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**Cued Speech Proficiency improves Segment and Cluster Accuracy in French-speaking
Children with Cochlear Implants**

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ABSTRACT

Objective: This paper aims to investigate to what extent Cued Speech proficiency may improve speech production in French-speaking children with cochlear implants. Although cochlear implants facilitate speech comprehension in children with profound hearing loss, the auditory information provided remains degraded and phonological processing can remain impaired. In face-to-face interaction, Cued Speech manual gestures can be used to complement auditory and visual speech information, with established benefits for children with hearing impairment. A Cued Speech education might also have longer-term impacts on the speech processing abilities of these children. It can thus be expected that production accuracy will also benefit from Cued Speech exposure.

Design: Thirty-two children with cochlear implants (20 with low Cued Speech proficiency vs 12 with high Cued Speech proficiency) and 81 children with typical hearing aged from 60 to 135 months took part in this study. Speech production was assessed using the picture-naming task from the EULALIES battery, which is designed to test the accuracy of spontaneous (non-imitative) phoneme production in isolated word context.

Results: The results reveal first, that early implantation facilitates the development of phonological skills, but also that adequate exposure to Cued Speech compensates for late implantation. A positive effect of Cued Speech proficiency was indeed observed on consonant, consonant cluster and vowel production in children with cochlear implants.

Conclusion: This study highlights long-term effects of Cued Speech education on speech production. This suggests that providing the child with a richer multimodal input might result in better specified phonological representations.

Keywords: French Cued Speech, Cochlear Implants, Speech Production, Phonological Representations

INTRODUCTION

Phonological representations are information about the sound structure of a language stored in long-term memory (Munson et al., 2012). The quality of these representations plays a key role in accurate speech perception and speech production, as well as in verbal short-term memory and in the development of reading and writing skills (e.g., Fowler, 1991; Campbell, 1992; Elbro, 1996; Leybaert & Charlier, 1996; Stackhouse et al., 1997; Rvachew et al., 2003). In children with typical hearing (TH), perceptual phonological knowledge emerges from the multisensory input provided by the linguistic environment (Beckman & Edwards, 2000). In orally-educated children with profound hearing loss, it is mainly built from lipreading, due to the lack of auditory information (Dodd, 1987; Burden & Campbell, 1994; Leybaert & LaSasso, 2010). However, in non-sense syllables, lipreading only gives access to 30% of phonemes (Bernstein et al., 2000). Therefore, through lipreading, children with profound hearing loss only have access to incomplete information and develop underspecified perceptual phonological knowledge (Charlier & Leybaert, 2000; Leybaert et al., 2011), which may impact oral language development and lead to persistent oral language disorders (Geers et al., 2016; Hansson et al., 2018).

To compensate for the degraded acoustic information in hearing impairment, Cornett (1967) developed Cued Speech (CS), a system initially designed to provide phonological information via manual gestures which are used to disambiguate visually identical phonemes in lip reading (e.g. /b/ and /m/). Providing Cued Speech gestures simultaneously with speech has been shown to improve sentence processing in children with hearing aids (Périer et al., 1990; Leybaert & LaSasso, 2010) and even in children with cochlear implants (Hage & Leybaert, 2005). Some studies have also demonstrated beneficial effects of Cued Speech exposure on the development of phonological representations in children with hearing impairment (e.g. Charlier & Leybaert,

2000; LaSasso et al. 2003) but very few studies have tested the long-term impact of a Cued Speech education on audio-only speech processing, or on speech production skills. Only two studies have evidenced that exposure to Cued Speech before cochlear implantation significantly improves speech intelligibility (Vieu et al., 1998; Kos et al., 2009). To our knowledge, there are no fine-grained study on the impact of Cued Speech exposure on the accuracy of phoneme production.

Cued speech perception

In the Cued Speech manual system (Cornett, 1967), all the consonant phonemes are cued with a specific handshape and all the vowels are cued with a specific hand placement around the face (see Appendix 1 for French Cued Speech). The manual cue helps disambiguating phonemes which have similar lip shapes. For example, consonants /b/ and /m/, which differ in manner of articulation, are visually identical, as they have similarly closed lips. Different Cued Speech handshapes are used to introduce a contrast between these two consonants: /m/ is produced with the addition of a handshape with all four fingers extended and the thumb raised whereas /b/ is accompanied with a handshape with all four fingers extended and the thumb hidden. Cuers will provide a manual cue for each phoneme of each word to convey spoken language to people with hearing impairment (Shull & Crain, 2010). Cued speech can be used with or without phonation (LaSasso, 2010). Although initially developed in English-speaking countries, Cued Speech has now been adapted to more than 60 spoken languages (Shull & Crain, 2010). Each of these spoken languages has its own Cued Speech system, adapted to its own phonological system (i.e., placements and configurations of the hand may therefore vary from one system to another). In 1998, Fleetwood and Metzger have argued that *Cued Language* and *cued speech* should be distinguished, just like language and speaking or language and signing. They suggested that the name *cued speech* refers to the communication modality whereas the name

Cued Language denotes a specific spoken language conveyed with manual and facial cues. However, previous studies commonly used the term Cued Speech to refer to modality and/or language. Given that all participants in this study are exposed to the same language, and to preserve backward-compatibility with previous literature, the term Cued Speech is used in this paper.

There are now well-established data that evidence the effectiveness of Cued Speech for speech perception. Speech comprehension has been shown to be improved in children with hearing impairment when audio-visual speech is presented together with manual cues (e.g. Ling & Clarke (1975) for Canadian English Cued Speech; Uchanski et al. (1994) for American English Cued Speech; Périer et al. (1990) for French Cued Speech), with benefits increasing with duration of exposure to Cued Speech (Clarke & Ling, 1976; Périer et al., 1990). Perception is improved even without auditory information (Nicholls & Ling, 1982). Early exposure to Cued Speech (before the age of two years) has been shown to be a key factor, particularly for pseudo-word perception (Alegria et al., 1999). But, even more remarkably, a few studies have also revealed that a Cued Speech education has longer-term effects, and that it helps children develop phonological skills that will help them better perceive lip gestures or audio-only speech information.

Impact of Cued Speech education on the development of phonological representations

A few studies suggest that a Cued Speech education not only helps speech perception when cues are added, but it also improves lipreading skills when cues are absent. As an example, Aparicio et al. (2012) revealed that adults who had received French Cued Speech in childhood had significantly better lipreading skills than those who had not received French Cued Speech. A few rare studies have even evidenced an effect of Cued Speech education on the perception of speech in audio conditions only. Kos et al. (2009) found that pre-implantation exposure to

French Cued Speech improves post-implantation phoneme identification based on audio only. The authors speculate that pre-implantation Cued Speech use promotes knowledge of the acoustic and temporal structure of the oral language, which would benefit the development of perceptual phonological knowledge, and in turn improve audio speech perception after implantation.

Additionally, Cued Speech exposure has also been shown to have a positive impact on phonological awareness and on linguistic competencies. Children who have been exposed to Cued Speech have better rhyme judgment performance (Charlier & Leybaert, 2000 for French Cued Speech) and rhyme generation skills (Paire-Ficout et al., 2003 and Leybaert et al., 2011 for French Cued Speech; LaSasso et al., 2003 for Cued American English), with performance equivalent to their peers with typical hearing if Cued Speech exposure has been intensive. Paire-Ficout et al. (2003) argue that Cued Speech reinforces the phonological input and helps better specify phonological contrasts, which facilitates access to the mental lexicon, making it easier to generate rhymes.

Overall, these studies indicate that exposure to Cued Speech sustains the development of accurate phonological representations, which leads to better perception, better phonological awareness, and supports reading, spelling and remembering (Colin et al. 2007; Leybaert & Charlier, 1996; Bouton et al., 2011; Trezek, 2017). In addition, it can be expected that perceptually-enriched phonological representations will provide more specified information to the speech production system, which has been shown to lead to more accurate realization of the sound units of the language for typically-developing children (Munson et al., 2012; Schwartz et al. 2012). Moreover, studies have also shown that perceptual training improves speech production in children with speech sound disorders (e.g. Rvachew et al., 2004). Therefore, it can be hypothesized that Cued Speech exposure, through its impact on speech perception and

on phonological representations, might also affect the speech production skills of children with hearing impairment.

Use of Cued Speech in children with cochlear implants

Cochlear implant is now a widely used remedial device available for congenital deafness and which partially restores access to speech sounds. However, although CI improves speech perception in children with profound hearing loss, the perception of some acoustic features can be altered (Bouton et al., 2012; Leybaert et al., 2016) and oral language development may be impacted (Hansson et al., 2018), with phonological skills that remain limited (Leybaert & LaSasso, 2010; Colin et al., 2017; Nittrouer et al., 2018). A few studies have demonstrated the benefit of using Cued Speech as a way to supplement speech perception in children with cochlear implants (Hage & Leybaert, 2005; Colin et al., 2017). As mentioned earlier, to our knowledge, only two studies have focused on the benefits of Cued Speech exposure on speech production. Vieu et al. (1998) and Kos et al. (2009) have shown that people who have been exposed to Cued Speech before cochlear implantation show significantly higher speech intelligibility scores. Moreover, Vieu et al. (1998) observed that children exposed to Cued Speech reached better speech intelligibility ratings than the other groups (i.e. children exposed to sign language and children with oral language only) before and after cochlear implantation. These preliminary observations on the positive impact of Cued Speech exposure on speech production suggest that perceptual phonological knowledge may be transferred to speech production skills. The present study thus investigates to what extent Cued Speech exposure may enhance speech production in children with cochlear implants. To our knowledge, this work is one of the first attempts to explore the impact of Cued Speech exposure on the development of phoneme production skills in children with cochlear implants.

RESEARCH QUESTION

The present study aims to investigate whether and how Cued Speech exposure may improve phoneme production in children with cochlear implants. To that aim, phoneme production in French children with cochlear implants is compared across different levels of Cued Speech proficiency. Indeed, as suggested by Colin et al. (2015; 2017), and more recently by Van Bogaert et al. (2023), the child's ability to decode manual cues (i.e. Cued Speech proficiency) is a key factor when studying the effects of Cued Speech exposure.

This study is focused on two main questions. First, the accuracy of French phoneme production by children with cochlear implants is evaluated and compared with data from children with typical hearing. Among other demographic factors, the effect of age at implantation is particularly examined. Indeed, the positive impact of early implantation has long been documented and several studies show that, by providing access to speech sounds in a crucial period for language development, implantation before 24 months improves the performance of children with cochlear implants (Boons et al., 2012; Colin et al., 2017; Berland et al., 2019; Sundarrajan et al., 2020, among others). Colin et al. (2017), among others, found that early implantation has a significant impact on the development of phonological skills and thus on reading and spelling. The second question concerns the impact of Cued Speech proficiency on the speech production skills of children with cochlear implants. The performance of children with high cue reading skills is contrasted with that of children with low cue reading skills. The final aim of the study is to test whether Cued Speech is a relevant tool to support oral communication development in children with cochlear implants.

MATERIAL AND METHODS

Picture-naming task

This study assessed the accuracy of consonant, consonant cluster and vowel production using a picture-naming task. This task is an adequate tool to assess a child's ability to produce phonological contrasts (Edwards & Beckman, 2008; Mahshie et al., 2015). The EULALIES battery (Meloni et al., 2017) consists of five tasks, each testing different levels of French speech processing, both in perception and production, with reference data on typically-developing children. The EULALIES picture-naming task used in this study tests the accuracy of spontaneous (non-imitative) phoneme production in isolated word context. It was designed to allow a comprehensive analysis of phonological skills, by including all French phonemes in different word positions, as well as a significant number of clusters and words varying in syllable structure and length. It includes 68 familiar words (Appendix 2). All French phonemes are targeted, with consonants elicited in word-initial, word-medial and word-final positions, as well as syllable-initially, syllable-finally, and in clusters. Word length varies from 1 to 4 syllables, presence of clusters is controlled, resulting in varying degrees of word complexity (simple words like “robe” /ʁɔb/ *dress* or complex ones like “hélicoptère” /elikoptɛʁ/ *helicopter*). Only familiar words are used, in order to lower the weight of lexical access, and to focus on the child's phoneme production abilities. For that purpose, high frequency of occurrence in oral French as indicated by the Lexique.org database (New et al., 2004) and accessibility to the youngest were used as criteria to select the stimuli (designating clothing, everyday objects, food, transportation modes, etc.). Children saw pictures displayed on a computer screen and had to spontaneously produce the name of each item. The pictures were clear photographs of familiar items and every attempt was made to elicit words without a model. In rare cases in which a child had difficulty identifying a picture, semantic or phonological cues were provided. When the child was unable to name the item despite the cues, the experimenter simply asked the child to repeat the word after her.

Data collection

Children sat in front of a computer screen on which the pictures to be named were displayed. They wore a SHURE headset microphone (Beta 54R) to record their productions and a backup microphone was also placed on the table in front of them. The experimenter was seated next to the child, also facing the computer screen. The audio data were recorded using a Zoom recorder (H4n Pro). Preliminary tests were administered: a digit span task (ODEDYS; Jacquier-Roux et al., 2002), and the morphosyntax production subtask of the ELO oral language test (Khomsî, 2001). These tests were used as inclusion tests for children with typical hearing: children whose scores were below the norm were excluded from the study.

The five tasks of the EULALIES battery were performed starting with the picture naming task. An auditory screening test was completed with children with typical hearing to eliminate a possible hearing disorder (perception at 20 dB on the frequencies 250, 500, 1000, 2000, 4000 and 8000 Hz, using an Electronica 9910 audiometer). For children with cochlear implants, French Cued Speech reading level was evaluated using the “Test d’évaluation de la réception du message oral par l’enfant sourd” (TERMO, Evaluation test for the reception of the oral message by the deaf child; Busquet & Descourtieux, 2000). TERMO is designed to assess lip-reading and cue reading skills in children with hearing impairment. A list of words and sentences (Appendix 3a) was presented by the experimenter using French Cued Speech gestures without phonation, in order to measure the children’s cue reading skills without the auditory modality. The children were asked to repeat the items vocally. Phonological accuracy of the children’s production was not evaluated, only lexical accuracy. Children were split into two subgroups according to their TERMO scores. Children in the CS- group had low cue reading skills: they could at most decode a few familiar words at slow speed (levels 3 and 4 of the TERMO scale, Appendix 3b). Children in the CS+ group had high cue reading skills: they could decode words and/or sentences at normal speed (levels 1 and 2 of the TERMO scale, Appendix 3b).

Parents completed a survey on their child's language development and background (multilingualism, age of first fitting, exposure to French Cued Speech, etc.). This provided further individual and environmental information and allowed us to eliminate any possible associated disorder. Data were collected anonymously, according to GDPR regulations.

Participants

Children with cochlear implants

Thirty-two children with profound hearing loss wearing cochlear implants and aged 60 to 135 months took part in this study (CI group). They had no additional diagnosed disability at the time of the study. Due to ethical regulations, information on the origin of the deafness was not available. Three quarters of the children were recruited during summer Cued Speech camps organized by the Association for the Promotion of French Cued Speech (ALPC: Association nationale pour la promotion et le développement de la Langue française Parlée Complétée) in 2018, 2019, 2022 and 2023 for families from all over France. The other quarter consisted of children recruited in schools in the Grenoble area in 2019 and 2022. All the children in the CI group had been exposed to French Cued Speech. As concerns exposure to French Sign Language (FSL), one child was regularly exposed to FSL at home (3.1%), 13 children were occasionally exposed to FSL at school or at home (40.6%) and 18 children had never been exposed to FSL (56.3%).

The CI group was divided into two subgroups according to Cued Speech proficiency (i.e. the level of Cued Speech reading skills, as described above). Table 1 describes the main demographic features by subgroups. Twenty children including nine girls had reached a low cue reading level ("CS-" group, age = 93.50 months, SD = 20.12) and twelve children including four girls demonstrated a high cue reading level ("CS+" group, age = 95.08 months, SD =

24.71). Seventeen children in the CS- group were fitted with two cochlear implants (bilateral) and three children were fitted with one cochlear implant and a hearing aid on the other ear (bimodal). In the CS+ group, eight children had a bilateral fitting and four children a bimodal one. The mean age at implantation was 29.15 months ($SD = 22.84$) for the CS- group and 29.67 months ($SD = 23.42$) for the CS+ group. Children were classified as early implanted and late implanted: early implantation refers to a first fitting with cochlear implant before 24 months whereas late implantation corresponds to a first implantation after 24 months. Therefore, some of our analyses contrast four groups of participants with CI: “CS- late” for children with late implantation and low cue reading skills ($n = 8$, age = 93.01 months, $SD = 17.50$), “CS+ late” for children with late implantation and high cue reading skills ($n = 5$, age = 92.83 months, $SD = 26.88$), “CS- early” for children with early implantation and low cue reading skills ($n = 12$, age = 93.85 months, $SD = 20.94$) and, “CS+ early” for children with early implantation and high cue reading skills ($n = 7$, age = 96.35, $SD = 20.97$).

Hearing age was defined as chronological age for the control group (i.e. children with typical hearing (TH)) and as duration of implant use (defined as the time elapsed since the first fitting with a cochlear implant) for children with cochlear implants (i.e. the CI group). The mean duration of implant use was 64.35 months ($SD = 27.52$) for the CS- group and 65.42 months ($SD = 30.41$) for the CS+ group. Strictly speaking, the children with cochlear implants had been first fitted with hearing aids before implantation, so their hearing age was in fact higher. However, in our analyses, hearing age was taken as the duration of implant use, because it was more comparable across children with profound hearing loss.

Finally, speech production in children with cochlear implants may also be influenced by socioeconomic status (SES). However, due to incomplete information on SES, a thorough examination of the influence of this factor is beyond the scope of this study. To reduce the

potential influence of SES, children with cochlear implants were recruited during summer Cued Speech camps that usually involve families with homogeneous SES.

Group	Gender	Chronological age (months)	Age at cochlear implantation (months)	Implantation period	Implantation type	French Cued Speech proficiency	Exposure to French Sign Language at home or at school (FSL)
CS- late	M	65	49	late	bimodal	Low	None
CS- late	M	74	28	late	bilateral	Low	Occasional
CS- late	F	79	24	late	bilateral	Low	Occasional
CS- late	M	94	60	late	bilateral	Low	None
CS- late	M	100	36	late	bilateral	Low	Occasional
CS- late	F	103	74	late	bimodal	Low	None
CS- late	M	111	26	late	bilateral	Low	None
CS- late	F	118	94	late	bilateral	Low	None
CS- early	F	64	15	early	bilateral	Low	Occasional
CS- early	F	72	12	early	bilateral	Low	None
CS- early	M	74	11	early	bilateral	Low	None
CS- early	M	77	14	early	bilateral	Low	None
CS- early	M	84	23	early	bimodal	Low	Occasional
CS- early	M	86	19	early	bilateral	Low	Occasional
CS- early	M	92	19	early	bilateral	Low	Occasional
CS- early	F	99	22	early	bilateral	Low	Occasional
CS- early	F	109	11	early	bilateral	Low	Occasional
CS- early	F	114	12	early	bilateral	Low	Occasional
CS- early	F	120	16	early	bilateral	Low	None
CS- early	M	135	18	early	bilateral	Low	Occasional
CS+ late	M	60	33	late	bimodal	High	Occasional
CS+ late	M	62	38	late	bimodal	High	None
CS+ late	M	107	44	late	bimodal	High	None
CS+ late	M	111	85	late	bilateral	High	None
CS+ late	F	125	37	late	bimodal	High	None
CS+ early	M	66	8	early	bilateral	High	None
CS+ early	F	77	12	early	bilateral	High	Important
CS+ early	M	86	20	early	bilateral	High	Occasional
CS+ early	F	91	22	early	bilateral	High	None
CS+ early	M	110	18	early	bilateral	High	None
CS+ early	F	118	10	early	bilateral	High	None
CS+ early	M	128	11	early	bilateral	High	None

Table 1: Demographics of children with cochlear implants

Children with typical hearing (TH group)

The CI group was compared with a group of 81 children with typical hearing (TH group) aged between 61 and 135 months and belonging to the large cohort of typical children in the EULALIES project (Meloni et al., 2017) (age = 90.17 months, SD = 20.44). This group consisted of 45 girls and 36 boys. None of the children were daily exposed to another language than French. TH children had never been exposed to French Cued Speech nor FSL. All the hearing participants were recruited in schools in Grenoble and Strasbourg (France).

This study was approved by the Grenoble Alpes Research Ethics Committee (CER Grenoble Alpes-Avis-2018-04-03-2-Amendment).

Data processing

The items produced by the children during the picture naming task were first transcribed and processed with the PHON software (Hedlund & Rose, 2016). Ten percent of the data were double-blindly transcribed, with an inter-judge agreement of 82.4%. All items which had transcription differences between judges were reviewed by five transcribers and consensus was obtained. No data were eliminated. Transcription methods and criteria were subsequently adapted for further annotation, with a list of diacritics facilitating narrow transcription. After alignment with the target, the number of errors per word was extracted with PHON (adding numbers of substitutions, elisions and epenthesis).

Statistical analyses

All graphs and statistical analyses were completed using the R software (R Core Team, 2019). The mean number of errors per participant on consonants, consonant clusters and vowels was compared between the groups (i.e. CS+, CS-, TH groups). Following the method described in Van Bogaert et al. (2023) a first step in the analysis consisted in assessing the relationship between the mean number of errors per participant and the chronological age in TH participants. A mixed exponential regression model with asymptote was then computed using the *nls* function because of the nonlinear effect of chronological age on number of errors. To situate the CS- and CS+ participants in relation to their typical hearing peers, a 95% prediction interval for the estimated TH curve was calculated using the *predFit* function from the *investr* package. The proportion of CS- and CS+ participants outside or inside the TH prediction interval was computed (see Table 2). A compliance test (*binom.test* function) was performed to assess

whether the proportion of CS- and CS+ participants outside the TH prediction interval was significantly different from 5% (the expected proportion in the TH group). Finally, for each child in the CS- and CS+ groups, the distance to the estimated number of errors in same age TH participants was calculated. A positive distance indicates a higher number of errors relative to the norm. A negative distance indicates fewer errors than the norm.

Linear Models (*lm* function in package *stats* in R) were then used to compare the distance to the TH norm across CS groups. Multiple comparison tests were run, (using the *glht* function in the *multcomp* package) to compare CS- and CS+ groups with each other and to compare each group to the TH norm (i.e. to distance = 0). Given the limited number of participants with representative variation on gender and exposure to French Sign Language, adding these two demographic factors did not improve model fit. These factors were therefore not further considered. Following the general consensus about the effect of age at implantation (cf. supra), the same statistical analyses were reiterated by splitting up CS participants into four subgroups: CS- late, CS- early, CS+ late and CS+ early. This allowed us to examine the effects of Cued Speech proficiency and implantation period (i.e. early (before 24 months) vs late (after 24 months)).

RESULTS

Figure 1 displays the mean number of errors for children with typical hearing as a function of chronological age, on consonants (1.a), clusters (1.b) and vowels (1.c). The solid line indicates the estimated mean number of errors and the dotted lines represent the 95% prediction interval. The triangle and round shapes represent data for CS children. Table 2 summarizes the number of participants of each group above the upper boundary of the prediction interval and Table 3 provides the mean distance to the estimated TH curve (solid line in Figure 1).

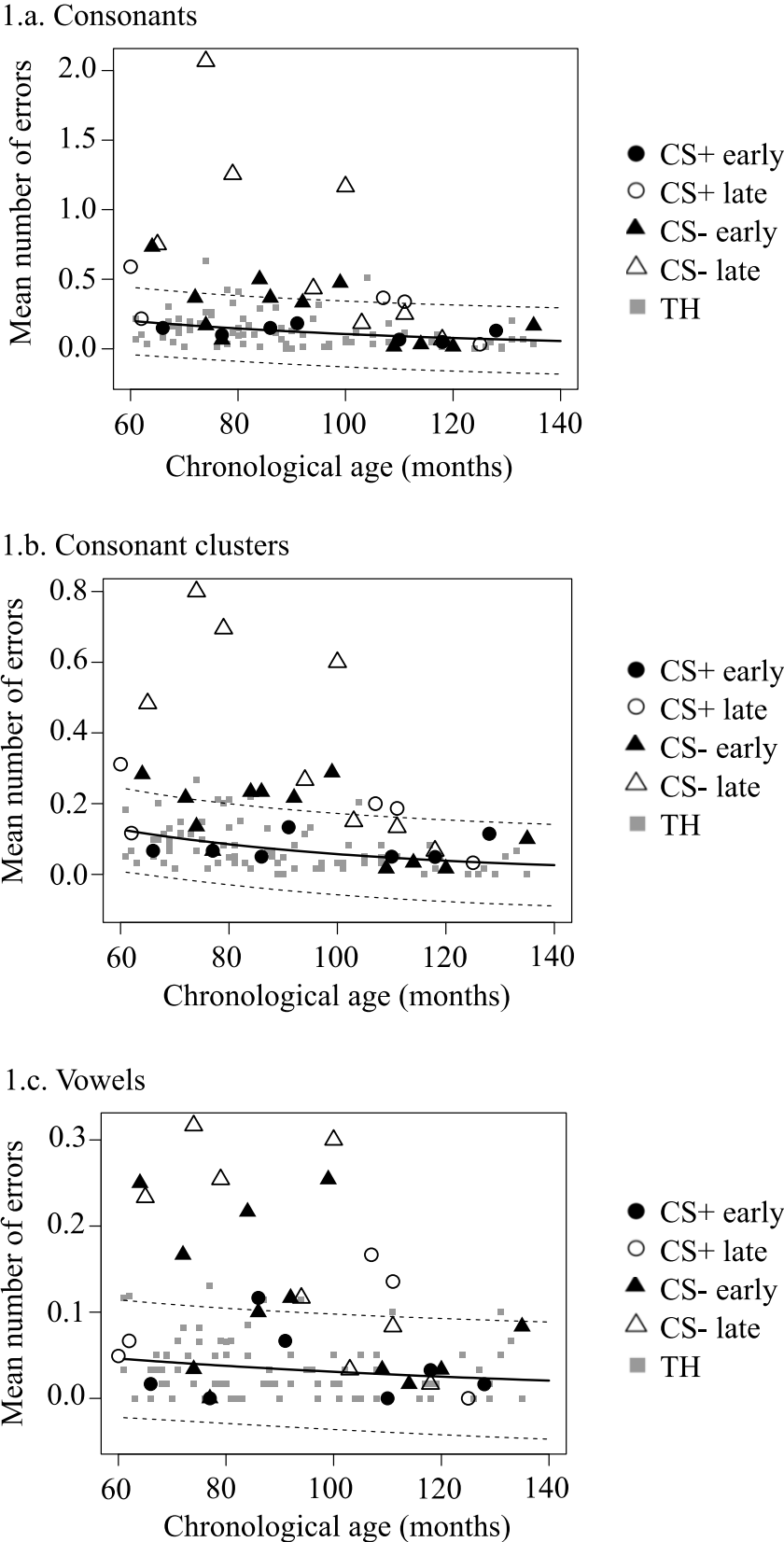


Figure 1: Mean number of errors by chronological age for consonant production in the five groups of children: CS- early (black triangles), CS- late (white triangles), CS+ early (black dots), CS+ late (white dots) and TH (grey squares). The solid line represents the estimated mean number of errors in the TH group and the dotted lines the 95% prediction interval.

CS-			CS+		
PCC	PCIC	PVC	PCC	PCIC	PVC
8/20	11/20	10/20	3/12	3/12	3/12

CS- late			CS- early			CS+ late			CS+ early		
PCC	PCIC	PVC	PCC	PCIC	PVC	PCC	PCIC	PVC	PCC	PCIC	PVC
5/8	5/8	5/8	3/12	6/12	5/12	3/5	3/5	2/5	0/7	0/7	1/7

Table 2: Proportion of participants above the upper boundary of the 95% TH prediction interval (i.e. with a higher number of errors than the norm).

CS-			CS+		
PCC	PCIC	PVC	PCC	PCIC	PVC
0.48	0.23	0.13	0.15	0.09	0.06

CS- late			CS- early			CS+ late			CS+ early		
PCC	PCIC	PVC	PCC	PCIC	PVC	PCC	PCIC	PVC	PCC	PCIC	PVC
0.74	0.33	0.16	0.25	0.14	0.11	0.23	0.16	0.07	0.05	0.04	0.04

Table 3: Mean distance to the estimated TH curve (a positive distance indicates a higher number of errors relative to the norm and a negative distance indicates fewer errors than the norm).

Number of consonant errors

The compliance test for consonant errors shows that the proportion of participants above the TH prediction interval is significantly different from 5% for the CS- group ($p < .001$, distance = 0.48). and for the CS+ group ($p = .020$, distance = 0.15). More precisely, the number of errors of CS- late, CS- early and CS+ late participants are significantly higher than the norm (respectively $p < .001$ with distance = 0.74, $p = .020$ with a distance = 0.25 and $p = .001$ with distance = 0.23). No significant difference is observed between CS+ early and TH participants (distance = 0.05). In other words, there is a high probability for a child with cochlear implants to produce a higher number of consonant errors than a typically-hearing child of the same age, unless he or she benefits from early implantation and a high level of Cued Speech proficiency.

Distance analysis reveals a group effect between CS- and CS+ participants. Multiple comparison tests show a significantly higher distance from the TH group for the CS- participants than for the CS+ participants ($p = .039$). Moreover, only the scores from the CS-

participants are significantly different from those of TH participants ($\rho = .001$). That is, children with a low cue reading level make significantly more consonant errors than children with typical hearing, and than children with a high cue reading level.

As concerns implantation period, multiple comparison tests reveal a significant between-group effect for the age at implantation in CS- group (Figure 2a): the CS- late group displays significantly higher consonant error rates than the CS- early group ($\rho = .010$). That is, later implanted children with a low cue reading level make significantly more consonant errors than children with a low cue reading level and an early implantation. No similar effect is found for children with high decoding skills, which suggests that Cued Speech proficiency may compensate for a late implantation.

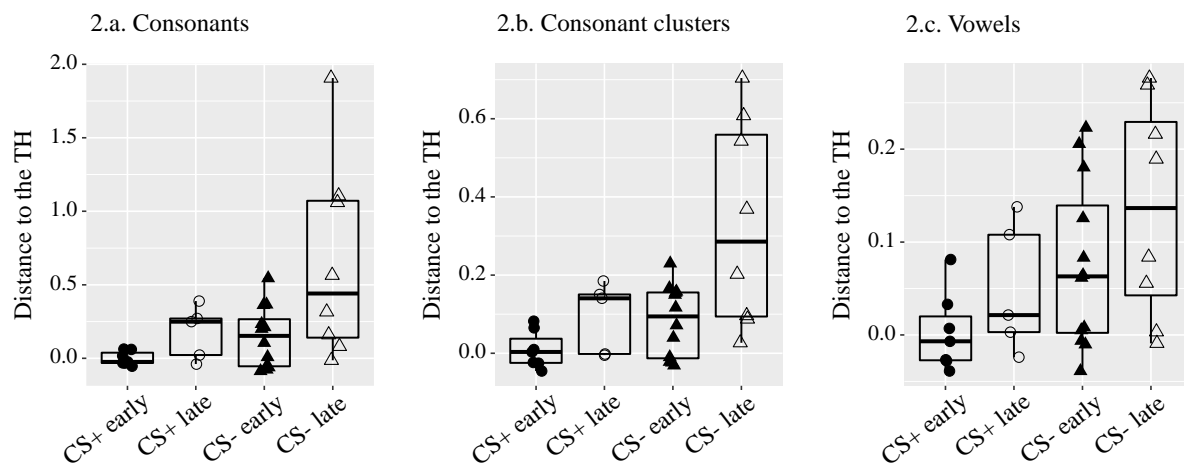


Figure 2: Distance to the estimated TH curve for consonants (left panel), consonant clusters (middle panel) and vowels (right panel) in the four groups of children with cochlear implants (CS+ early, CS+ late, CS- early and CS- late). A positive distance indicates a higher number of errors relative to the norm and a negative distance indicates fewer errors than the norm.

As concerns the benefits of Cued Speech proficiency in early vs late implanted children, a moderate between-group effect is observed (Figure 2a): the CS+ late group displays lower consonant error rates than the CS- late group ($\rho = .056$). That is, with a similarly late implantation, children with high cue reading skills show less inaccuracy in consonant production than children with a low cue reading level. No significant difference was found

between CS- early and CS+ early groups. Overall, only CS- late participants display significantly more errors than the norm ($p < .001$).

Number of consonant cluster errors

As observed for consonant errors, the compliance test for consonant cluster errors shows that the proportion of participants above the TH prediction interval is significantly different from 5% for the CS- group ($p < .001$) and for the CS+ group ($p = .020$). More precisely, the scores of the CS- late, CS- early and CS+ late participants are significantly different from typical scores (respectively $p < .001$, $p < .001$ and $p = .001$). No significant difference is observed between CS+ early participants and TH participants. This suggests that a child with cochlear implants may produce a higher number of consonant cluster errors than a typically-hearing peer, unless he or she benefits from early implantation and a high level of Cued Speech proficiency.

Distance analysis reveals a group effect between CS- and CS+ participants. Multiple comparison tests show a significantly higher distance to the TH norm for the CS- participants than for the CS+ participants ($p = .021$) but only CS- participants are significantly different from the norm ($p < .001$). Children with a low cue reading level make significantly more consonant cluster errors than children with typical hearing, and than children with a high cue reading level. Concerning implantation period, multiple comparison tests reveal a significant between-group effect for the age at implantation in CS- group (Figure 2b): CS- late participants display significantly higher consonant cluster error rates than CS- early participants ($p = .002$). No similar effect is found for children with high Cued Speech proficiency. A significant between-group effect is also observed for cue reading skills in later implanted children (Figure 2b): the CS- late group exhibits significantly higher cluster error rates than the CS+ late group ($p = .018$). With a similarly late implantation, children with high cue reading skills show less inaccuracy in consonant cluster production than children with a low cue reading level. No

significant difference was found between the CS- early group and the CS+ early group. Only the CS- late group is significantly different from the norm ($p < .001$).

Number of vowel errors

As observed for consonant and consonant cluster errors, the compliance test for vowel errors shows that the proportion of participants above the TH prediction interval is significantly different from 5% for CS- ($p < .001$) and CS+ participants ($p = .020$). More precisely, the scores of CS- late, CS- early and CS+ late participants are significantly different from the norm (respectively $p < .001$, $p < .001$ and $p = .023$). No significant difference is observed between CS+ early participants and TH participants. A child with cochlear implants may produce a higher number of vowel errors than a typically-hearing peer, unless he or she benefits from early implantation and a high level of Cued Speech proficiency.

Distance analysis reveals a group effect between CS- and CS+ participants. Multiple comparison tests show a significantly higher distance to the norm for the CS- participants than for the CS+ participants ($p = .012$) but only the scores of the CS- participants significantly differ from the norm ($p < .001$). When focusing on the implantation period, multiple comparison tests reveal no significant between-group effect for the age at implantation nor for cue reading skills (Figure 2c). However, CS- late and CS- early groups are significantly different from TH group (respectively $p < .001$ and $p = .023$) whereas no difference was found between CS+ groups and TH group.

DISCUSSION

The present study aimed to investigate the combined effects of cochlear implantation and of Cued Speech proficiency on speech production in children. To that aim, phoneme production (i.e., consonants, consonant clusters and vowels) was assessed in a group of children with

cochlear implants with different degrees of Cued Speech proficiency, and compared with typically hearing peers. Overall, our data suggest that the picture-naming task from the EULALIES battery can be used for the assessment of speech production in children with cochlear implants. Two main questions were explored. The first question relates to the specificities of speech production in children with cochlear implants as compared to children with typical hearing. The second addresses the impact of Cued Speech proficiency on phoneme production skills of children with cochlear implants.

French phoneme production in children with cochlear implants

In order to determine the impact of hearing impairment remedied with cochlear implantation on speech production skills, the production scores of children with cochlear implants who had little to no Cued Speech proficiency (CS- group) were first compared to the ones of children with typical hearing (TH group). A clear result of our study is that the children in the CS- group show significantly higher error rates than children with typical hearing for consonant production, consonant cluster production, and vowel production (i.e. significantly higher distance to the TH norm). This observation suggests that cochlear implant alone does not allow children with hearing impairment to develop speech production skills akin to those of their peers with typical hearing. It pleads for the view that children with cochlear implants need specific additional support to develop successful oral communication.

However, age at implantation was also found to have an effect on the production scores. Early age at implantation significantly decreases error rates for consonant and consonant cluster production. The CS- early group (children with low French Cued Speech abilities who have been implanted before 24 months of age) consistently show lower error rates than the CS- late group (children with low French Cued Speech abilities who have been implanted after 24 months of age). Early implantation allows CS- children to reach accuracy scores that are higher

than the ones of their later-implanted peers, and that are close to those of children with typical hearing (Fig. 1 and Fig. 2). This finding is consistent with those of Sundarrajan et al. (2020), who demonstrated that early implantation improves consonant production accuracy in children with hearing impairment. In their study, early implantation reduced acquisition delay and led to acquisition stages patterning with those of children with typical hearing. However, in the present study, no similar effect was found for vowel production: both the CS- late and CS- early groups displayed a higher number of errors than the TH group. This suggests that early implantation alone is not sufficient to reach typical vowel production abilities.

Positive impact of Cued Speech proficiency on speech production in children with CIs

The second purpose of this study was to examine the specific impact of Cued Speech proficiency on speech production abilities in children with cochlear implants. To that end, we compared two groups of children with cochlear implants with high vs. low cue reading skills.

The results show that children with cochlear implants and high cue reading skills (CS+ group) display less speech production deficits than their peers with low cue reading skills (CS- group), even though their error rates are higher than that of their peers with typical hearing for consonants, consonant clusters and vowels.

A more detailed examination of our results reveal that Cued Speech proficiency may even compensate for late implantation. No significant difference was found between CS+ groups (i.e. early vs late) for consonant, consonant cluster and vowel production. In addition, CS+ late children show significantly lower error rates to their CS- late peers for consonant and consonant cluster production. The disadvantage of late implantation appears to be cancelled: CS+ late children reach error rates close to those of their CS+ early peers.

These observations highlight the benefits of Cued Speech proficiency on speech production in French-speaking children with cochlear implants. A high cue reading level is associated with a lower number of phoneme production errors, akin to that of peers with typical hearing. Cued Speech provides additional information that can be used by the child to enhance the encoding of phonological information. Children who benefit from Cued Speech exposure may develop fuller phonological specification. In turn, this may translate into better specified phonological plans and articulatory gestures. In other words, phonological representations enriched by multimodal perceptual cues could lead to improved speech production skills.

Limitations

Although this study supports the claim that Cued Speech proficiency improves speech production in children with cochlear implants and that this might stem from a transfer from perceptually-enhanced phonological representations to speech production, other factors may influence speech production skills in children with cochlear implants. The type of implantation (i.e., bilateral vs bimodal, that is, whether the children are wearing two cochlear implants, or one cochlear implant with a contralateral prosthesis) has been shown to have an effect on speech perception (Sturm et al., 2020) and might, therefore, shape speech production as well. In this study, the low number of participants with bimodal implantation (i.e. three children in the CS- group and four children in the CS+ group) does not allow us to compare between types of implantation. Error rates of CS- late children are always significantly higher than those of CS+ late children, although all but two children in the CS- late group wear two cochlear implants whereas only one child in the CS+ late group does. Regarding the group of children with early implantation, for whom no effect of Cued Speech proficiency was found, only one child in the CS- early group had a bimodal fitting. Moreover, Boons et al. (2012) found no difference between bilateral and bimodal fittings when early implantation is considered. Our present

observations therefore seem to warrant the claim that Cued Speech proficiency can compensate for the difference in equipment in late implantation.

Another potentially influential factor that should be examined is duration of French Cued Speech exposure. It has been shown that the longer the exposure, the higher the decoding skills are (Clarke & Ling, 1976; Périer et al., 1990; Leybaert & Charlier, 1996). Moreover, exposure to sign language has been shown to positively impact reading abilities in hearing-impaired children (Niederberger, 2004; Niederberger & Prinz, 2005; Padden & Ramsey, 2012). It may also impact phonological skills and, therefore, have an effect on segmental accuracy.

Finally, socioeconomic status may impact the development of speech production in children with cochlear implants. Even though the potential influence of SES on our data was reduced by recruiting the majority of the participants during summer Cued Speech camps that involve families with homogeneous SES, it would be relevant to further examine the influence of this factor on speech production.

Not all of these factors were considered in our analyses because of the relatively small sample size. Since our groups were too heterogeneous with regard to these variables, it was not relevant to integrate them into our models. Further exhaustive studies, with more data, should take these factors into account in order to more precisely describe the impact of French Cued Speech exposure on speech production in children with cochlear implants.

CONCLUSION

This study is one of the first to evaluate the contribution of Cued Speech proficiency to speech production in children with cochlear implants. The results revealed a more accurate production of consonants, consonant clusters and vowels in twelve children with cochlear implants who had been adequately exposed to French Cued Speech, relative to 20 children with lower cue

reading skills. As found in many studies, the results indicate that early implantation facilitates the development of phonological skills (Colin et al., 2017; Berland et al., 2019; Sundarrajan et al., 2020), but also that adequate exposure to Cued Speech, as measured by a high level of cue reading, compensates for late implantation.

As argued by several research teams (e.g., Leybaert et al., 2011), French Cued Speech is functionally beneficial during face-to-face interaction, as it provides visual access to all the French phonemes. The results of the present study highlight its longer-term effects on speech processing that extend onto speech production. This transfer can be explained by the fact that better perceptual access provides better phonological representations. These data may inform clinical intervention as well as family practice by revealing the crucial role of visual cues for optimal learning of speech production and processing. This preliminary evidence should be extended to larger samples and replicated across ages of participants, controlling for social economic status and language abilities in addition to cue reading skills.

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Appendix 2: List of items for the picture-naming task from the EULALIES battery
(Meloni et al., 2017, 2022)

oreiller	fourchette	camion	livre
locomotive	tigre	bonhomme	ordinateur
hippopotame	hibou	yaourt	crocodile
indien	escargot	aquarium	ciseaux
griffe	médicament	langue	bibliothèque
jambe	uniforme	menton	téléphone
docteur	citron	zèbre	robe
hélicoptère	neige	ventilateur	enveloppe
machine à laver	main	extraterrestre	stade
supermarché	capuche	tomate	toboggan
huile	parapluie	euro	cinéma
pyjama	chaussette	couverture	ongle
peigne	avion	rhinocéros	chocolat
farine	biberon	dentiste	voiture
œuf	grenouille	pieuvre	fraise
poisson	éléphant	ours	loup
déguisement	aspirateur	gare	vétérinaire

Appendix 3a: TERMO test tasks

Word repetition		Sentence repetition	
Before 8 years old	After 8 years old	Before 8 years old	After 8 years old
poire	volcan	Les enfants jouent dans le jardin	Tu as de belles chaussures
feu	refrain	Les poules pondent des oeufs	La statue s'élève sur la place
bateau	tailleur	Non, c'est trop difficile !	Non, c'est trop difficile !
couteau	tissu	La télévision est cassée	Le courrier est arrivé en retard
marché	signal	Le garçon va à l'école	La lampe est suspendue au plafond
champignon	soldat		
oiseau	départ		
malade	boucher		
savon	berger		
ananas	moineau		

Appendix 3b: French Cued Speech proficiency scale

4	3	2	1
Score < 4 in word repetition	Score < 6 in word repetition	Score > 5 in word repetition and < 4 in sentence repetition	Score > 5 in word repetition and > 3 in sentence repetition