t*-test t(29) = 1.6, p = 0.06). For the PSEs (Fig. 4) no effect was observed for group, neither for condition and nor for their interaction.

We next consider the Audio-visual experiments, where both tones and lights were presented, either together or "in conflict". Fig. 5A and B show example psychometric curves for a typically developing child (A) and a congenitally deaf child with hearing restored (B), for three levels of conflict. The relative delays of the "auditory standard" are shown by the colour-coded arrows above the graphs. Here we are interested both in the threshold (again given by the standard deviation) and also the point of subjective equality (PSE), given by the mean of the error functions (the 50% point). The bisection PSEs clearly depend on the conflict, but in a different way for the typical and the deaf child with restored hearing. For the typical child, the curves are not superimposed but follow the auditory stimulus, suggesting high auditory weighting for the task. The blue curve (positive conflict +Δ) moves to the left, and the green (negative conflict −Δ) moves to the right, both tending to align with the auditory standards. For the child with restored hearing, the pattern was quite different: the green curve is virtually superimposed on the red (suggesting equal auditory and visual weights) while the blue shifts to the right, away from the auditory standard, towards the symmetrically opposite visual standard. This suggests that the auditory weighting is much less. The arrows below the functions show the MLE predictions from the thresholds, following Eqs. (1) and (2). In both cases, the MLE predicts the direction of the shift in the graphs.

The pattern observed in the example psychometric functions generalized to the subject pool. For each subject, we calculated the relative weighting for vision and audition, following the procedure detailed in Section 2 (Eqs. (2) and (6)). Fig. 6 plot these weights calculated from the PSEs and thresholds. The typical group showed a strong auditory dominance, with average weights calculated from the PSEs of 0.82 (weights of 1 mean total auditory dominance, 0.5 equal weighting to vision and audition). A main effect was observed for group (chi-square (1) = 5.34, p = 0.02) and for condition (chi-square (1) = 5.12, p = 0.02) and for their interaction group X condition (chi-square (1) = 3.99, p = 0.04). A Bonferroni post hoc test revealed that in typical children weights predicted from thresholds were higher than those predicted from PSEs (two tailed, paired *t*-test t(40.8) = −2.57, p = 0.02). In deaf children with restored audition, weights predicted from thresholds were not different from those predicted from PSEs (two tailed, paired *t*-test t(15.46) = 0.28, p = 0.78). Weights predicted from thresholds were significantly higher in typical that in deaf children with hearing restored (two tailed, paired *t*-test t(10.31) = −2.45, p = 0.03).

Fig. 3 replots the threshold for the Audio-visual presentation, together with the Bayesian predictions for the combination (Eq. (3)). There is some evidence for multisensory gain in the group of deaf children with restored hearing (Fig. 3). A Bonferroni post hoc test revealed that in hearing restored children, bimodal thresholds did not significantly differ from the MLE prediction (two-tailed, paired *t*-test t(8) = 1.30, p = 0.40), consistent with Bayesian predicted gain. Contrarily, in the typical group bimodal thresholds were significantly higher from the MLE prediction (two-tailed, paired *t*-test t(21) = 2.50, p = 0.04) and were not lower than auditory thresholds (two tailed, paired *t*-test t(21) = −0.63, p = 1) consistent with previous research (Gori et al., 2012b).

We then asked if the auditory thresholds and weights predict the important practical auditory skill of language acquisition. Linguistic skills were assessed in all patients by trained clinicians, who made routine assessments of language based on expressive vocabulary, receptive vocabulary, expressive grammar and receptive grammar (see Section 2). Fig. 7A plots language performance against auditory weight (calculated from PSE) for all subjects. Language performance correlated strongly with auditory weights (linear fit, r = 0.92; p = 0.02, see Fig. 7A). There was a clear cut-off in performance: almost all subjects with weights less than 0.74 had poor language (close to 0, indicating all language tests failed), while those with higher weights had good language (close to 4, indicating all the 4 tests performed within the typical range). The difference between these two groups was highly significant (one tailed, paired *t*-test, t(7) = 6.25, p < 0.0001). Fig. 7B plots language performance against audio bisection thresholds. Again, language correlated well with auditory thresholds (linear fit, r = 0.76; p = 0.016), and again there was a clear division, with lower thresholds (less than 150 ms) predicting better language performance (one tailed, paired *t*-test, t(8) = 6.25, p < 0.0001).

No significant correlations were observed between language skills and age at first prosthesis (Fig. 8A, linear regression, r = 0.33; p = 0.37), age of cochlear implantation (Fig. 8B, linear regression, r = 0.07; p = 0.85) or time since implantation (Fig. 8D, linear regression r = 0.5; p = 0.2). Marginally significant correlations were observed with time since first prosthesis (Fig. 8C, linear regression, r = 0.57; p = 0.1)0 and with chronological age (Fig. 8E, linear regression, r = 0.6 p = 0.06). Similarly, no correlations were observed between audio weight and chronological age (Fig. 9A, linear fit, r = 0.30; p = 0.35) and marginally between audio threshold and chronological age (Fig. 9B, linear fit, r = 0.70; p = 0.05). When corrected by for age, no correlation was found between language capabilities and audio weight (linear fit, r = 0.56 p = 0.12) and audio thresholds (linear fit, r = 0.41 p = 0.27). All this suggests that the correlations between language skills and auditory weights and thresholds are not simple by products of the time since implantation of the device, or other confounding variables.

## 4. Discussion

The absence of one sensory modality – such as audition – during the first period of life (expecially during the first three years of age) modifies the typical manner of interacting with the environment in humans and in animals (Cardon et al., 2012; Friston, 2009; Kral and Sharma, 2012; Lazard et al., 2014; Merabet and Pascual-Leone, 2010; Strelnikov et al., 2010). Our results support this idea showing that the lack of earlier auditory experience impacts on temporal discrimination in deaf children with restored hearing: they do not show the auditory dominance that typical children do. More importantly, we observed a strong correlation between language capabilities and both auditory thresholds and auditory dominance; no significant correlations were found between audio weights or thresholds and age. No correlation was found between language capabilities and audio weight and audio thresholds, when corrected for age. This suggests a strict link between audio dominance in Audio-visual temporal integration and language capability. Restored hearing children who gave more weight to the auditory modality (and had lower audio temporal thresholds), similar to typical children, performed better at the language tests than those who gave lower weight to the auditory modality (and had higher audio thresholds).

Compensatory theories (Bavelier et al., 2006; Bavelier and Neville, 2002) hold that the absence of audition can enhance the sensitivity of the remaining senses, such as vision (Bavelier et al., 2006; Bavelier and Hirshorn, 2010; Neville and Lawson, 1987). This benefit is particularly evident for the visual cues that are integrated with the auditory sense in typical subjects (Bavelier et al., 2006). Temporal audio signals are typically integrated with visual signals for many everyday tasks, such as reading and speaking. Speech, for example, is a bimodal percept for which it is necessary to associate the audio information of the voice with the visual information of the lip movements of a speaker (Schorr et al., 2005). Time discrimination in the deaf is particularly interesting because audition is fundamental for time perception of brief interval times, dominating the final percept in many multisensory temporal tasks (e.g. Bresciani and Ernst, 2007; Burr et al., 2009). The lack of audition in the first years of life can impact on the typical link that is naturally established in the first period of life (Adams, 2016; Gori et al., 2012b; Nardini et al., 2008; Petrini et al., 2015).

Audio-visual multisensory integration and in particular auditory temporality is fundamental for efficient language acquisition

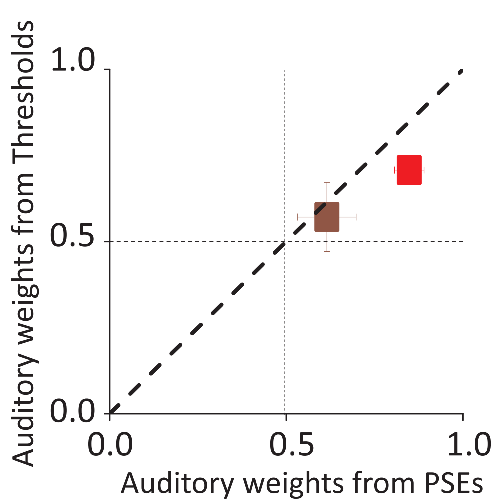


**Fig. 6. Average auditory weights predicted from PSEs against from thresholds.** Average auditory weights predicted from thresholds against from PSEs. The typical group (N = 22) is presented in light red and the deaf group in dark red (N = 9). (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article).

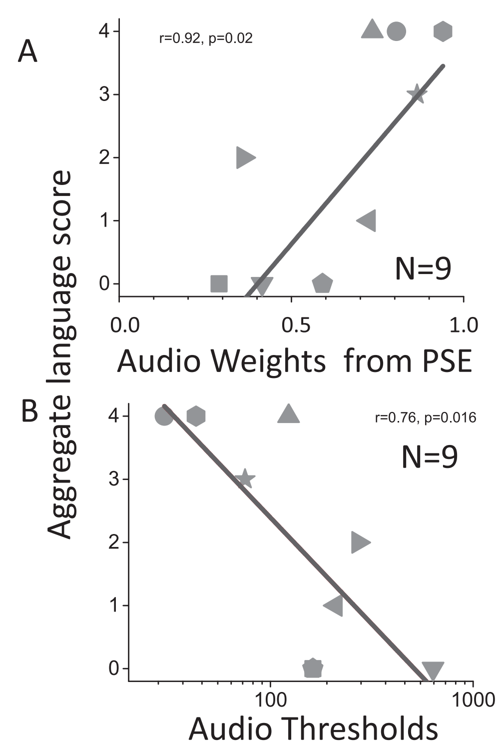


**Fig. 7. Audio weighting and precision predict language skills. A.** Language performance as a function of auditory weights (derived from PSEs). **B.** Language performance as a function of audio bisection thresholds. Continuous red lines represent the linear regression of the data. See Section 2 for a full description of how language was evaluated.

(Megevand et al., 2013; van Wassenhove, 2013; van Wassenhove et al., 2005, 2007). Audio-visual temporal links that are established naturally (Desantis and Haggard, 2016; Martin et al., 2015; VanRullen et al., 2014) have a key role on the role of audition on language development (Cardon et al., 2012 for a review). Auditory experience in the first period of life is fundamental for the maturation and organization associated with speech perception and production (see Kral, 2007 for a review). Many studies have investigated Audio and visual integration, showing specific processing for Audio-visual speech integration. For example it has been shown that visual speech speeds up the cortical processing of auditory signals (van Wassenhove et al., 2005). Several researchers have investigated visual-auditory multisensory integration in deaf children and adults with cochlear implants by studying speech perception (e.g. Bergeson et al., 2010; Doucet et al., 2006; Rouger et al., 2008; Schorr et al., 2005).

Since speech perception is complex, both for the auditory and visual systems, we investigated simple and semantically neutral stimuli. It is well known that the lack of audition directly impacts language capabilities of deaf children with restored hearing. Auditory experience in the first period of life (Levanen et al., 1998), is fundamental for the maturation and organization associated with speech perception and production (see Kral (2007) for a review), and for the calibration of the remaining visual and somatosensory modalities (Bavelier et al., 2006; Bavelier and Hirshorn, 2010; Gori, 2015; Gori et al., 2010, 2012c). The use of cochlear implant can recover auditory skills but it has been shown that impacts of pitch-based discriminations (Galvin et al., 2007; Kang et al., 2009; Kong et al., 2004; McDermott, 2004; Zeng et al., 2008, 2014).

Our data indeed suggest that the reacquisition of auditory input cannot immediately compensate for the changes produced by the previous lack of audition, even though the children in our group were implanted before 7 years of age, towards the end of the sensitive period of plasticity (Giraud and Lee, 2007; Lee et al., 2007; Sharma et al., 2009). Our results agree with the idea that restoration of audition requires the brain to develop new functional interactions with the other modalities, such as vision in this specific temporal domain. Only when the deaf children with restored hearing had acquired auditory dom-

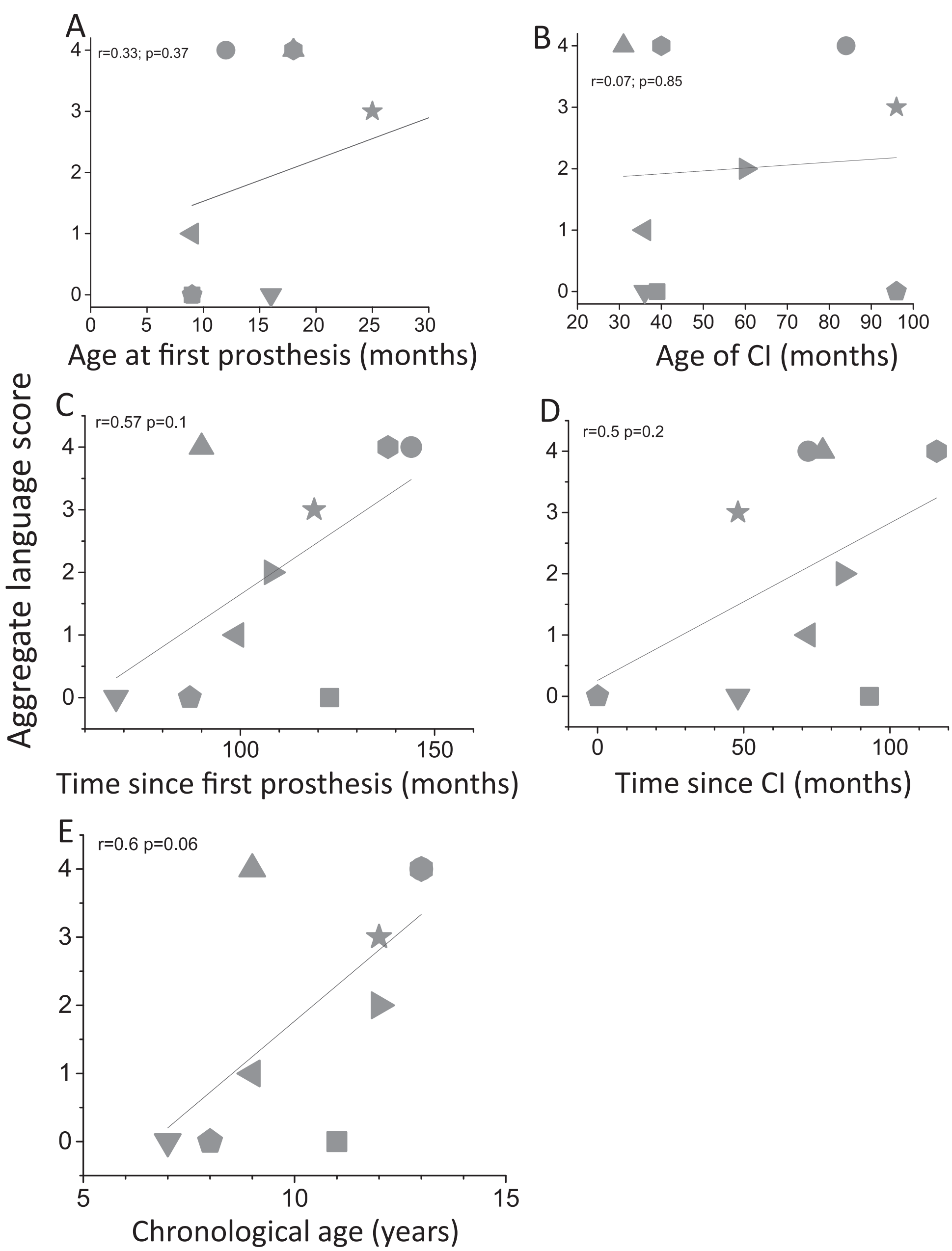


**Fig. 8. Audio thresholds as a function of auditory experience.** Auditory thresholds as a function of: **A.** Age of CI (months); **B.** Age at first prosthesis (months); **C.** Time since CI (months); **D.** Time since first prosthesis (months). **E.** Chronological Age. All data have been fitted with liner functions.

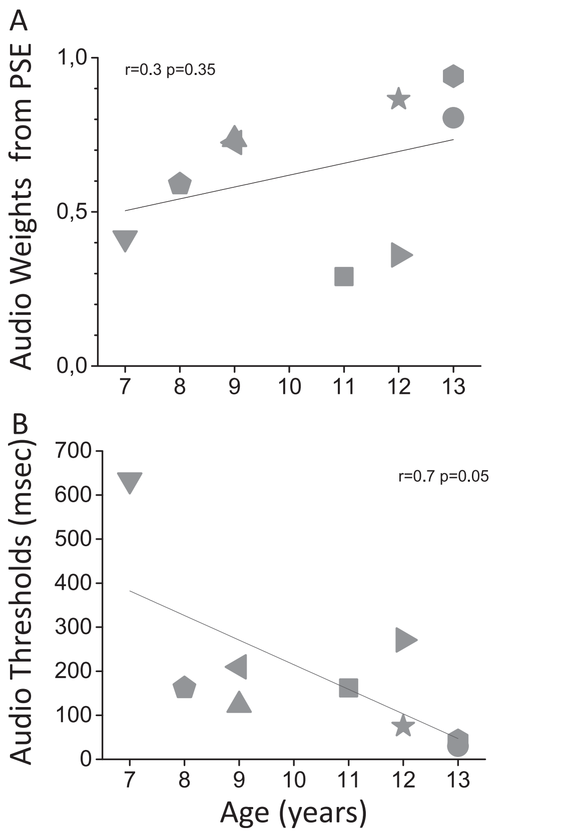


**Fig. 9. Audio thresholds as a function of age.** A) Auditory weights as a function of age. B) Auditory bisection thresholds as a function of age. All data have been fitted with liner functions.

inance for temporal bisection, similar to the typical children, did they acquire good language skills.

There is now some evidence, both from animal and human studies, that in profoundly deaf individuals auditory cortical areas are functionally colonized by the visual system (Bavelier and Hirshorn, 2010; Buckley and Tobey, 2011; Campbell and Sharma, 2016; Fine et al., 2005; Finney et al., 2001, 2003; Kok et al., 2014). On the other hand, other authors have reported little change of the auditory neural structures in deaf animals (e.g. Clemo et al., 2016) and others show very few novel connections between visual and auditory cortices as a result of deafness (e.g. Butler et al., 2016; Chabot et al., 2015). On restoration of auditory sensory input, the auditory system would need to regain use of the colonized areas to function as the brain does in normal hearing listeners (e.g. Buckley and Tobey, 2011; Doucet et al., 2006). This process could involve considerable rewiring of neural circuitry, which could be expected to proceed slowly. It is also likely that Audio-visual multisensory interactions are important in consolidating the reacquisition of neural territory for auditory processing. Many works highlighted a strong and significant correlation between the improvement of language capabilities and auditory experience, suggesting that the auditory-visual system is highly plastic, but that changes take time to develop (Doucet et al., 2006; Goh et al., 2001; Tyler et al., 1997; Chilosi et al., 2013, 2014). This is true especially for speech restoring precision even after a period of sensory deprivation (Cardon et al., 2012 for a review). Our data show no significant correlations between auditory thresholds and duration of auditory experience (from the time at first prosthesis use) and duration of cochlear implant (although there was a non-significant tendency). Since all the subjects tested in this work received the implantation outside or around the end of the window of time during which complete recovery of function is often observed around 3 years (Levanen et al., 1998), a strong link between time of implant and function would not be expected.

Some studies using speech tasks (e.g. MgGurk effect; McGurk and MacDonald, 1976) also show a positive correlation between cross-modal reorganization and age at which the cochlear implant was inserted (e.g. Bergeson et al., 2010; Doucet et al., 2006; Rouger et al., 2008; Schorr et al., 2005). Here we did not observe any correlation between temporal precision and age at which the implant was inserted. A possible explanation of this discrepancy could be that in our task we used a simple temporal discrimination task in which typically hearing children and adults show a clear auditory dominance (Burr et al., 2009; Gori et al., 2012b), and probably a standard auditory prosthesis can be enough to learn to interpret such information.

Multisensory optimal integration of different modalities develops during childhood through to scholastic years (Gori et al., 2008, 2012a, 2012c). In terms of Bayesian integration, we found that the group of hearing restored deaf children showed optimal integration between audition and vision. This integration is revealed by both auditory weights and bimodal thresholds, and is well predicted by the Bayesian model. Contrarily, controls show auditory dominance in the temporal Audio-visual bisection task, with little evidence of integration (in agreement with Burr et al. (2009), Gori et al., 2012b). Typical bimodal thresholds were significantly different from the Bayesian prediction. These results suggest that the auditory modality is important in constructing an optimal integrated sense of time (as sustained by many authors e.g. Burr et al., 2009; Gori et al., 2012b; Nava et al., 2009). Previous data showed that the lack of audition in deaf people can impact on visual time perception (as suggested by many authors e.g. Nava et al., 2009). This result is also in agreement with the idea that early auditory experience is fundamental for the calibration of the remaining visual and somatosensory modalities (Bavelier et al., 2006; Dye and Bavelier, 2010; Gori, 2015; Gori et al., 2010, 2014, 2012c).

The changes in patterns of multisensory gain and audio dominance observed in this study are also relevant to theories of cross-modal calibration (e.g. Gori et al., 2008, 2012a; Vercillo et al., 2015). The auditory dominance found in typical children could reflect a process of auditory calibration on visual temporal perception (Gori et al., 2012b). Although most recent work on multi-sensory interactions has concentrated on sensory fusion, investigating the efficiency of the integration of information from different senses, an equally important, but somewhat neglected aspect is sensory calibration. We suggest that the reason why young children do not integrate sensory information is that during the early years of development, when their body is undergoing rapid changes affecting in various ways the different sensory channels, they use each channel individually to calibrate the senses to physical reality (Burr and Gori, 2012). Our interpretation is that, in the same way that the more precise sense has the highest weight for sensory integration, the most important property for sensory calibration is accuracy, which is defined in absolute terms as the vicinity of a measurement to its true physical value. Precision, conversely, is a relative measure defined as the degree of reproducibility or repeatability between measurements, usually defined as the standard deviation of the distribution. A good deal of evidence in support of the cross-sensory calibration idea comes from specific patient groups, including congenitally blind children and adults (Gori et al., 2010, 2014), and children with cerebral palsy dyskinesia (Gori et al., 2012c). In deaf children, the process of cross-modal calibration would not prevail. We think that the results presented here support this model showing that in absence of the audio information in the first period of life impacts on the development of an audio based temporal reference system. On the other hand, when audition is restored, perhaps there is no impediment to Audio-visual integration. Audio-visual integration may be fundamental to restoring many important temporal properties to hearing.

In agreement with this idea we observed that in older children the auditory system dominates again, suggesting that the temporal resolution has matured, probably with the help of Audio-visual integration (although no correlation was observed between audio dominance and audio experience). A speculation is that vision might be used as a support to calibrate time when audition is absent (in agreement with e.g. Doucet et al., 2006; Giraud et al., 2001; Green et al., 2005; Tyler et al., 1997). The weighting given to auditory signals in integration of basic multisensory stimuli has important practical implications. Research in adults with cochlear implants has shown that cross-modal reorganization from the visual modality is linked to deficits in speech perception performance (Lazard et al., 2014; Lazard et al., 2013). Importantly, cochlear implants seem to be less successful in restoring language if the operation occurs after subjects have learned sign language, which can colonize the auditory cortex (Lee et al., 2001). We found a very strong correlation between auditory weighting of multisensory signals and quality of language: those who gave more weight to audition had better language skills. Similarly, auditory thresholds for the temporal bisection task were also a good predictor of language skills. On the other hand, in this study we found no negative correlation between language ability and age of first prosthesis or cochlear implant. A possible explanation is that none of our subjects used sign language, and were trained in the use of lip reading, which relies on the oral language.

To conclude, our results suggest that temporal auditory dominance can be important for the development of language skills and that simple semantically unrelated stimuli, such as the bisection task used here, could be used as language screening.

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