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Surgical, speech, and hearing outcomes at five years of age in internationally adopted children and Swedish-born children with cleft lip and/or palate

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ABSTRACT

Internationally adopted children (IAC) with a cleft lip and/or palate (CL/P) tend to arrive with un-operated palates at an age at which their Swedish-born peers have completed their primary palate surgery. Our aim of the present study was to analyze surgical, speech and hearing outcomes of IAC at age 5 and compare with those of a matched group of Swedish-born children. Fifty children with CL/P born in 1994-2005 participated in the study. Twenty-five IAC were matched according to age, sex and cleft type with 25 Swedish-born children. Audio recordings were perceptually analyzed by two experienced, blinded speech-language pathologists. Hearing and speech statuses were evaluated on the same day for all children. Surgical timing and complications as in fistulas and requirement for secondary velopharyngeal (VP) surgery, speech evaluation results, and present hearing status were analyzed for all children of age 5 years. Results showed that primary palatal surgery was delayed by a mean of 21 months in IAC. IAC had a higher prevalence of velopharyngeal impairment that was statistically significant, a higher fistula rate, and experienced more secondary surgery than Swedish-born peers. Hearing loss due to middle ear disease was slightly more common among IAC, whereas the rate of treatment with tympanostomy tubes was similar between the two groups. In conclusion, IAC with CL/P represent a challenge for CL/P teams because of the heterogeneous nature of the patient group and difficulties associated with delayed treatment, and the results show the importance of close follow-up over time.

Introduction

The number of internationally adopted children (IAC) with cleft lip and/or palate (CL/P) has increased since the 1980s in Sweden as in many other countries. Research on IAC with CL/P has been mainly focused on describing and analyzing treatment results in comparison with those in native cohorts. Studies have shown that IAC with CL/P arrive to their new country at a later age and in most cases with un-operated palates. This leads to late primary palatal surgery higher fistula and revision rates [1-5]. A high frequency of velopharyngeal incompetence (VPI) and consequently less favorable speech results have also been described [2-4,6,7]. The effect of the timing of primary palatal surgery on optimal speech outcomes for CL/P patients is still debated. A firstlanguage switch is a further challenge for IAC [8]. A high prevalence of middle ear disease and hearing loss in children with CL/P is well known and complicates the situation further [9]. Studies of adopted children in general have reported additional health problems that are unknown at the time of adoption, such as impaired developmental and nutritional statuses [10-13].

Previous studies on IAC have described a heterogeneous group of patients, which makes comparing data with native cohorts challenging [14]. The inherent fact that medical data from the native countries on surgery, speech, hearing and general medical history are uncertain

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and even missing for IAC makes investigations complex and comparisons difficult. The evaluations regarding surgery are especially challenging, since data regarding timing for pre-adoptive surgery are uncertain or missing and unknown methods for repair are used.

To perform reliable comparative studies of surgical, speech, and hearing outcomes between a heterogeneous group and native-born children with CL/P, it is beneficial if the compared cohorts are matched in terms of age, sex, and CL/P type. To conform to the rigorous standards of scientific investigation, it is also fundamental to compare cohorts using audio or video recordings with standardized tests of articulation and perceptual analyses performed by blinded listeners [14].

Pet et al. [7] presented a retrospective study in 2018 in which speech and surgical outcomes were compared between IAC and non-adopted children. The authors concluded that adoptees were significantly more likely to develop moderate-to-severe VPI and trended toward increased secondary speech surgery. No statistically significant difference was found regarding compensatory misarticulations, compromised intelligibility, and nasal air emission between the groups. The study demonstrated an association but not a causal relationship between delayed repair and inferior outcomes regarding speech and surgery in IAC. Another study was recently published by Morgan et al. [6] on IAC and speech production skills. In this study, IAC demonstrated poorer articulation

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skills than did their non-adopted peers. Interestingly, regression analysis of factors associated with articulation and the presence of cleft-related errors revealed that age at assessment and velopharyngeal status were significantly correlated with articulation outcomes, whereas age at primary palate repair was not. However, in the study by Pet et al. [7], speech evaluation data were only available for little over half of the IAC and native-born children. Moreover, in both studies mentioned, age at assessment differed widely for IAC and the native-born children.

In an attempt to perform a comparative study and meet the scientific criteria previously mentioned, a Swedish study by Larsson et al. was published in 2017 on children with unilateral cleft lip and palate (UCLP) [8]. IAC from China and Swedish-born non-adopted children were matched in terms of age and cleft type and compared at 3 years of age. Significantly fewer correct consonants and a higher prevalence of glottal articulation and VPI were found in the IAC group compared to the non-adopted children. Furthermore, many IAC had to be excluded from the study due to sparse speech material, indicating severe speech difficulties at age 3. Hearing was also taken into consideration and revealed a low prevalence of hearing loss in the IAC group (16.7% vs. 41.2%) when compared to a group of Swedish-born children with the same cleft type.

It is well known that children with CLP have a high risk of otitis media with effusion (OME). Flynn et al. [9] presented a study in 2009 of children with a UCLP, where the prevalence of OME (74.7%) was higher in the UCLP group than in children without clefts (19.4%) at 1-5 years of age. Hearing loss associated with OME was also more prevalent in children with clefts than in children without clefts and OME [9], an unfavorable situation that may lead to delayed speech development. Few studies have compared OME and hearing loss between IAC and native-born children. Swanson et al. showed in 2014 that 25% of IAC with CL/P in the years 1997-2011 had conductive hearing loss and 26% had a history of ear infections, but no group for comparison was described and age at assessment was missing [2]. A study by Werker et al. [15] recently published a study on IAC versus nativeborn children with CL/P, middle ear findings, and hearing status during childhood [16]. No differences were found except for more tympanic membrane perforations in the IAC group.

Most previous studies on IAC lack comparison groups, age at assessment varies greatly, and standardized speech evaluations are often missing. This in a group of children that so far seems to be at great risk of speech difficulties and need for secondary surgeries. Impaired hearing due to OME may cause delayed speech development, and it is beneficial to include hearing in future studies on IAC. This study was therefore designed including matched controls, hearing data and standardized speech evaluations at the same age for all children.

Our aim was to analyze the surgical timing, complications as in requirements for secondary velopharyngeal (VP) surgery, speech evaluation results, and present hearing status for all children at age 5 years.

The following research questions will be addressed:

Is there a difference in timing of primary surgery, fistula frequency and the need for secondary VP surgery between IAC and Swedish-born children with CL/P?

- Is there a difference in frequency of velopharyngeal competence, glottal articulation and retracted oral articulation between IAC and Swedish-born children with CL/P?
- Is there a difference in hearing loss and treatment with tympanostomy tubes between IAC and Swedish-born children with CL/P and a reference group of 5-year-olds without CL/P?

Materials and methods

Evaluation of surgical status (medical records including pictures from each visit) was made with regard to cleft type, type and timing of surgery, the number of surgical interventions, fistula frequency and the need for speech improving surgery as in secondary VP surgery. Examination regarding surgical status, speech assessment, and otological and audiological investigations was performed on the same day for all children according to a standard follow-up protocol at the routine 5-year visit. Data were then analyzed and compared in the two matched groups of IAC and non-adopted Swedish-born children, including a further Swedish-born reference group without cleft in the comparison for hearing.

Participants

Internationally adopted children

Thirty-seven consecutive IAC with CL/P born between 1994 and 2005 treated at Sahlgrenska University Hospital were identified; 28 of 37 families gave written informed consent for study participation. Three children were excluded: Two children could not be matched to a control, and in one case, the audio tape recording was missing. The final study group of IAC with CL/P consisted of 25 adopted children: 11 girls and 14 boys. One of the IAC with an isolated cleft palate (CP) had Pierre Robin sequence. Sixteen IAC originated from China (64%), four from India (16%), four from Eastern Europe (16%), and one from South America (4%). Fifteen had a UCLP, seven a bilateral cleft lip and palate (BCLP), and three an isolated CP.

Swedish-born children

Twenty-five non-adopted Swedish-born children with CL/P were matched according to cleft type, sex and age, as close as possible. Two children in the CP group had Pierre Robin sequence, and one child in the UCLP group had Van der Woude syndrome. All children were treated according to the Gothenburg standardized two-stage surgical protocol at Sahlgrenska University Hospital [17]. For hearing investigation, a further reference group of 5-year-old children without CL/P and with Swedish language background and no known anomalies was included. These children were investigated in 2005 as part of a comparative UCLP study [9]. All groups are shown in Table 1.

Gothenburg two-stage surgical protocol

Three surgeons in the cleft team performed the palate repairs, but the majority of repairs were performed by one surgeon. The general principal of the Gothenburg two-stage surgical protocol is

3 1			
Cleft type	IAC $n = 25$	Swedish-born children $n = 25$	Reference group-hearing $n = 20$
3CLP/UCLP/CP/none	7/15/3/0	7/15/3/0	0/0/0/20
Male/Female	14/11	14/11	9/11
Age at assessment (years)	5 (4:11–5:3)	5 (4:11–5:3)	5 (+/-2 weeks)

IAC: internationally adopted children; Reference group-hearing: children aged 5 years without cleft lip/palate used as a reference for hearing evaluation; BCLP: bilateral cleft lip and palate; UCLP: unilateral cleft lip and palate; CP: isolated cleft palate.

early closure (6 months) of the soft palate to stimulate speech development, followed by hard palate closure at 2–3 years in order to cause as little trauma to the hard palate as possible and hence achieve optimal growth.

The adopted children were included in the Gothenburg twostage palatal protocol [17] and received the required surgical treatment after 2–3 months of adjustment to their new living situation. Hard palate repair would be planned 6 months after soft palate repair and lip-nose repair, but due to circumstances such as difficulties in general health status, a delay could occur. Within each surgical protocol, a variation exists to some extent, and the deviations are explained. In some cases of wide bilateral clefts, the hard palate was, for technical reasons, closed in two procedures. In cases of narrow clefts in IAC, a one-stage procedure was performed.

Lip-nose repair

Lip-nose repair was performed together with soft palate repair (SPR) at 5-6 months.

In selected cases of wide clefts, lip adhesion was performed at the time of SPR.

Final lip-nose repair would normally follow at 12 months but this was not the case in this study population.

Soft palate repair

The soft palate was repaired at 5–6 months together with lip-nose repair, in accordance with the Gothenburg two-stage protocol, leaving a residual cleft in the hard palate.

Hard palate repair

Closure of the residual cleft in the hard palate (HPR) was planned at 24–36 months of age. The so-called tuck-in was performed, where an incision at the cleft border, followed by subperiosteal dissection, raised the palatal mucosa on the cleft side and allowed for a vomer flap to be tucked in and sutured. For wide bilateral residual clefts, the hard palate repair was performed in two stages, and timing was in that case represented by the second and final surgery.

Secondary surgery

Before secondary VP surgery (speech-improving), the patients were evaluated with a comprehensive panel of speech assessments, including audio recording, nasometry, videofluoroscopy, and/or nasoendoscopy. When secondary VP-surgery was needed, soft palate re-repair with intravelar veloplasty was performed. In the majority of children, the investigation regarding speech-improving surgery is usually initiated after 5 years of age and the final decision is taken together with the families. Hard palate fistulas were closed with one or two mucoperiosteal flaps over a sutured nasal layer.

Speech

Speech recording

All participants were audio-recorded at the routine visit to the cleft clinic at age 5 years (4 years 11 months–5 years 3 months) according to a standardized procedure. The audio equipment used was a Panasonic Digital Audio Tape (DAT) SV-3800, Sony Walkman DAT TCD-D8, or Frontier Tascam HD-P2 with either an AKG acoustics C 407/B or Sony ECM-MS957 condenser microphone.

Speech material

For recordings performed before 2005, children repeated short, standard sentences in Swedish with only oral sounds, sentences with combinations of oral and nasal consonants, and sentences with only nasal sounds. From 2005 onward, the standard sentences from the test SVANTE (Swedish Articulation and Nasality Test) were used [18]. If one child in each matched pair was not able to repeat the sentences, the words from an unpublished articulation test (before 2005) and words from SVANTE (from 2005) were used for both children in the pair. In total, 19 pairs repeated the sentences and six pairs named pictures.

Speech assessment

All speech samples were edited in Praat software and only the speech material that was used for assessment was saved and was presented in a random order so that the two speech-language pathologists (SLP) from the same team were blinded to the origin of the children. Both SLPs had substantial experience of working with children with CL/Ps. Before the rating was performed, the SLPs underwent a training session. Six speech samples were rated and discussed by the SLPs during this session. Retracted oral articulation (palatal/velar articulation for dentals) and glottal articulation (glottal plosives for oral consonants) were rated on a five-point scale [19]: 0 = not present, 1 = single occurrence, 2 = several times, 3 = occurs many times, 4 = occurs all the time. The overall perceptual assessment of velopharyngeal function was rated on a three-point scale: 0 = competent velopharyngeal func-1 = marginally incompetent, 2 = incompetent [20,21] tion 'Marginally incompetent' meant signs of minor symptoms of VP function, but not to an extent where surgery was considered. The SLPs were simultaneously sitting in the same room while listening with high-quality headphones to each speech sample through their own computer. After listening to one speech sample, an individual rating was given. Immediately afterward, a consensus assessment was performed. If disagreement occurred, the SLPs discussed the recordings and listened again until a consensus was reached. The consensus agreement was used as the result, and the individual assessments were used for calculating inter-rater reliability. Sixteen speech samples (30%) were duplicated and rerated to calculate intra-rater reliability. Reliability was assessed by point by point agreement. The levels of inter-rater reliability for the two SLTs were 92% for glottal articulation, 78% for retracted oral articulation, and 82% for velopharyngeal function. Both listeners had an intra-rater reliability of 100% for glottal articulation: 87% vs 73% for retracted oral articulation and 67% vs 80% for velopharyngeal function.

Hearing

The otological and audiological investigations were performed at the 5-year visit, i.e. on the same day as the speech and surgery assessment for all children. A control group of 20 children from the general pediatric population without CL/P and without any known disability was assessed in the same manner at age 5 years (±2 weeks). The children were examined by microscopic otoscopy to assess the status of the external ear, ear canal, tympanic membrane, and middle ear. Pneumatic otoscopy was performed when deemed appropriate in the clinical setting. Age and developmentally appropriate psychoacoustic testing and acoustic impedance testing (tympanometry) were performed on the same day. ISO 389, ISO 8253–1, and ISO 8253–2 standards were used. The type of hearing impairment was defined according to hearing levels with a screening level of 20 dB, tympanometry findings, and clinical investigation findings. Hearing loss was defined according to clinical definitions: pure-tone average (0.5-4 kHz) > 20 dB hearing level. The number of children and ears with *in situ* tympanostomy tubes was reported.

Statistical analysis

The differences in speech variables between the adopted children and their matched controls were calculated by the Wilcoxon signed-rank test to compare the ranks between each matched pair for speech variables. Means, standard deviations, percentages, and minimum and maximum values were calculated for each surgical variable. Fisher's exact test was used to calculate differences regarding audiometry screening and hearing status between the three groups. SPSS version 21.0 (IBM Corp., New York, USA) was used for all analyses. A statistically significant difference was considered to be established at p<.05.

Ethical considerations

Approval was obtained from the Regional Ethical Review Board in Gothenburg, 774–10, 2011. All families gave written informed consent for study participation before data collection.

Results

Surgery

Pre-adoptive surgery

Six of 25 adopted children had received complete soft and hard palatal surgery before adoption. All of these children were from the UCLP group (Table 2).

Timing of soft and hard palate surgery

Two IAC were repaired in one stage. Five IAC with BCLPs were repaired in two stages for the hard palate because of wide clefts. Six IAC received pre-adoptive palatal surgery (numbers 1, 6, 11,

 Table 2. Surgical status of IAC at the first visit to the cleft clinic.

 Surgical status

	Un-repaired	Lip-nose	+SPR	+SPR $+$ HPR	
BCLP (7)	5	1	1	0	
ULCP (15)	1	7	1	6	
CP (3)	3	_	0	0	
Total 25	9	8	2	6	

IAC: internationally adopted children; BCLP: bilateral cleft lip and palate; UCLP: unilateral cleft lip and palate; CP: isolated cleft palate; SPR: soft palate repair; HPR: hard palate repair.

16, 23, and 25). Data regarding this surgery were uncertain or missing, and they were excluded from the analysis (and Figures 1 and 2).

The mean age at SPR was 26 months (SD 9.2, range 13–47) for IAC and 5 months (SD 1.0, range 4–8) for Swedish-born children (Figure 1). There was a statistically significant difference in the timing of SPR between the two groups (p<.001).

The mean age at hard palate repair (HPR) was 36 months (SD 9.0, range 14–51) for IAC and 33 months (SD 9.7, range 12–51) for Swedish-born children (Figure 2). No statistically significant difference was found between the groups regarding the timing of HPR (p=.13). All Swedish-born children underwent surgery according to the Gothenburg protocol [19,22,23].

Secondary palatal surgery

Eight secondary palatal surgeries had been performed in seven out of 25 IAC (28%) at five years of age. Four UCLP, where two children had received palatal surgery in China, two BCLP where both children had received palatal surgery in Sweden, and one CP, also repaired in Sweden. Two IAC (Table 2) arrived with preadoptive soft palate repair. Status of the soft palates was evaluated as completely insufficient at time of surgery in Sweden and considered as 'de novo' SPR, and is therefore also present in Figure 1. Three were secondary VP surgery (soft palate re-repairs at 27 months, 30 months, and 58 months), where one child simultaneously received a hard palate fistula repair. One additional child in the IAC group was diagnosed with VPI and recommended to undergo surgery, which the family declined. Three IAC received hard palate fistula repairs, and in two cases, (both BCLP), twice. In the group of Swedish-born children, only one child of 25 (4%) underwent soft palate re-repair due to VPI, at three years and eleven months: the child had UCLP and Van der Woude syndrome.

Speech

Velopharyngeal function

A statistically significant difference was found regarding the velopharyngeal function, where the occurrence of VPI was higher among the adopted children than their matched Swedish-born peers (p=.004) (Figure 3).

Glottal articulation

The frequency of glottal articulation is reported in Figure 4. No statistically significant difference was found between the adopted children and their matched Swedish-born peers (p=.204).



IAC ■CG

Figure 1. Age at soft palate repair for the two matched groups. IAC: internationally adopted children; Swe-born: Swedish-born children. Data regarding timing of preadoptive surgery for IAC are missing and therefore excluded in the figure.



IAC with each matched control



Figure 2. Age at hard palate repair for the two matched groups. IAC: internationally adopted children; Swe-born: Swedish-born children. Data regarding timing of pre-adoptive surgery for IAC are missing and therefore excluded in the figure.





Figure 4. Glottal articulation.

Retracted oral articulation

The frequency of retracted oral articulation is reported in Figure 5. Most of the children received a score of 0. No statistically significant difference was found between the adopted children and their matched Swedish-born peers (p=.235).

Hearing

At the time of the protocol-based investigation, 48% of the IAC and 36% of Swedish-born children had hearing loss in one or both ears due to middle ear disease. There was no statistically significant difference between these groups (p=.57). The hearing loss was mild in all of the affected children. In the reference group of 20 children from the general population without CL/P, the prevalence of mild hearing loss was 15%. Thus, the IAC group had a significantly higher prevalence of mild hearing loss than the reference group (p<.05).

Tympanostomy tubes were present in all three study groups at age 5 years (38% in the IAC group, 42% in the Swedish-born group, and 7.5% in the hearing reference group) (Figure 6).



Discussion

The IAC received palatal surgery at a mean of 21 months later than the non-adopted Swedish-born children and had received more additional surgery by age 5 years. Thirty-two percent of IAC received secondary surgery: four fistula repairs and four soft palate re-repairs. In the Swedish-born group, only one child underwent secondary surgery, including one re-repair of the soft palate and one fistula repair in the same child with Van der Woude syndrome.

The difference in pre-adoption repair between BCLP and UCLP was substantial. All children with BCLP arrived with un-repaired palates, and only one child had undergone lip-nose repair previously. In the UCLP group, all children except one arrived with some kind of repair, seven lip-nose repairs, one soft palate repair, and seven complete palate repairs. This observation also correlates with several earlier studies yielding similar results [2–5]. The reason for the discrepancy is unclear. However, since BCLP is considered more challenging to repair [24], the choice to postpone surgery to a more resourceful cleft center could be a possible explanation.

In this study, there was a statistically significant difference in the timing of SPR between the two groups but not with respect to hard palate repair. The Swedish-born children underwent SPR according to the standardized treatment plan at a mean age of 5 months. A major difference between Swedish-born children and IAC was that the mean age at SPR was 21 months in the latter. The protocol called for hard palate repair at 24–36 months; hence, the IAC generally arrived in time to undergo this procedure. This explains why no significant difference between the groups



■ Per cent of children with PTA> 20 dB. L/R or both ears

Figure 6. Audiometry screening data and hearing status at age 5 years. A statistically significant difference (p=.027) was found between the reference group and IAC group for mild hearing loss (Fisher's exact test). IAC: internationally adopted children; Swe-born: Swedish-born children; PTA (L/R): Pure Tone Average (left/right).

regarding hard palate closure was observed, 36 months for IAC vs 33 months for Swedish-born children.

A statistically significant difference was found between the groups with respect to velopharyngeal incompetence at age 5 years, with a higher frequency among the adopted children (p<.001). We did not find any statistically significant difference between IAC and Swedish-born children with respect to glottal articulation or retracted oral articulation. These results are similar to the study by Larsson et al. [8].

Overall, the two matched CL/P groups had a high prevalence of mild hearing loss. The precise negative impact of hearing loss during the sensitive period of speech development and a first language switch in children with CL/P is unknown. A unique aspect of this study was that hearing was compared between the two matched groups of IAC and Swedish-born children but both groups were also compared to children of the same age without CL/P at the same center. The differences found in hearing loss due to middle ear disease (IAC 48% vs. Swedish-born children 36% vs. reference group-hearing 15%) were interesting considering the results shown in the study of Ma et al. 2016, in which the overall rate of hearing impairment in Chinese children with non-syndromic clefts was considerably lower (17%) than that in most Western studies (24%-50%) [25]. Although their study included older children (6-15 years), the results could indicate that ethnicity and health care setting influence the auditory status of children with CL/P [26]. In this study, the prevalence of tympanostomy tubes was similarly high for both IAC and Swedish-born children (38% vs. 42%). Hence, under-treatment did not seem to be a possible explanation for the differences in mild hearing loss between the groups. Only 3 of 40 ears were treated with tympanostomy tubes in the reference group.

Many studies have described difficulties regarding general health, malnutrition, development, and associated anomalies in IAC [10–13,26,27]. Importantly, these factors could potentially affect treatment outcomes. Swanson et al. [2] observed improvements in all growth percentiles soon after adoption. As this suggests improvements in nutritional status and well-known corresponding impacts on wound healing, the authors raised the question of whether a pre-operative catch-up growth period should be considered for IAC. This was not specifically studied in this material since larger cohorts would be necessary to draw conclusions on this issue. In this study, we encountered one adopted

child with Pierre Robin sequence in the CP group. In the Swedishborn group, we observed two children with Pierre Robin sequence in the CP group and one child with Van der Woude syndrome in the UCLP. We chose to include these children since they did not affect the results. The only Swedish-born child that underwent additional surgery had Van der Woude syndrome.

A lack of accurate pre-adoptive medical information for IAC is a limitation of research in this field and is unfortunately difficult to compensate for [26]. Different surgical techniques and surgeons were used in both groups, which complicates comparisons between the groups further. It is also worth noting that the oro-facial anatomy of the two groups was different due to differences in ethnic background [28-30]. How this might affect the surgical results and thus the function of the palate is not clarified. In this study, 64% of IAC originated from China, a lower number than reported in most previous studies [2,5,31]. This difference could be related to the fact that the children were born in 1995-2005 and therefore arrived before the peak of adoptees with CL/P from China in 2008-2011, which has been reported in many cleft centers [2-5,8,31]. Regarding speech assessment, the SLP were from the same team. Since the assessments were meticulous, inter- and intra-rater reliability was calculated and the SLP were blinded to the origin of the children, we considered this to be of less importance. Studies at later ages are planned and of interest, since the decisions regarding secondary VP surgery not yet were made for all children in the study at this age, and the present speech data indicate possible future difficulties.

In conclusion, this study showed that IAC presented with a significantly higher prevalence of velopharyngeal incompetence, required more surgery, had more palatal fistulas and need for secondary VP surgery than their matched Swedish-born peers at age 5 years. Furthermore, the IAC had a slightly higher prevalence of hearing loss compared to their matched peer, but also a significantly higher prevalence of hearing loss compared to children without clefts at this age. It is therefore of particular importance to closely follow this group of children over time.

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Disclosure statement

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

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