Received: 24 May 2022 | Accepted: 23 February 2023

DOI: 10.1111/1460-6984.12869

International Journal of Language & Communication Disorders

RESEARCH REPORT

Longitudinal data on speech outcomes in internationally adopted children compared with non-adopted children with cleft lip and palate

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Funding information

This research was supported by funds administered by Uppsala University Hospital.

Abstract

Background: At the beginning of the 21st century, international adoptions of children with cleft lip and/or palate increased dramatically in Sweden. Many children arrived partially or totally unoperated, despite being at an age when palatoplasty has usually been performed. To date, the speech development of internationally adopted (IA) children has been described up to age 7–8 years, but later development remains unstudied.

Aims: To investigate speech development between ages 5 and 10 years in children born with cleft lip and palate (CLP) adopted from China and to compare them with non-adopted (NA) children with CLP. A secondary aim was to compare the frequencies of secondary palatal surgery and number of visits to a speech and language pathologist (SLP) between the groups.

Methods & Procedures: In a longitudinal study, 23 IA children from China were included and matched with 23 NA children born in Sweden. Experienced SLPs blindly reassessed audio recordings from routine follow-ups at ages 5 and 10 years. Velopharyngeal function (VPF) was assessed with the composite score for velopharyngeal competence (VPC-Sum) for single words and rated on a three-point scale (VPC-Rate) in sentence repetition. Target sounds in words and sentences were phonetically transcribed. Per cent correct consonants (PCC) were calculated at word and sentence levels. For in-depth analyses, articulation errors were divided into cleft speech characteristics (CSCs), developmental speech characteristics (DSCs) and s-errors. Information on secondary palatal surgery and number of visits to an SLP was collected.

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Outcomes & Results: VPF differed significantly between the groups at both ages when assessed with VPC-Sum, but not with VPC-Rate. Regardless of the method for assessing VPF, a similar proportion in both groups had incompetent VPF but fewer IA than NA children had competent VPF at both ages. IA children had lower PCC at both ages at both word and sentence levels. More IA children had CSCs, DSCs and s-errors at age 5 years, and CSCs and s-errors at age 10. The development of PCC was significant in both groups between ages 5 and 10 years. The proportion of children receiving secondary palatal surgery did not differ significantly between the groups, nor did number of SLP visits.

Conclusions & Implications: CSCs were more persistent in IA children than in NA children at age 10 years. Interventions should target both cleft and DSCs, be comprehensive and continue past the pre-school years.

KEYWORDS

articulation, international adoption, longitudinal, velopharyngeal function

WHAT THIS PAPER ADDS

What is already known on this subject

- At the beginning of the 21st century, IA children with cleft lip and/or palate arrived in Sweden partially or totally unoperated, despite being at an age when palatoplasty has usually been performed. Studies up to age 7–8 years show that adopted children, compared with NA peers, have poorer articulation skills, demonstrate both cleft-related and developmental articulation errors, and are more likely to have velopharyngeal incompetence. Several studies also report that adopted children more often require secondary palatal surgery due to fistulas, dehiscence or velopharyngeal incompetence compared with NA peers.

What this paper adds to existing knowledge

- This longitudinal study provides additional knowledge based on longer follow-ups than previous studies. It shows that the proportion of children assessed to have incompetent VPF was similar among IA and NA children. It was no significant difference between the groups regarding the proportion that received secondary palatal surgery. However, fewer IA children were assessed to have a competent VPF. Developmental articulation errors have ceased in most IA and all NA children at age 10 years, but significantly more adopted children than NA children still have cleft-related articulation errors.

What are the potential or actual clinical implications of this work?

- Speech and language therapy should target both cleft-related and developmental articulation errors. When needed, treatment must be initiated early, comprehensive, and continued past the pre-school years, not least for adopted children.

INTRODUCTION

At the beginning of the 21st century, the number of internationally adopted (IA) children with special needs, including those born with cleft lip and/or palate (CLP), increased dramatically in, for example, Sweden and the United States, with the vast majority originating from China (Goldstein et al., 2014; Hansson et al., 2012; Kaye et al., 2019; Shay et al., 2016; Sullivan et al., 2014; Swanson et al., 2014). The incidence of CLP in Sweden is 2/1000 live births (Hagberg et al., 1998), which means approximately 150–200 children per year. In 2007, 24 children with CLP were adopted to Sweden. In 2009 the number had increased to 98 children, representing 14% of the total number of adopted children 2009 (Stålhand, 2010). Mean age at arrival between 20 and 30.5 months has been reported (Goldstein et al., 2014; Kaye et al., 2019; Larsson et al., 2017; Swanson et al., 2014). The cleft teams met new challenges as many of the IA children arrived partially or totally unoperated despite being at an age when non-adopted (NA) children would have had their palatoplasty done. Moreover, IA children undergo a language shift and are just settling in with their new families at the time of their first palatoplasty, usually performed within 3 months after arrival (Hansson et al., 2012; Larsson et al., 2020; Shay et al., 2016).

Velopharyngeal insufficiency (VPI) has been associated with increasing age at primary palatoplasty in IA children (Follmar et al., 2015; Pet et al., 2018; Sullivan et al., 2014). It has also been shown in several studies that more IA than NA children need secondary surgery due to fistulas, dehiscence, or VPI (Hansson et al., 2012; Morgan et al., 2018; Swanson et al., 2014), but not always at a significant level (Pet et al., 2018). In addition, IA children with CLP have been described as having poorer language skills (Kaye et al., 2019; Scherer et al., 2018) and articulation (Larsson et al., 2017, 2020; Morgan et al., 2018) compared with NA children with CLP.

Larsson et al. (2017) found that 3-year-old IA children (n = 14) with unilateral cleft lip and palate (UCLP) had significantly more articulation errors than their NA peers (n = 18). Furthermore, more IA children than NA children were judged to have an incompetent velopharyngeal function (VPF). A comparison between IA and NA children at age 5 years showed that IA children more often than NA children exhibited articulation errors related to the cleft as well as errors related to development, and 52% of IA children had incompetent VPF compared with 25% of NA children (Larsson et al., 2020). Similar results were shown when a comparison between IA and NA children with cleft palate, with or without a cleft lip, was made in three age groups (3–4, 5–6 and 7–8 years) (Morgan et al., 2018). In all age groups, IA children had poorer articulation skills than NA children. At ages 7–8 years, cleft-related articulation errors were noted in just over half of the IA group compared with just under one-third of the NA US-born children. A larger proportion of the IA children than their NA peers also exhibited developmental phonological errors at ages 7–8 years. Morgan et al. (2018) did not find a significant correlation between age at adoption and articulation outcomes. However, time with sufficient VPF was associated with better articulation skills in both groups. Typically developed NA children without CLP in Sweden score a mean of 95.8% (SD = 4.8, range = 76–100%) correct consonants at word level as 5-year-olds, 97.8% (SD = 3.3, range = 88–100%) as 7-year-olds, and 98.8% (SD = 3.2, range = 86–100%) as 10-year-olds (Lohmander et al., 2017b). A longitudinal study of children with UCLP adopted from China (Larsson et al., 2022) reported a significant improvement between 3 and 7–8 years of age in both per cent consonants correct (PCC) and VPF. Despite the significant improvement reported, 94% of the children had PCC scores > 2 SD below typically developed NA children without CLP at ages 7–8 years. Moreover, developmental speech characteristics (DSCs), to some extent, were still present at ages 7–8 years in 77% of the children.

To date, articulation and VPF have been described and compared between IA and NA children up to age 7–8 years, but what happens after that is still unknown. There is also a lack of knowledge about how articulation and VPF develop over time, as only a few longitudinal studies, including IA children, have been published. The results from previous studies are consistent: IA children have poorer articulation skills and more often have VPI than their NA peers. More studies at later ages are required to understand the full extent of the consequences of the delayed speech development previously described at earlier ages. This study aimed to investigate how VPF and articulation skills develop in a longitudinal perspective between ages 5 and 10 years in children with CLP adopted from China compared with children with CLP born in Sweden. A secondary aim was to compare VPF, articulation, the proportions of secondary palatal surgery, and the number of visits to a speech and language pathologist (SLP) between IA and NA children.

The following research questions were posed:

How do VPF and articulation develop over time?

Are there differences between IA and NA children regarding (1) VPF, (2) articulation, (3) need for secondary palatal surgery or (4) number of visits to an SLP?

METHODS

The Regional Ethics Committee in Uppsala approved this study (Reference no.: 2017/457).

Participants

Information about the participants regarding cleft type, age at adoption, surgery, tympanostomy tubes and number of visits to an SLP was retrieved from their medical records. IA children born in the period 2000–09 and treated for CLP at Uppsala University Hospital were reviewed. Only children originating from China were included, as IA children from other countries were rare during the period of increasing international adoptions to Sweden. This resulted in a larger group of IA children with similar ethnicity, geographical and linguistic origin, and thus rendered a more homogenous group. The other inclusion criteria were unilateral or bilateral CLP. A consecutive series of 39 patients, 25 with UCLP and 15 with bilateral cleft lip and palate (BCLP), met the inclusion criteria. A total of 16 children were excluded due to missing or incomplete audio recordings. Thus, 23 children were included, 17 with UCLP and six with BCLP, six girls and 17 boys. The mean age at adoption was 1;9 (years;months) (SD = 7 months, range = 0;11–3;4). The IA children were matched with Swedish-born children (NA) based on cleft type and sex. If an adopted child had surgery in Sweden, he/she was also matched on the surgical technique for closure of the soft palate, as the surgical methods used changed during the study period. The NA group was selected from a consecutive series of 105 NA children with UCLP (n = 76) or BCLP (n = 29) born in the period 2000–10 and treated at Uppsala University Hospital. A random selection was made to prevent bias if more than one Swedish-born child with a complete audio recording matched an adopted child. One included NA child had Van der Woude syndrome.

A total of 12 IA children (52%) were treated with tympanostomy tubes at a mean age of 2;7 (range = 1;1–3;9) and a mean of 8 months after adoption (range = 2–15 months). A total of 16 NA children (70%) were treated with tympanostomy tubes at a mean age of 1;0 (range = 3–27 months).

Surgical treatment

The standard procedure at Uppsala University Hospital since 1984 has been to close the palate in two stages: soft palate closure (SPC) at age 6–8 months and hard palate closure (HPC) at age 2 years. Before 2008, SPC was performed with an intra-velar veloplasty reinforced by the palatopharyngeal muscle (Henriksson et al., 2005). Since 2008, intra-velar veloplasty has been performed (Sommerlad, 2003, 2006). Speech-improving surgery was performed with a pharyngeal flap. Before such surgery, the patients were evaluated with a speech assessment, nasoendoscopy and, when possible, nasometry. The order of operations was the same in both groups, but the timing differed. A total of 15 (65%) in the IA group and 11 (48%) in the NA group had their secondary alveolar bone grafting before the assessment at age 10 years. See Table 1 for surgical details.

Speech material

Audio recordings from routine follow-ups at ages 5 and 10 years were used for phonetic transcription, rating of hypernasality and perceived VPF. The audio recordings were made with Zoom H4n or a PC with Soundswell software (Saven Hitech, Stockholm, Sweden) and a condenser microphone (Røde NT4, Sydney, Australia or Philips SpeechMIke Classic 6264). Mean ages at recording were 5;2 and 10;1 in the IA group (ranges = 4;11–5;8 and 9;8–10;7) and 5;1 and 10;1 in the NA group (ranges = 4;11–5;5 and 9;7–10;5). The speech material consisted of 59 words and 13 sentences from the Swedish Articulatory and Nasality Test (SVANTE) (Lohmander et al., 2017b). The 59 words and the first eight sentences each include target consonants particularly vulnerable to cleft-related speech difficulties (plosives /p/, /b/, /t/, /d/, /k/ and /g/, and voiceless fricatives /s/, and /f/). The first nine words also include high vowels, (/i:/, /u:/ and /u:/). Three sets of speech material were edited from the recordings: (1) 59 single words, each including one target consonant; (2) a nine-word string (the first nine monosyllabic words edited to a string of words with no pause between them) including high vowels for assessment of hypernasality; and (3) 12 sentences used for phonetic transcription of target consonants and rating of VPF—the 13th consisted only of nasals and was therefore excluded. The first eight sentences, including four target consonants each, were phonetically transcribed. All 12 were included in the rating of VPF. Picture naming or reading from a list was used to elicit the words, and sentences were repeated after the SLP. The audio recordings (.wav files) were edited using Praat (Boersma & Weenink, 2018) to contain only the speech from the child. The audio files were then randomized and assigned codes to ensure blinded assessments. The audio files were listened to through high-quality headphones and could be replayed as many times as needed.

TABLE 1 Primary surgeries

	Internationally adopted	Non-adopted
Surgery before adoption, *n* (%)	12 (52%) lip 7 (30%) lip + palate	–
Age at SPC, mean (SD, range)	2;0 (7, 1;1–3;9)	0;7 (1, 0;6–0;9)
Age at HPC, mean (SD, range)	2;6 (7, 2;0–4;3)	2;1 (2, 1;10–2;8)
Secondary alveolar bone grafting, mean (SD, range)	9;8 (9, 8;3–10;11)	10;0 (10, 8;10–11;8)

Notes: Mean ages and ranges are given as years;months, SD is given in months.
HPC, hard palate closure; SPC, soft palate closure.

Speech assessment

Calibration. Before rating hypernasality and a perceptual rating of velopharyngeal competence (VPC-Rate) and the phonetic transcription of target sounds in words and sentences, two training sessions were performed to calibrate the SLPs. Four SLPs with 6–18 years of experience with patients with cleft palate were involved in the assessments, three in the assessment of hypernasality and VPC-Rate, and two in the phonetic transcriptions. SLPs 1 and 2 were from the same centre the participants were recruited from, and SLPs 3 and 4 were from two other centres in Sweden. SLPs 1–3 spent 1.5 h calibrating their ratings of hypernasality and VPC-Rate. They were instructed first to decide if hypernasality and VPF were within normal limits and, if not, rate the degree of hypernasality as mild, moderate or severe, and VPF marginally incompetent or incompetent. They listened to one file at a time, rated them individually and then compared their ratings. SLPs 1 and 4 spent 4 h listening to and discussing their phonetic transcriptions. They listened to five words or one sentence at a time and then compared their transcriptions. In cases of disagreement in the transcriptions or ratings, the SLPs discussed until reaching consensus.

VPF. Two variables to assess VPF was used: VPC-Sum and VPC-Rate (Lohmander et al., 2017a). VPC-Sum was used to assess perceived VPF in words, and VPC-Rate was used to assess perceived VPF in connected speech. VPC-Sum is a composite score encompassing (1) perceptual ratings of hypernasality rated on a four-point scale (normal, mild, moderate, severe); (2) perceptual signs of VPI from transcriptions (nasal emission and weak pressure consonants); and (3) active non-oral consonant errors from transcriptions. The three variables each generate a score of 0–2. Ratings of hypernasality translate to scores as follows: 0 = 0, 1 = 1, and 2–3 = 2. The number of perceptual signs of VPI and active non-oral errors in transcriptions translates to a score as follows: 0–2 occurrences = 0, 3–5 occurrences = 1, and ≥ 6 occurrences = 2. The three scores were added together to calculate VPC-Sum of between 0 and 6 for each child. A score of 0–1 indicates competent VPF, 2–3 indicates marginally incompetent VPF, 4–6 indicates incompetent VPF, and VPC-Sum is reported as competent, marginally incompetent and incompetent VPF.

VPC-Rate was rated on a three-point scale (competent, marginally incompetent and incompetent). The SLPs rated hypernasality for all children and then VPF, first at age 5 years and then at age 10 years. If a child consistently used glottal articulation, VPF was rated as incompetent. The score for each child was classified based on the majority decision of the three SLP ratings. In case of total disagreement, the middle value was chosen. All audio files, including 30% for intra-rater agreement, were rated by SLPs 1–3.

Phonetic transcription. SLPs 1 and 4 performed a semi-narrow transcription, in this study defined as a phonetic transcription using the International Phonetic Alphabet (IPA) (2015) and the symbols for nasal escape, velopharyngeal friction, weak articulation, voicing, devoicing and active nasal fricative from the extended IPA (2008) symbols for disordered speech. The target sounds in the 59 words and eight sentences were transcribed for all participants by SLP one. A total of 30% of the audio files in each group at each age were randomly selected for intra-rater agreement. For interrater agreement, 50% of the sound files from ages 5 and 10 years were randomly selected and transcribed by SLP 4.

The transcriptions were used to calculate PCC (Shriberg & Kwiatkowski, 1982)[1], where all articulation errors were given the same weight. A consonant was scored correct if the place and manner of articulation were correct. Signs of VPI were disregarded. PCC was calculated at word and sentence levels. Articulation errors were divided into cleft speech characteristics (CSCs) and DSCs, as described in the Scandcleft project (Willadsen et al., 2017). Three or more occurrences signified an error. CSCs were subdivided into non-oral errors (glottal or pharyngeal plosive or fricative, nasal for unvoiced stop or fricative, and active nasal fricative) and oral errors (retracted/backed to palatal/velar/uvular place of articulation and double articulation). Velar fronting (a velar consonant is replaced with a dental or alveolo-dental consonant), stopping (a fricative is replaced with a plosive), and voicing errors (difficulties differentiating between voiced and voiceless consonants) were considered DSCs. An articulation error

TABLE 2 Intra-rater agreement for hypernasality and rating of velopharyngeal competence at ages 5 and 10 years

		Hypernasality		VPC-Rate	
		5 years	**10 years**	**5 years**	**10 years**
Intra-rater	Rater 1	50%	93%	93%	93%
	Rater 2	79%	64%	93%	93%
	Rater 3	71%	93%	93%	86%

Note: VPC-Rate, rating of velopharyngeal competence.

could be counted as both a CSC and a DSC. The consonant /s/ was scored as correct when realized as /s/. All other oral realizations of /s/ that were still realized as a fricative (*θ* ȿ ç ɬ x) were scored as incorrect and categorized as an s-error. Although they may be categorized as CSCs or DSC, s-errors were reported separately.

TABLE 3 Differences between phonetic transcriptions considered an agreement

/ʔ b͡ʔ/ (glottal versus glottal reinforcement)
/ʔ ∅[a] /
/c k/ /ɟ g/

[a]*Notes*: Omission of target consonant.

Statistical analysis

Non-parametric analyses were used for group comparisons. The chi-square test was used to compare VPC-Sum and VPC-Rate between IA and NA children. Mann–Whitney *U*-tests were used to calculate differences in PCC between the groups. The categorical data related to CSCs and DSCs (three or fewer occurrences), the need for speech-improving surgery and number of visits to an SLP were calculated with the Fisher exact test. Statistical analyses were performed in IBM SPSS Statistics, version 28. The level of significance was set at $\alpha < 0.05$.

Agreement

Interrater agreement for hypernasality and VPC-Rate was calculated as the frequency of (1) agreement between all three raters, (2) agreement between two raters and (3) no agreement. Regarding hypernasality at age 5 years, two out of three raters agreed in 97% of the cases (all raters agreed in 38%), with no agreement in 3% of the cases. Two out of three raters agreed in 98% of the cases at age 10 years (all raters agreed in 48%), with no agreement in 2% of the cases. Regarding VPC-Rate, two out of three raters agreed in all cases at both ages (all raters agreed in 65% at age 5 years and 77% at age 10 years). Intra-rater agreement was measured as agreement point by point for both hypernasality and VPC-Rate. Information about intra-rater agreements is shown in Table 2.

Point-by-point comparisons also calculated the inter- and intra-transcriber agreement for the phonetic transcriptions. The agreement was calculated at word and sentence levels at each age and reported as median values. Comparisons were made for correctly articulated target sounds, symptoms of VPI, and the presence of non-oral articulation. In calculating inter- and intra-transcriber agreement, minor differences were considered an agreement (Table 3). Table 4 provides detailed information about inter- and intra-transcriber agreements.

RESULTS

VPF

VPC-Sum. There was a significant difference between the groups at age 5 years ($p = 0.031$). At age 10 years, the difference between the groups remained significant ($p = 0.003$). Excluding children with pharyngeal flaps from the statistical analyses at the respective ages did not change the results.

VPC-Rate. In contrast to the results for VPC-Sum, VPC-Rate did not differ significantly between IA and NA children at age 5 years ($p = 0.492$) or age 10 years ($p = 0.264$). The results did not change when children who received pharyngeal flaps before the assessments were excluded from the statistical analyses. The proportion of children judged with incompetent VPF decreased in both groups regardless of the method used to evaluate VPF. The results for VPC-Sum and VPC-Rate are shown in Figure 1. Changes in the assessments of VPF between ages 5 and 10 years are shown in Tables 5–8

PCC

The PCC scores at word level at age 5 years did not differ significantly between IA and NA children ($U = 180.00$, $z = -1.86$, $p = 0.063$). After excluding one outlier with significantly lower PCC (NA), a recalculation changed the result to a significant difference ($U = 158.00$, $z = -2.16$, $p =$

TABLE 4 Inter- and intra-transcriber agreement at word and sentence levels at ages 5 and 10 years: median percentages (minimum–maximum)

		Inter-transcriber agreement		Intra-transcriber agreement	
Age (years)	**Agreement on**	**Word level**	**Sentence level**	**Word level**	**Sentence level**
5	Correct/incorrect articulation	88% (69–98)	89% (71–100)	95% (84–100)	91% (83–100)
	Symptoms of VPI	85% (49–97)	77% (47–100)	95% (81–100)	87% (74–100)
	Presence of non-oral articulation	96% (76–100)	95% (74–100)	98% (82–100)	99% (83–100)
10	Correct/incorrect articulation	97% (73–100)	97% (74–100)	98% (85–100)	100% (89–100)
	Symptoms of VPI	95% (56–100)	97% (54–100)	92% (74–100)	94% (74–100)
	Presence of non-oral articulation	100% (90–100)	100% (100)	100% (95–100)	100% (97–100)

Note VPI, velopharyngeal insufficiency.

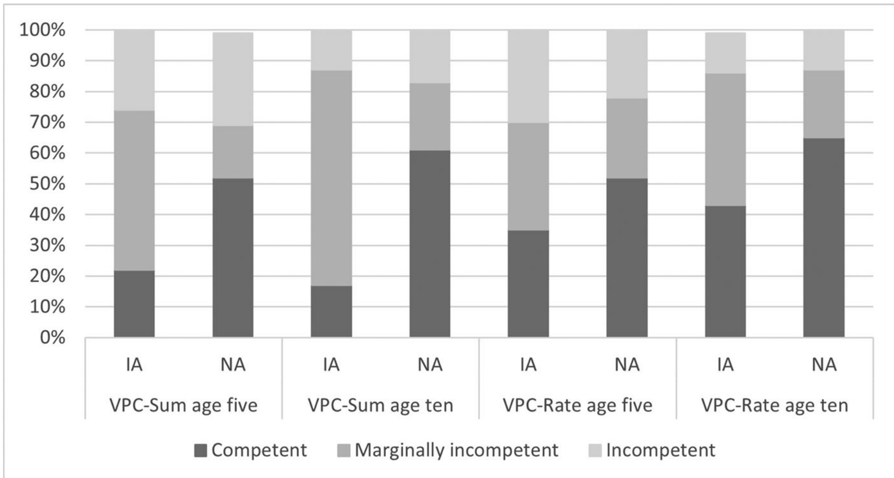

FIGURE 1 Velopharyngeal function at ages 5 and 10 years for internationally adopted (IA) and non-adopted (NA) children.

0.031). At sentence level, the difference was significant ($U = 156.50$, $z = -2.38$, $p = 0.017$).

At age 10 years, there was a significant difference regarding PCC at word level ($U = 164.50$, $z = -2.27$, $p = 0.023$) between IA children and NA children, but not at sentence level ($U = 212.50$, $z = -1.25$, $p = 0.212$). A recalculation after excluding outliers with significantly lower PCC, one in each group for PCC at word level and two in each group for PCC at sentence level did not change the results (Figure 2).

The development of PCC at word and sentence levels between ages 5 and 10 years was significant in both groups (IA: $p < 0.000$ and < 0.000, respectively. NA: $p = 0.001$ and 0.003, respectively). If scores within 1 SD (91%) from the age-specific norms are regarded as age-appropriate, 17% of IA children reached this level at age 5 years. The corresponding proportion among NA children was 35%. A total of 57% of IA children and 39% of NA children scored > 4 SDs below the mean score in typically developed children without CLP. At age 10 years, 30% of IA children and 65% of NA children performed at an age-appropriate level if −1 SD is accepted (95.5%), but 30% in the IA group and 9% in the NA group performed < 4 SDs below the age-specific norm.

Consonant errors

There was no significant difference between IA and NA children regarding CSCs at age 5 years ($p = 0.065$). Oral retracted articulation and non-oral articulation were more common in the IA group (43% and 52%, respectively) than in the NA group (26% and 30%, respectively), but not at

TABLE 5 Changes in the assessment of velopharyngeal function in internationally adopted children between 5 and 10 years of age using sum, score for velopharyngeal competence

		VPC-Sum at age 10			
		0	**1**	**2**	**Total**
VPC-Sum at age 5	0	1	4	0	5
	1	3	8	1	12
	2	0	4	2	6
Total		4	16	3	23

Notes 0 = competent VPF, 1 = marginally incompetent VPF, 2 = incompetent VPF.
VPC-Sum, score for velopharyngeal competence; VPF, velopharyngeal function.

TABLE 6 Changes in the assessment of velopharyngeal function in internationally adopted children between 5 and 10 years of age using rating of velopharyngeal competence

		VPC-Rate at age 10			
		0	**1**	**2**	**Total**
VPC-Rate at age 5	0	5	3	0	8
	1	3	4	1	8
	2	2	3	2	7
Total		10	10	3	23

Notes 0 = competent VPF, 1 = marginally incompetent VPF, 2 = incompetent VPF.
VPC-Rate, rating of velopharyngeal competence; VPF, velopharyngeal function.

TABLE 7 Changes in the assessment of velopharyngeal function in non-adopted adopted children between 5 and 10 years of age using Sum, score for velopharyngeal competence

		VPC-Sum at age 10			
		0	**1**	**2**	**Total**
VPC-Sum at age 5	0	8	4	0	12
	1	2	0	2	4
	2	4	1	2	7
Total		14	5	4	23

Notes 0 = competent VPF, 1 = marginally incompetent VPF, 2 = incompetent VPF.
VPC-Sum, score for velopharyngeal competence; VPF, velopharyngeal function.

a significant level ($p = 0.353$ and 0.231, respectively). An in-depth analysis of non-oral error types showed that substitution or reinforcement with glottal plosive was more common in IA children (52%) than in NA children (17%), and this difference was significant ($p = 0.029$). Active nasal fricative was less common in the IA group (4%) than in the NA group, but the difference was not significant (13%, $p = 0.608$).

TABLE 8 Changes in the assessment of velopharyngeal function in non-adopted children between 5 and 10 years of age using rating of velopharyngeal competence

		VPC-Rate at age 10			
		0	**1**	**2**	**Total**
VPC-Rate at age 5	0	8	2	2	12
	1	4	1	1	6
	2	3	2	0	5
Total		15	5	2	23

Notes: 0 = competent VPF, 1 = marginally incompetent VPF, 2 = incompetent VPF.
VPC-Rate, rating of velopharyngeal competence; VPF, velopharyngeal function.

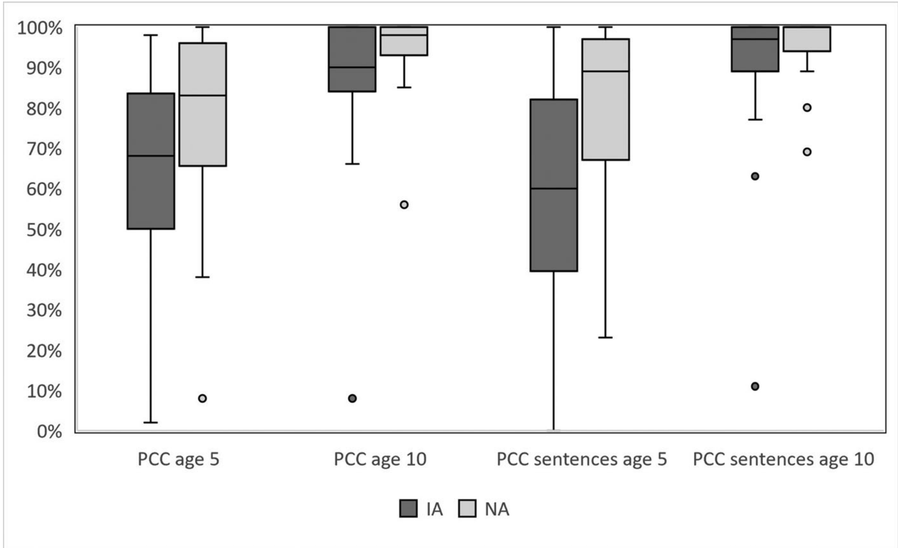

FIGURE 2 Median per cent consonants correct (PCC) at word level and sentence level at ages 5 and 10 years for internationally adopted (IA) and non-adopted (NA) children.

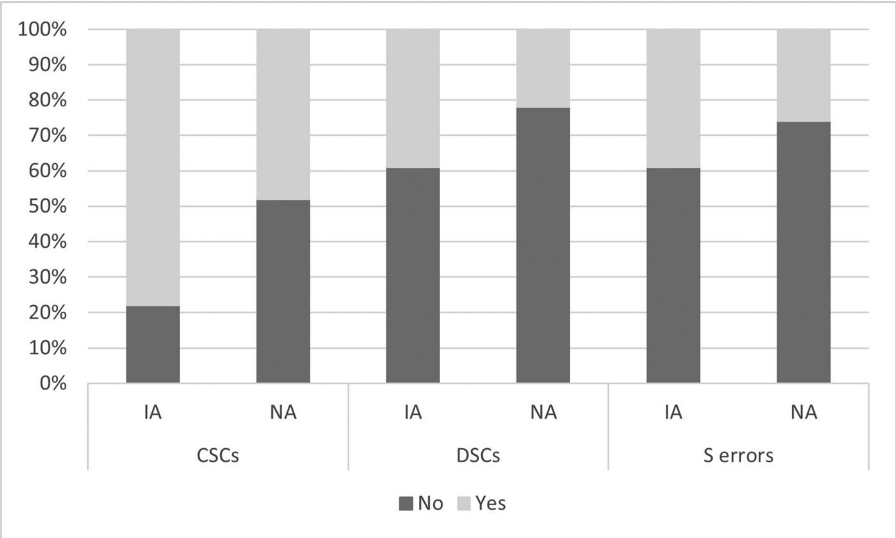

FIGURE 3 Proportions of internationally adopted (IA) and non-adopted (NA) children with articulation errors divided into cleft speech characteristics (CSCs), developmental speech characteristics (DSCs) and s-errors at age 5 years.

At age 5 years, DSCs did not differ significantly between IA and NA children ($p = 0.337$) nor did s-errors ($p = 0.265$). When comparing subtypes of DSCs, velar fronting (IA 22%, NA 13%, $p = 0.699$), stopping (IA 30%, NA 17%, $p = 0.491$), and voicing errors (IA 9%, NA 4%, $p = 1.0$), no significant differences were found between the groups (Figure 3).

At age 10 years, CSCs differed significantly between the groups ($p = 0.023$). Oral retracted articulation remained in 26% of IA children and 9% of NA children ($p = 0.243$), while non-oral articulation was still present in 22% of IA children and 4% of NA children ($p = 0.187$). Glottal plosive was present in 13% of IA children and 0% of NA children ($p = 0.233$), while active nasal fricative was present in 17% of IA children and 4% of NA children ($p = 0.346$).

Regarding DSCs, two IA children still had minor difficulties at age 10 years, one with voicing errors and one with stopping. s-errors did not differ significantly between the groups ($p = 0.414$) (Figure 4).

Secondary palatal surgery

For details on secondary palatal surgeries, see Table 9. One NA child had a re-repair of the soft palate due to dehiscence at age 1;5 and later received a pharyngeal flap (age 3;4). The difference between the proportion of IA and NA children receiving speech-improving surgery was not significant ($p = 0.530$).

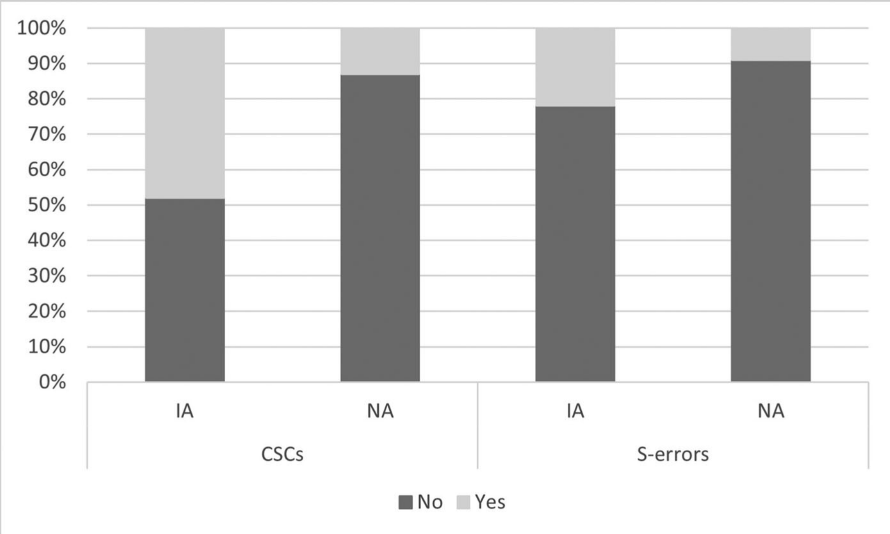

FIGURE 4 Proportions of internationally adopted (IA) and non-adopted (NA) children with articulation errors divided into cleft speech characteristics (CSCs) and s-errors at age 10 years. Only two IA children had developmental speech characteristics (DSCs) (not shown).

TABLE 9 Secondary palatal surgeries

	Internationally adopted	Non-adopted
Secondary surgery due to dehiscence, *n* (age)	–	1 (1;5)
Fistula repair, *n* (mean age, SD, range)	4 (3;9, 10, 3;1–4;10)	3 (3;5, 11, 2;5–4;2)
Pharyngeal flap < age 5 years, *n* (mean age, SD, range)	5 (4;2, 8, 3;3–4;9)	2 (3;8, 6, 3;4–4;1)
Pharyngeal flap age 5–10 years, *n* (mean age, SD, range)	4 (6;5, 13, 5;3–7;7)	4 (6;2, 17, 5;3–8;3)

Note: Mean ages and ranges are given as years;months, SD is given in months.

Visits to SLP

Mean number of visits to an SLP at age 5 years was 19 (range = 4–35, $n = 20$) in the IA group and 14 in the NA group (range = 3–30, $n = 18$). Between 5 and 10 years of age, the mean number of visits was 12, 5 (range = 2–44, $n = 21$) in the IA group and 8 in the NA group (range = 1–31, $n = 18$). No significant differences were detected between the groups at either age ($p = 0.450$ and 0.335, respectively).

DISCUSSION

This study investigated how articulation and VPF developed longitudinally between ages 5 and 10 years in IA and NA children. Also, it compared VPF, articulation, the proportion of secondary palatal surgery and number of visits to an SLP between these groups. It adds new knowledge of how articulation and VPF develop over time in IA children and how their speech ability is at an older age than previously described. The study demonstrated that developmental articulation errors had ceased at age 10 years in both groups. However, cleft-related articulation errors were still present in significantly more IA than NA children, and despite the significant development noted, the IA children as a group did not catch up.

Based on previous studies, a somewhat unexpected finding was that the proportion of IA children with incompetent VPF at age 5 years in this study was smaller than reported earlier (Larsson et al., 2020; Scholin et al., 2020). In past studies, as many as half of IA children had incompetent VPF compared with 30% in the present study. The difference between IA and NA children with incompetent VPF was also considerably larger in past studies than in the present study. The proportion of NA children assessed to have incompetent VPF was similar in all three studies, indicating that VPF was assessed with a similar definition of the scale steps, and differences in the assessment can, therefore, not explain the difference between the studies. Further, mean age at arrival to Sweden, at SPC and HPC, and the proportions of secondary surgery were also similar in the studies and cannot explain the difference between them. The three studies differed concerning included cleft types. Larsson et al. (2020) included only children with UCLP, while Scholin et al. (2020) included children with UCLP, BLCP, and isolated cleft palate. However, it is not certain that this can explain the different outcomes regarding VPF.

Another interesting finding was the discrepancy between VPC-Sum and VPC-Rate, especially in IA children at age 10 years. VPC-Sum revealed a significant difference between the groups at both ages, while VPC-Rate did not even show a trend towards a significant difference. Lohmander et al. (2017a) reported the level of agreement between the two measurements to be 65.2%, in line with what was found in this study. As the VPC-Rate is based on connected speech, which is more like speech in real-life, it is considered to have high external validity. However, similar proportions of children with incompetent VPF in both groups at both ages were identified using both measurements, and the difference lay in the distribution of competent and marginally incompetent VPF. VPC-Sum turned out to be more sensitive than VPC-Rate. This is probably explained by the fact that VPC-Sum is based on transcriptions of target consonants where occurrences of passive and active speech deviations are counted, and VPC-Rate is based on an overall assessment of connected speech. Regardless of the method used to evaluate VPF, those rated with competent VPF were noticeably fewer in the IA group at both ages. With more children in both groups receiving speech-improving surgery, VPF improved between 5 and 10 years.

Children in the IA group scored lower PCC than NA children at both 5 and 10 years, at word and sentence levels. However, most IA and NA children scored below an age-appropriate level. For IA children at ages 5 and 7–8 years, it has previously been reported that only one child scored an age-appropriate PCC, and 16 of the 17 participants scored > 2 SD below the age-specific norm. Further, at ages 7–8 years, most children scored > 4 SD below the age-specific norm (Larsson et al., 2022). As many as 57% in the IA group and 35% in the NA group in the present study scored > 4 SD below the mean score in typically developed children without CLP at age 5 years. Despite significant improvement in both groups, only 30% of the IA children and 65% of the NA children scored at an age-appropriate level, indicating that they still had considerable articulatory difficulties compared with peers without CLP. It is reasonable to believe that the extensive articulation difficulties seen in some children in both groups at age 5 years and mainly in the IA group at age 10 years affected intelligibility and, thus, the opportunity to participate in various contexts.

As PCC does not reveal the character of the articulation error, and different articulation errors affect speech perception differently (Sell & Sweeney, 2020), we chose to divide them into different categories for further analysis, as described previously (Larsson et al., 2020; Morgan et al., 2018; Willadsen et al., 2017). Both CSCs and DSCs were common in both groups at age 5 years, but almost twice as many IA children as NA children presented with some type of CSC and/or DSC. In this study, the proportion of children in each group with some type of CSC (IA 78%, NA 48%) was similar to that reported by Larsson et al. (2020). However, the results differ with respect to DSCs. Larsson et al. (2020) reported that 92% of their IA group and 65% of their NA group presented with articulation errors related to development. In the present study, the corresponding proportions at age 5 years were 39% in the IA group and 22% in the NA group. A difference in voicing errors largely explained this discrepancy. A total of 76% of IA children and 55% of NA children presented with such errors in the study by Larsson et al. (2020), compared with 9% in the IA group and 4% in the NA group in the present study at age 5 years; at age 10 years, only one child (IA) had voicing errors. Voicing errors were reported by Morgan et al. (2018) to be common in both adoptees and non-adoptees at ages 7–8 years, and in the longitudinal study of IA children, 71% still had voicing errors at ages 7–8 years (Larsson et al., 2022). One explanation for the difference between the present study and earlier work (Larsson et al., 2020, 2022; Morgan et al., 2018) may be difficulties differentiating between a voiced plosive and a voiceless plosive articulated with reduced pressure. Another explanation may be that the SLP focuses more on errors related to VPI, for example, weak pressure consonants, or more phonological aspects, for example, voiced plosives.

Substantial development was seen from age 5 to 10 years in both groups, and errors related to development were seen in only two children (IA) at age 10 years. This suggests that even though DSCs have been reported to be present at ages 7–8 years (Larsson et al., 2022; Morgan et al., 2018), these errors are not as persistent as CSCs. Oral retracted articulation is a common articulation error in children with CLP (Lohmander-Agerskov, 1998), especially when delayed HPC is practiced. In the study by Larsson et al. (2017), oral retracted articulation was more common in NA children than in IA children at age 3 years. The fact that NA children had a residual cleft in the hard palate for a more extended period after SPC was given as an explanation for the results. At age 5 years, the situation was reversed, and oral retracted articulation was noted more frequently in the IA group (Larsson et al., 2020). However, one-third also exhibited velar fronting, suggesting an unstable sound system. The trend of IA children more often having oral retracted articulation than their peers were also seen in the present study, and at age 5 years, approximately one-sixth of the IA children simultaneously had velar fronting. At age 10 years, no child exhibited velar fronting, but more IA than NA children still had oral retracted articulation, suggesting that this articulation error was more persistent in IA children. Unfortunately, about a quarter of the IA children also still had issues with non-oral articulation, which is associated with VPI and impairs intelligibility even more than retracted oral articulation (Lohmander-Agerskov, 1998). As the IA children had their surgery later than the NA children, the articulation pattern may already have been established at the time of the first surgery, possibly making it more challenging to correct. As shown earlier (Larsson et al., 2017, 2020; Morgan et al., 2018; Scherer et al., 2018; Scholin et al., 2020), the results of this study indicated that IA children have more articulation errors than NA peers with CLP. Further, the results suggested that articulation errors related to the cleft are more persistent in IA children, remaining well into school age. Considering the articulation difficulties seen in both groups, one could have expected to see that reflected in the number of visits to an SLP. However, the mean number of visits to an SLP in both groups was relatively low, especially between ages 5 and 10, and did not differ even though more IA children had articulation difficulties. Providing more intervention may not always be possible since it depends partly on the child's maturity, ability and willingness to participate, and accessibility to an SLP. However, as IA children seem at a higher risk of more persisting and severe articulation difficulties than NA children, it is crucial, when needed, to instigate therapy early and not have a "wait and see" approach. Many children, not least in the IA group, still at the age of 10, had severe speech difficulties. They would probably have benefited from more comprehensive therapy, ideally from an early age, and continued through the early school years.

Our results contradict many of those reported earlier on secondary palatal surgeries, which have been reported to be required considerably more often in IA children than in NA children (Hansson et al., 2012; Morgan et al., 2018; Swanson et al., 2014). Although the proportion was somewhat higher in the IA group in the present study, it did not differ significantly between the groups. This finding should be interpreted cautiously due to the relatively small sample and possible differences in surgical indications between centres. However, differences in surgical results between centres that appear in a subset of challenging-to-operate patients cannot be ruled out.

Several methodological considerations need to be pointed out. In earlier studies, deletion of consonants and replacement with glottal plosive have been reported separately (Larsson et al., 2017, 2020). This is recommended (Sell & Sweeney, 2020), as deletion of consonants can be interpreted as developmental immaturity (Dodd et al., 2003). However, it has also been shown that it is hard to distinguish between consonant deletion and replacement with a glottal plosive (Willadsen et al., 2017). For this reason, we counted each occurrence of consonant deletion as an elicited target sound and categorized it as an occurrence of glottal plosive.

s-errors occur in just under 30% of Swedish 5-year-olds without cleft palate (Lohmander et al., 2017b), and some earlier studies have reported PCC adjusted for age, with s-errors regarded as correct (Klinto et al., 2014, 2016; Larsson et al., 2020). However, in this study, all s-errors were scored as incorrect. s-errors were present in 39% of IA children and 26% of NA children at age 5 years. The initial aim was to divide s-errors into CSCs and DSCs, but they proved hard to categorize in some cases. Furthermore, when scrutinizing the transcriptions, it was found that the main transcriber had only used the sign for alveolo-palatal fricative (ɕ) and never the sign for palatal fricative (ç). As the alveolo-palatal fricative is a Swedish phoneme and consequently would have been categorized as a DSC, and the palatal fricative is a common CSC, the decision was made not to categorize s-errors as CSCs or DSCs, but to report them separately.

Another methodological consideration in this study is the lack of information on hearing status. Children with cleft palate often exhibit otitis media with effusion, associated with mild conductive hearing loss (Flynn et al., 2009), at least at pre-school age. No child in the study had a severe hearing impairment requiring a hearing aid, but we cannot rule out that some had mild conductive hearing loss. However, the impact of hearing on speech outcomes is sparsely studied. Mild hearing loss was demonstrated to impact the presence of canonical babbling at 10 months, and hearing level at age 18 and 30 months was significantly correlated with PCC at age 36 months (Lohmander et al., 2021), indicating mild conductive hearing loss has an impact on the early development of consonant production. On the other hand, hearing was not associated with speech outcomes at age 3 years (Klinto et al., 2014) or age 5 (Fitzpatrick et al., 2021).

An initial aim of this study was to include an assessment of intelligibility, which would have given important information about the impact of different speech deviations. Recording of spontaneous speech was sparse for most children in both groups at age 5 years, often due to the children's unwillingness or inability to elaborate answers despite encouragement from the SLP who met the children or the SLP's inability to understand the children, which put the conversation to a stop. At age 10 years, the figures had improved, but a considerable proportion of recordings still did not include enough spontaneous speech to enable assessment of intelligibility.

The limited sample size and the heterogeneity in the IA group may have affected the results, and as small studies are vulnerable to outliers, the results should be interpreted cautiously. However, the results in this study point in the same direction as results already reported and can therefore be considered reliable.

CONCLUSIONS

The IA children in the present study did not have VPI to a greater extent than the NA children, but fewer were judged to have a fully competent VPF. Developmental articulation errors seemed to have ceased at age 10 years, also in most IA children. However, both groups presented with developmental articulation errors at age 5 years, and intervention should thus target both developmental and cleft-related articulation errors. It is evident from the results that IA children have greater and more severe articulation difficulties up to the age of 10 and that they probably require more extended intervention than NA children.

ACKNOWLEDGEMENTS

The authors gratefully acknowledge SLPs Malin Appelqvist Gajsek at Uppsala University Hospital, Kristina Klintö at Lund University and Justin Weinfeld at Sahlgrenska University Hospital for helping with the transcriptions and assessment of velopharyngeal function. Parts of the results was presented at the 14th International Cleft Congress Edinburgh, Scotland, UK, July 2022.

CONFLICT OF INTEREST STATEMENT

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

DATA AVAILABILITY STATEMENT

The datasets generated during and/or analysed during the current study are available from the corresponding author upon reasonable request.

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ENDNOTES

[1] Shriberg and Kwiatkowski (1982) originally developed the measure of PCC in conversational speech. A modified version of PCC in single-word samples has been used to assess articulation skills in children born with cleft palate (Lohmander & Persson, 2008; Scherer et al., 2008; Larsson et al., 2020).

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How to cite this article: Okhiria, Å, Persson, C., Johansson, M.B., Hakelius, M. & Nowinski, D. (2023) Longitudinal data on speech outcomes in internationally adopted children compared with non-adopted children with cleft lip and palate. *International Journal of Language & Communication Disorders*, 58, 1440–1453. https://doi.org/10.1111/1460-6984.12869