Better speech outcomes after very early palatal repair? – A longitudinal case-control study in Ugandan children with cleft palate

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Abstract

Introduction. Children born with cleft palate with or without cleft lip (CP±L) tend to use less oral pressure consonants and more glottal sounds in their babbling. The purpose of very early palatal repair (i.e., one-stage palatal closure prior to 6 months of age) is to make the palate functional before the onset of speech acquisition to reduce the anchoring of wrong patterns in the child's developing phonological system. As a result, less compensatory articulation errors are expected to be present. Currently, no detailed longitudinal speech outcomes after very early palatal closure are available. This study aimed to provide longitudinal speech outcomes in Ugandan children with CP±L who received palatal closure prior to the age of 6 months.

Methods. Ten children with CP±L were assessed at a mean age of 5 and 10 years old. Speech understandability, speech acceptability, resonance, nasal airflow and articulation were perceptually rated by two experienced speech-language pathologists. Velopharyngeal function was estimated using the velopharyngeal composite score (VPC-sum). Information regarding speech therapy, fistula rate, and secondary (speech) surgery was collected. The outcomes were compared with the longitudinal outcomes of an age- and gender-matched control group of 10 Ugandan children without CP±L.

Results. Speech understandability and acceptability improved significantly over time in the group with CP±L (all *p's* ≤ .05, all Z's > -2.43). At both test dates, significantly worse judgments were found for the group with CP±L compared to the control group for these variables and variables related to passive speech errors (all *p's* ≤ .05, all Z's > 2.49). A statistically significant difference with the control group was found for the presence of compensatory articulation errors at the age of 5 years but not at the age of 10 years, indicating a catch up by the children with CP±L.

Conclusion. Whether a one-stage palatal closure prior to the age of 6 months is more favorable for speech outcomes compared to one-stage palatal closure at 12 months is still not clear. Speech of the children with CP±L improved over time, but significantly differed from the control group at the age of 5 and 10 years old. Limited access to health care facilities and possible influence of malnutrition on wound healing need to be considered when interpreting the results. Whether palatal closure prior to the age of 6 months is transferable to other countries is subject for further research, including both longitudinal and prospective designs with larger samples.

1. Introduction

Children who are born with a cleft palate with or without cleft lip (CP±L) often present with speech difficulties. At the age of 6 months, infants with cleft palate may already show a different speech development compared to typically developing children (Peterson-Falzone et al., 2016). More specifically, delayed onset and lower frequencies of canonical babbling and the infrequent production of oral pressure consonants are observed in babies with unrepaired or partially repaired cleft palate (Chapman et al., 2001; Peterson-Falzone et al., 2016; Scherer et al., 2008; Stout et al., 2011; Willadsen & Albrechtsen, 2006). It has been hypothesized that less oral-motor practice of these pressure consonants due to an open palate in the babbling phase may result in less phonological knowledge about these consonants and the anchoring of wrong patterns in the child's developing phonological system (Jørgensen & Willadsen, 2020; Lohmander & Persson, 2008; Russell & Grunwell, 1993; Willadsen & Albrechtsen, 2006). Consequently, compensatory articulation errors (e.g. active nasal fricatives, pharyngeal or laryngeal productions) often persist after palatal closure (Lohmander & Persson, 2008; Nyberg et al., 2014). In addition, velopharyngeal insufficiency may remain after palatal closure resulting in passive speech problems (e.g. hypernasality, audible nasal airflow and nasalized or weak pressure consonants as a result of difficulty in creating intra-oral pressure during the production of these consonants). Cleft related speech errors are often referred to as cleft speech characteristics (CSCs) (John et al., 2006). These speech disorders may severely affect a child's speech understandability and speech acceptability which impacts psychosocial functioning (Bruneel et al., 2019; Murray et al., 2010).

Currently, the generally accepted protocol for palatal repair in Europe and the USA is a one-stage palatal closure at the age of 12 months (Shaw et al., 2019). As a result, most children with CP±L still pass through their babbling phase with an open palate. Studies including this population mention the presence of compensatory articulation errors at the age of 5 years (Nyberg et al., 2014; Sell et al., 2015; Willadsen et al., 2017). More specifically, 10-26% of the included children showed posterior oral articulation errors and 15-22% of them showed non-oral articulation errors. Most of them were, however, referred for speech therapy intervention before this age and additional velopharyngeal surgery was performed when indicated.

Although speech outcomes generally improve over time, speech in children born with CP±L often still differs from the speech of children born without CP±L (Jørgensen & Willadsen, 2020; Lohmander & Persson, 2008; Nyberg et al., 2014). In this light, some authors advocated that the closure of the soft and hard palate prior to 6 months of age results in better speech outcomes (Copeland, 1990; De Mey et al., 2006; Desai, 1983; Doucet et al., 2013; Kaplan, 1981). The purpose of palatal closure prior to the onset of speech acquisition is to optimize the possibility to establish normal (neuro-)motor speech patterns and, as a result, minimizing the development of compensatory articulations (Dorf and

Curtin, 1982). Only a few authors reported on intelligibility, articulation and/or resonance after one-stage palatal closure prior to the age of 6 months (Copeland, 1990; De Mey et al., 2006; Kaplan et al., 1982; Ysunza et al., 1998). Kaplan et al. (1982) found excellent speech (based on judgments of nasality, phonation and intelligibility without providing definitions or rating methods for these concepts) in 52% (11/21) of the included children with an age range of 2 to 7 years. Copeland (1990) performed speech analyses in 100 children with CP±L (mean age 5.5 years, range 3.8-6.3 years) based on spontaneous speech, an articulation assessment and counting from 1 to 10. Based on a visual-analogue scale, 87% (87/100) were judged to be very easy or easy to understand (i.e., score of > 5 on a 10 cm scale). Of these 87 children, 57 had normal articulation and 77 had normal resonance. Ysunza et al. (1998) described articulation to be 'below normal limits' for 35 children at an age of 4 years, although none of them showed compensatory articulation errors. They found significantly better articulation in children who received palatal closure at 6 months compared to children who received palatal closure at 12 months, but there was no significant difference between both groups regarding the presence of velopharyngeal insufficiency (Ysunza et al., 1998). Only De Mey et al. (2006) reported longitudinal results for 18 children with unilateral cleft lip and palate (UCLP) who received a one-stage lip and palatal closure at 3 months. At the age of 3 years, 63% had good intelligibility, 30% had normal articulation and 30% had normal resonance. At the age of 6 years, speech had improved with 80% showing satisfactory intelligibility, 40% having normal articulation and 40% having normal resonance. Unfortunately, definitions or rating methods for these concepts were not provided. Moreover, no detailed information was reported about how many children with good intelligibility had also normal articulation and/or resonance. At a mean age of 5 years, one child had received a velopharyngoplasty and eight children (44%) had received a fistula repair for phoniatric reasons. Speech therapy was provided in 63% (11/18) of the children between the age of 3 and 6 years.

Recently, two cross-sectional studies reported speech outcomes in Ugandan children with CP±L who received palatal closure at a mean age of 3 months. One study included 11 children with CP±L at a mean age of 5 years (range 3-7 years) (Luyten et al., 2013), the other included 24 children with CP±L at a mean age of 8 years (range 6-12 years) (Bettens et al., 2020). Outcomes were compared with an age- and gender-matched control group who presented with normal speech. At the age of 5 years, hypernasality was present in 18% of the children with CP±L, audible nasal airflow in 27% and compensatory articulation errors in 55%. No information regarding speech understandability, speech acceptability and the presence of passive articulation errors was provided. At the age of 8 years, 29% was judged as having hypernasality, 50% as having audible nasal airflow, 16% as having compensatory articulation and 46% as having passive articulation errors. Normal speech understandability was observed in 42% of the children with CP±L and 38% were judged with normal speech acceptability following the definitions provided by Henningsson et al. (2008). The presence of compensatory

articulation errors (i.e., posterior oral and non-oral articulation errors) had decreased over time resulting in the absence of a statistically significant difference with the control group at the age of 8 years. In the study by Luyten et al. (2013), none of the children with CP±L had been enrolled in speech therapy at a mean age of 5 years, whereas 25% of the children with CP±L in the study by Bettens et al. (2020), had received speech therapy at a mean age of 8 years. This may suggest the positive effect of speech therapy intervention on the presence of compensatory articulation errors. On the other hand, velopharyngeal insufficiency had become worse, given the increased amount of children with CP±L presenting with hypernasality and audible nasal airflow in the study by Bettens et al. (2020). Conclusions about the evolution of speech over time need to be done carefully because of the cross-sectional design of the studies. Luyten et al. (2014) also compared the speech of Ugandan children with CP±L who received a palatal closure prior to the age of 6 months (mean 3 months, range 2-6 months) with those of Belgian children with CP±L who received their palatal closure after the age of 6 months (mean 11 months, range 9-15 months). At a mean age of 5 years (range 3-7 years), no significant differences were found regarding resonance, audible nasal airflow, and the presence of compensatory and passive articulation between both groups. The authors concluded that at a mean age of 5 years, the articulation and resonance characteristics following palatal repair before and after 6 months of age were comparable. Important group differences were noticed regarding the enrollment in speech therapy and insertion of ventilation tubes. Half of the group with CP±L who received their palatal closure after the age of 6 months (i.e., Belgian group) was enrolled in speech therapy to reduce cleft related speech disorders and 75% of them received ventilation tubes due to otitis media in contrast to none of the children who received their palatal closure prior to the age of 6 months (i.e., Ugandan group). No information was provided regarding the need for speech therapy or the need for ventilation tubes in the Ugandan group with CP±L. It could be assumed that (some) of the Ugandan children with CP±L were in need of intervention but did not receive it because these services were not available.

Currently, no longitudinal results have been published about speech outcomes after one-stage palatal closure before the age of 6 months including a detailed analysis of all speech characteristics (i.e., understandability, acceptability, resonance, audible nasal airflow and articulation) and a profound description of the used speech assessment protocol (i.e., rating scales, raters, speech samples and reliability). The present study aimed to provide longitudinal speech outcomes in Ugandan children with CP±L who received palatal closure prior to the age of 6 months using a retrospective longitudinal matched case-control study design. Speech outcomes included perceptual ratings of speech understandability, speech acceptability, resonance, nasal airflow and articulation at a mean age of 5 and 10 years using an internationally accepted assessment protocol (John et al., 2006). Velopharyngeal function was estimated using the velopharyngeal composite score (VPC-sum, Lohmander et al. (2009))

based on perceptually rated speech outcomes. Information regarding speech therapy, fistula rate, and secondary (speech) surgery was collected. The longitudinal speech outcomes were compared with those of an age- and gender-matched control group of Ugandan children without CP±L with a comparable language and cultural background. It was hypothesized that speech improved over time in all children, with the speech of the children with CP±L still differing from the speech of children without CP±L at the age of 10 years.

2. Methods

This study was approved by the *blinded for review* Research Ethics Committee (0611-2017) and the *blinded for review* (HS 2448).

2.1 Participants

Between January 2011 and September 2014, our research unit regularly visited the Comprehensive Rehabilitation Services in Uganda (CoRSU) hospital in Entebbe, a city in the southeast of Uganda, in the context of a VLIR-UOS project (ZEIN2009EL28). We provided speech assessments and speech therapy in children with CP±L in collaboration with local staff. Within this project, speech samples from Ugandan children with and without CP±L were collected by our research unit. Children with CP±L were seen at CoRSU hospital (N = 207). Children without CP±L were recruited from an orphanage located near CoRSU hospital (N = 75). These data, collected between January 2011 and September 2014, were used to select eligible participants for the current study. In addition, the data were used for the speech analyses to determine speech outcomes at the first test date (i.e., test date 1, T1).

Criteria for inclusion into the group with CP±L were (1) born with an isolated cleft palate with or without cleft lip, (2) performed full speech assessment with good cooperation according to age at a previous consultation (i.e., T1), (3) multilingual with English as a second language, (4) receiving educational instruction in English, (5) received one-stage primary palatal closure before the age of 6 months at CoRSU hospital, (6) palatal closure included the Sommerlad technique with radical muscle repositioning and bilateral lateral Langenbeck-type releasing incisions when indicated (Hodges, 2010), (7) in case of a cleft lip, lip closure was performed simultaneously with the palate repair, and (8) lip closure was done by using a modified Millard repair in unilateral clefts and a modified Mulliken repair in bilateral clefts (Hodges, 2010). Children who received a vomer flap with delayed (after the age of 6 months) hard palate closure were excluded. Children needed to be multilingual with English as a second language as all speech assessments were conducted in English, due to a large variation in native languages in Uganda. English is one of the official languages in Uganda, deeply rooted in media, administration and education (Mpuga, 2003). A total of 34/207 children fulfilled the criteria and their

caregivers were contacted via phone. After they received information about the current study aim, they were invited to let their child participate voluntarily. Ten children presented at CoRSU hospital in November 2018 or March 2019. A child was excluded if he/she was not attending school, showed insufficient English proficiency, had severe speech difficulties not related to the cleft (e.g., stuttering), had self-reported hearing problems at the moment of testing, had nasal congestion due to a cold or allergy at the moment of testing, or was not able to perform the assessments. None of the children were excluded based on these criteria. Speech analyses were performed on the collected data to determine speech outcomes at the second test date (i.e., T2).

Six boys and four girls with CP±L were included in this study. Six children were born with a unilateral cleft lip and palate, two with a bilateral cleft lip and palate and two with a cleft palate only. Palatal closure was performed at a mean age of 2.8 months (SD 1.14, range 1-5 months). The mean age of the children at T1 was 4.5 years (SD .85, range 4-6 years), the mean age of the children at T2 was 9.7 years (SD 1.64, range 8-13 years).

Based on this experimental group, the database including 75 children without CP±L was used to select ten children for the control group. Speech samples of a fully performed speech assessment with good cooperation according to age at a previous test moment (i.e., T1) needed to be available. Other inclusion criteria were: (1) no (history of) craniofacial anomalies, (2) multilingual with English as a second language, and (3) receiving educational instruction in English. Children with nasal congestion at the moment of testing were excluded. The caregivers of the eligible children were provided with information about the current study aim and were invited to let the child participate voluntarily. Data were collected and speech analyses were performed on these data to determine speech outcomes at the second test date (i.e., T2).

Five boys and five girls were included in the control group. Although we tried to match by gender, no data of a sixth boy with a suitable age were available in the database. Age-matching was based on group means, taking the age-range of the group with CP±L into account. The mean age of the children at T1 was 4.0 years (SD .82, range 3-5 years), the mean age of the children at T2 was 10.1 years (SD 1.52, range 9-13 years). Based on a Mann-Withney U test, no significant difference was found regarding the age of the group with and without CP±L (T1: $U = 65.5$, $p = .247$; T2: $U = 44.5$, $p = .684$).

During an interview with the child and the child's primary caregiver, information regarding age, gender, language, school attendance, nasal regurgitation and speech therapy was obtained. The children's medical records (only children with CP±L) were screened to collect medical information (e.g., age at palatal closure, secondary surgery and presence of oral fistula).

2.2 Perceptual speech assessment

The same speech samples were collected at two different test moments, i.e. T1 at 5 years and T2 at 10 years. Participants were asked to repeat the oral and nasal sentences from the MacKay-Kummer Simplified Nasometric Assessment Procedures (SNAP) test (Kummer, 2005) and the high frequency English words cued by pictures of the Photo Articulation Test – Third Edition (PAT-3) (Lippke et al., 1997). High-quality audio(visual) recordings were made using a unidirectional condenser microphone (Samson C01U) and a Sony Handycam HDR—UX1 with built-in high quality microphone.

Two speech-language pathologists (SLPs) with 3 and 8 years of experience in rating cleft palate speech characteristics perceptually rated the audio and video samples. One of them was officially trained in using the Cleft Audit Protocol for Speech- Augmented (CAPS-A) outcome tool (John et al., 2006; Sell et al., 2009). They were both native speakers of Dutch with professional proficiency in English. They independently rated all samples in a randomized sequence using a standard pair of over-ear headphones (Sennheiser EH150). Intra-rater reliability was verified by randomly presenting 30% (12/40) of the speech samples a second time during the same rating session.

The definitions and ordinal rating scales described by Henningsson et al. (2008) were used to evaluate speech understandability and speech acceptability. Hypernasality, hyponasality, audible nasal emission, nasal turbulence, voice and grimace were perceptually judged using the definitions and ordinal rating scales of the CAPS-A outcome tool (John et al., 2006; Sell et al., 2009). The evaluation of these variables was based on sentence repetition (SNAP test, Kummer (2005)).

To evaluate the articulation, 48 of the 72 items from the PAT-3 were phonetically transcribed by both SLPs. Only those words that included the 18 consonants (i.e., /p/, /b/, /t/, /d/, /k/, /g/, /s/, /z/, /f/, /v/, /h/, /w/, /j/, /l/, /r/, /n/, /m/, and/ŋ/) that occur both in English and Luganda (Luyten et al., 2013), were included in this transcription. This was done because Luganda was the first language in 85% of the children. For each consonant, word initial, medial and final position was transcribed if that position was present in English (e.g., no word initial position of /ŋ/). Following the classification of CSCs provided by John et al. (2006), articulation errors were assigned to one of the following categories: anterior CSCs (i.e., dentalization, lateralization, palatalization), posterior CSCs (i.e., double articulation, velar or uvular articulation), non-oral CSCs (i.e., pharyngeal articulation, glottal articulation or reinforcement, active nasal fricative) or passive CSCs (i.e., weak or nasalized consonants, nasal realization of plosives, suspected passive nasal fricative, gliding of fricatives). When at least one compensatory speech error (i.e., anterior CSC, posterior CSC or non-oral CSC) was detected that had an impact on speech understandability and/or acceptability, the child was judged to be in need of speech therapy.

Velopharyngeal functioning was estimated based on the perceptual judgment of four speech variables of the CAPS-A tool resulting in the velopharyngeal composite score (VPC-sum) (Lohmander

et al., 2009). More specifically, VPC-sum was calculated based on the presence or absence of hypernasality, audible nasal airflow, non-oral CSCs and passive CSCs, resulting in a VPC-sum score between 0 and 4. Based on this score, velopharyngeal function was considered to be sufficient (score 0 or 1), borderline insufficient (score 2) or insufficient (score 3 or 4). Instrumental assessment by nasoendoscopy or videofluoroscopy is necessary to complement this perceptual estimation. However, no such instrumental equipment was available at CoRSU hospital at that moment.

2.3 Statistical analysis

IBM SPSS Statistics software version 25.0 (IBM Corp., Armonk, NY) was used for the statistical analysis of the data. To determine the intra-rater and inter-rater reliability of the dichotomous variables, Cohen's kappa was used. Intraclass Correlation Coefficients (ICCs) were calculated for the ordinal speech variables. A two-way mixed model with absolute agreement was used to determine intra-rater reliability following the interpretation by Altman (1991). Inter-rater reliability of the ordinal speech variables was verified using a two-way random model with absolute agreement. Percentages of absolute agreement were calculated within each rater and between raters to complement the ICCs as limited variance might cloud the interpretation of ICC measurements (Hallgren, 2012).

Wilcoxon paired-signed rank tests compared the results between T1 and T2 for the group with CP±L and for the control group separately. This test was chosen because of the paired characteristics of the data and the small sample. To detect a possible difference between the children with and without CP±L, Mann-Whitney U tests were performed on the data of T1 and on the data of T2. A Mann-Whitney U test was chosen because of the absence of paired data and the small sample. All significance levels were set at $\alpha = .05$.

3. Results

3.1 Medical information

Table 1 provides medical information of the children in the group with CP±L. Secondary surgery was performed in 5/10 children before T1. This surgery included fistula closure (n = 2), palatal re-repair (n = 1), palatal re-repair with fistula closure (n = 1) or palatal re-repair followed by a buccal flap (n = 1). One child received fistula closure between T1 and T2 at the age of 4 years.

At T1, 4/10 children with CP±L reported nasal regurgitation of whom one had a fistula. A fistula was also present in a another child without the presence of nasal regurgitation. Only one of the two children with a fistula at T1 presented without a fistula at T2. This was due to fistula closure between both test dates. This child did not reported nasal regurgitation anymore at T2. However, at T2, two

Table 1. Medical information for the included children with CP±L

Child	**Gender**	**Cleft type**	**Age at palatal closure (in months)**	**Fistula present at test date 1?**	**Nasal regurgitation present at test date 1?**	**Fistula present at test date 2?**	**Nasal regurgitation present at test date 2?**	**Secondary surgery**	**Age at secondary surgery (in years)**
1	female	CP	3	no	no	no	no	Palatal re-repair Buccal flap	0.5 1.5
2	male	UCLP	3	no	yes, on food	no	no	N/A	
3	female	CP	2	no	no	no	no	Palatal re-repair	2
4	male	UCLP	2	no	no	yes	yes, on liquids and food	N/A	
5	male	UCLP	4	no	no	no	no	N/A	
6	female	BCLP	3	no	no	no	no	Palatal re-repair + fistula closure	3
7	male	UCLP	1	no	yes, on liquids and food	no	no	Fistula closure	4
8	male	UCLP	3	no	yes, on liquids	yes	yes, on liquids	N/A	
9	female	UCLP	2	yes	no	yes	yes, on liquids and food	Fistula closure	4
10	male	BCLP	5	yes	Yes, on liquids and food	no	no	Fistula closure	1

CP: cleft palate; UCLP: unilateral cleft lip and palate, BCLP: bilateral cleft lip and palate

N/A: not applicable

children had a fistula who did not present with a fistula at T1. Both of them also reported nasal regurgitation at T2, but not at T1 (Table 1).

Two children with CP±L had received speech therapy due to cleft speech characteristics between T1 and T2. One child produced an active nasal fricative as a substitute for the sounds /s/ and /z/, the other child showed pharyngeal and glottal productions of respectively fricatives and plosives. They were both enrolled in an intensive speech therapy camp of one week and received six hours of therapy.

3.2 Perceptual speech assessments

Reliability. Overall, good to very good intra-rater agreements were found based on ICC and Cohen's Kappa, although some exceptions were noticed. Rater 2 showed poor intra-rater agreement based on ICCs or Cohen's Kappa for the variables 'audible nasal emission' (ICC = 0.00), 'posterior CSCs' (ICC = 0.00), 'non-oral CSCs' (ICC = 0.42) and 'non-cleft speech errors' (Kappa = .29). Rater 1 also showed poor intra-rater agreement for the variable 'audible nasal emission' based on ICC (ICC = -0.09). On the contrary, percentages of absolute agreement for these variables were good (rater 1: audible nasal emission: 83%; rater 2: audible nasal emission: 92%, posterior CSCs: 92%, non-oral CSCs: 83%, non-cleft speech errors: 92%), indicating good reliability as the presence of limited variance in these variables probably resulted in low ICCs. For all other variables, ICC or Cohen's Kappa ranged from .77 to 1.00 for rater 1 and from .65 to 1.00 for rater 2 (Table 2). Regarding inter-rater reliability, overall good to very good agreements were found based on ICC or Cohen's Kappa (Table 3). Only for the variables 'audible nasal emission', 'hyponasality', and 'non-cleft errors' poor agreement was noted. However, the percentage of absolute agreement for these variables was high, indicating good reliability as the presence of limited variance for these variables probably resulted in a low ICC. Nevertheless, 'non-cleft errors' also showed low percentages of absolute agreement, indicating low inter-rater reliability for this variable. Analyses were continued based on the ratings of rater 1, the most experienced SLP, due to better intra-rater reliability compared to rater 2.

Perceptual speech variables. Table 4 provides descriptive statistics (i.e., median values and interquartile ranges) and statistical values of the comparison over time for the children with CP±L. Significant improvements were found in the children with CP±L for the outcome variables 'speech understandability, 'speech acceptability', 'non-oral CSCs', 'non-cleft speech errors' and 'need for speech therapy' between T1 and T2. At T1, eight children with CP±L (80%) were judged to be in need of speech therapy due to at least one compensatory speech error. At T2, three of them (30%) still needed speech therapy. Of the eight children who were in need of speech therapy at T1, two had

Table 2. Intra-rater reliability

Speech parameter	Rater 1		Rater 2	
	ICC/ κ	% Absolute agreement	ICC/ κ	% Absolute agreement
Speech understandability**	.85	58	.90	92
Speech acceptability**	.91	75	.80	83
Hypernasality**	.99	92	.86	75
Hyponasality**	N.A.	100	N.A.	100
Audible nasal emission*	-.09	83	.00	92
Nasal turbulence**	N.A.	100	.80	92
Voice*	1.00	100	1.00	100
Grimace*	1.00	100	1.00	100
Anterior oral CSCs**	.86	83	.67	67
Posterior oral CSCs **	N.A.	100	.00	92
Non-oral CSCs **	N.A.	100	.42	83
Passive CSCs **	.87	92	.78	83
Non-cleft speech errors	.75	92	.29	83
VPC-sum**	.77	92	.65	92
Indication for speech therapy*	.80	92	1.00	100

Abbreviations: K, Cohen's kappa; ICC, intraclass correlation coefficient; CSCs, cleft speech characteristics; VPC-sum, velopharyngeal composite score; N.A., not applicable - ICC impossible to calculate due to zero variance or negative covariance.

*Dichotomous parameters: Cohen's kappa (κ)

**Ordinal parameters: Two-way mixed ICC, absolute agreement, single measures

Interpretation by Altman (1991): <0.40 poor; 0.40-0.59 moderate; 0.60-0.74 good; 0.75-1.00 very good.

Table 3. Inter-rater reliability

Speech parameter	ICC/ κ	95% CI	Interpretation by Altman (1991)	Mean % absolute agreement
Speech understandability**	.93	.87 - .97	very good	75
Speech acceptability**	.92	.83 - .96	very good	73
Hypernasality**	.90	.80 - .95	very good	75
Hyponasality**	.00	-.89 - .47	poor	98
Audible nasal emission**	-.11	-1.16 - .42	poor	90
Nasal turbulence**	.56	.18 - .77	moderate	88
Voice*	.68	N/A	good	93
Grimace*	N/A	N/A	N.A.	98
Anterior oral CSCs**	.49	.03 - .73	moderate	58
Posterior oral CSCs**	.45	-.04 - .71	moderate	90
Non-oral CSCs**	.82	.66 - .91	very good	83
Passive CSCs**	.67	.40 - .84	good	70
Non-cleft errors*	.15	N/A	poor	63
VPC-sum**	.89	.79 - .94	very good	85
Need for speech therapy*	.57	N/A	moderate	85

Abbreviations: K, Cohen's kappa; ICC, intraclass correlation coefficient; CSCs, cleft speech characteristics; VPC-sum, velopharyngeal composite score; N/A, not applicable, ICC impossible to calculate due to zero variance.

*Dichotomous parameters: Cohen's kappa (κ)

**Ordinal parameters: Two-way mixed ICC, absolute agreement, single measures

Table 4. Descriptive statistics of the perceptually rated speech parameters for the children with CP±L (*n* = 10) at test date 1 (mean age 5y) and test date 2 (mean age 10y).

Perceptually rated speech parameter	***Test date***	**Scale** (%)					***Z***	***p***
		WNL	*Mild*	*Moderate*	*Severe*			
Speech understandability	1	0	40	20	40		-2.62	**.009***
	2	40	20	40	0			
Speech acceptability	1	0	30	40	30		-2.43	**.015***
	2	30	40	20	10			
		Absent	*Borderline*	*Mild*	*Moderate*	*Severe*		
Hypernasality	1	40	30	0	0	30	-.214	.831
	2	30	20	0	20	30		
		Absent	*Mild*	*Marked*				
Hyponasality	1	100	0	0			.000	1.000
	2	100	0	0				
		Absent	*Occasional*	*Frequent*				
Audible nasal emission	1	90	10	0			-1.000	.317
	2	70	30	0				
Nasal turbulence	1	80	20	0			-1.518	.129
	2	60	30	10				
		Absent	*Present*					
Grimace	1	100	0				-1.000	.317
	2	90	10					
		Normal	*Distinctive or abnormal voice quality*					
Voice	1	80	20				-.447	.655
	2	90	10					
		No consonants affected	*1-2 consonants affected*	*≥3 consonants affected*				
Anterior CSCs	1	40	30	30			-.264	.792
	2	20	60	20				

Posterior CSCs	1	70	30	0	-1.414	.157
	2	90	10	0		
Non-oral CSCs	1	30	40	30	-2.333	**.020***
	2	80	10	10		
Passive CSCs	1	40	30	30	.000	1.000
	2	30	50	20		
		Sufficient VPF	*Borderline deficit*	*Insufficient VPF*		
VPC-sum	1	60	30	10	-0.857	.391
	2	60	40	0		
		Absent	*Present*			
Non-cleft speech errors	1	10	90		-2.126	**.033***
	2	30	70			
Need for speech intervention	1	20	80		-2.236	**.025***
	2	70	30			

Abbreviations: WNL, within normal limits; CSCs, cleft speech characteristics; VPC-sum, velopharyngeal composite score; VPF, velopharyngeal function

*Wilcoxon paired-signed rank test, $p < .05$

Table 5. Descriptive statistics of the perceptually rated speech parameters for the children without CP±L (*n* = 10) at test date 1 (mean age 5y) and test date 2 (mean age 10y).

Perceptually rated speech parameter	***Test date***	**Scale** (%)					***Z***	***p***
		WNL	*Mild*	*Moderate*	*Severe*			
Speech understandability	1	70	30	0	0		-1.732	.083
	2	100	0	0	0			
Speech acceptability	1	100	0	0	0		.000	1.000
	2	100	0	0	0			
		Absent	*Borderline*	*Mild*	*Moderate*	*Severe*		
Hypernasality	1	90	10	0	0	0	-1.000	.317
	2	100	0	0				
		Absent	*Mild*	*Marked*				
Hyponasality	1	90	10	0			-1.000	.317
	2	100	0	0				
		Absent	*Occasional*	*Frequent*				
Audible nasal emission	1	100	0	0			.000	1.000
	2	100	0	0				
Nasal turbulence	1	100	0	0			.000	1.000
	2	100	0	0				
		Absent	*Present*					
Grimace	1	100	0				.000	1.000
	2	100	0					
		Normal	*Distinctive or abnormal voice quality*					
Voice	1	90	10				-.557	.577
	2	70	30					
		No consonants affected	*1-2 consonants affected*	*≥3 consonants affected*				
Anterior CSCs	1	50	50	0			-.447	.655
	2	40	50	10				

Posterior CSCs	1	100	0	0	.000	1.000
	2	100	0	0		
Non-oral CSCs	1	100	0	0	.000	1.000
	2	100	0	0		
Passive CSCs	1	80	20	0	-1.414	.157
	2	100	0	0		
		Sufficient VPF	*Borderline deficit*	*Insufficient VPF*		
VPC-sum	1	100	0	0	.000	1.000
	2	100	0	0		
		Absent	*Present*			
Non-cleft speech errors	1	50	50		-1.732	.083
	2	90	10			
Need for speech intervention	1	100	0		.000	1.000
	2	100	0			

Abbreviations: WNL, within normal limits; CSCs, cleft speech characteristics; VPC-sum, velopharyngeal composite score; VPF, velopharyngeal function

*Wilcoxon paired-signed rank test, $p < .05$

received speech therapy intervention between T1 and T2. At T2, one of them was still referred for additional speech therapy. To verify if the significant improvements between T1 and T2 in the group with CP±L were related to speech therapy, analyses were rerun without the inclusion of the two children who were enrolled in speech therapy between T1 and T2. Significant improvements were still found for the variables 'speech understandability' (p = .020), 'speech acceptability' (p = .026), 'non-oral CSCs' (p = .034), and 'non-cleft speech errors' (p = .023). In the control group, no significant improvements were found between T1 and T2 (all p's > .05, all Z's < .00, Table 5).

Significant differences were found for the group with CP±L compared to the control group at T1 as well as at T2 for the variables 'speech understandability', 'speech acceptability', 'hypernasality', 'passive CSCs', 'developmental speech errors' and 'VPC-sum' (Table 6). Children with CP±L, as a group, were judged to be less understandable, had less acceptable speech, showed more resonance and articulation errors and had more perceived velopharyngeal insufficiency. The results for 'presence of nasal turbulence' only significantly differed at T2. Significantly more 'non-oral CSCs' were observed in the group with CP±L compared to the control group at T1 but not at T2, indicating that the children with CP±L as a group caught up when they became older. Even when the two children who received speech therapy between T1 and T2 were excluded from the analyses, no significant results were found between the children with and without CP±L at T2 for this variable (non-oral CSC's: p = .264). This is also represented in the number of children with CP±L that were in need for speech therapy. At T1, significantly more children with CP±L were in need for speech therapy compared to the control group, whereas at T2 no significant group difference could be found anymore.

Table 7 provides more detailed information about the observed CSCs per child with CP±L at T1 and T2. In 8/9 children who showed compensatory CSCs at T1, less compensatory CSCs were observed at T2. Due to the small sample size, it is difficult to draw strong conclusions. However, in 3/4 children lateral or palatal articulation was not present anymore at T2, although none of these children had ever followed speech therapy. Information regarding satisfaction with speech at T2 was available for eight children based on caregiver report. At T2, 87.5% (7/8) were satisfied with the speech of their child.

4. Discussion

This study aimed to provide longitudinal speech outcomes in Ugandan children with CP±L who received palatal closure prior to the age of 6 months. Speech outcome variables were collected in 10 Ugandan children with CP±L at a mean age of 5 and 10 years old and compared with those of an age- and gender-matched control group of children without CP±L. Speech understandability and speech acceptability significantly improved over time in the group with CP±L, but not in the control group.

Table 6. Comparison of the outcome parameters of the children with and without CP±L at the age of 5 and at the age of 10 years old.

	Test date 1 – Children with vs. without CP±L		**Test date 2 – Children with vs. without CP±L**	
	Z	***p***	**Z**	***p***
Speech intelligibility	3.891	**<.001***	2.801	**.005***
Speech acceptability	4.051	**<.001***	3.111	**.002***
Hypernasality	2.490	**.013***	3.114	**.002***
Hyponasality	1.000	.317	.000	1.000
Audible nasal emission	1.000	3.17	1.831	.067
Nasal turbulence	1.451	.147	2.164	**.030***
CSCs anterior	.626	.531	1.011	.312
CSCs posterior	1.831	.067	1.000	.317
CSCs non-oral	3.127	**.002***	1.451	.147
CSCs passive	1.985	**.047***	3.139	**.002***
Non-cleft speech errors	2.644	**.008***	2.669	**.008***
VPC-sum	3.418	**.001***	2.500	**.012***
Need for speech therapy	3.559	**.002***	1.831	.067

Abbreviations: CSCs, cleft speech characteristics; VPC-sum, velopharyngeal composite score

*Mann-Whitney U test, $p < .05$

Table 7. Detailed comparison of cleft speech characteristics (CSCs) in the group with CP±L at 5 and 10 years old.

Child	**CSCs on T1**	**CSCs on T2**	**Speech therapy between T1 and T2**	**Improvement compensatory articulation?**	**Satisfied with speech at T2?**
1	Dentalization/interdentalization: /t/, /d/, /l/, /s/ Glottal reinforcement: /t/, /k/ Active nasal fricative: /s/	Weak/nasalized consonants: /p/, /b/	no	yes	yes
2	None	Dentalization/interdentalization: /l/, /t/	no	NA	NA
3	Palatal articulation: /s/ Weak/nasalized consonants: /b/, /f/	Dentalization/interdentalization: /s/, /t/ Weak/nasalized consonants: /b/, /v/	no	yes	yes
4	Dentalization/interdentalization: /t/, /d/, /l/ Palatal articulation: /s/, /z/, /n/ Backing to velar/uvular: /d/ Glottal articulation: /t/ Weak/nasalized consonants: /b/	Dentalization/interdentalization: /n/, /z/ Weak/nasalized consonants: /p/, /t/	no	yes	yes
5	Backing to velar/uvular: /n/ Glottal articulation: /t/, /k/ Weak/nasalized consonants: /p/, /v/ Gliding: /g/	Dentalization/interdentalization: /l/ Weak/nasalized consonants: /p/, /t/	no	yes	yes
6	Dentalization/interdentalization: /d/	Dentalization/interdentalization: /n/	no	yes	NA

	Glottal articulation: /k/, /g/ Glottal reinforcement: /t/ Weak/nasalized consonants: /p/, /b/, /f/, /v/ Nasal realization of plosives: /b/	Glottal reinforcement: /t/, /b/ Weak/nasalized consonants: /p/, /b/, /s/, /z/			
7	Lateral articulation: /s/, /z/ Palatal articulation: /t/, /d/ Backing to velar/uvular: /n/, /t/ Glottal articulation: /k/ Weak/nasalized consonants: /d/	Dentalization/interdentalization: /d/, /n/ Backing to velar/uvular: /t/ Weak/nasalized consonants: /p/	no	yes	yes
8	Glottal articulation: /p/, /b/, /t/, /d/, /k/, /g/, /f/ Glottal reinforcement: /t/, /d/, /f/, /v/, /s/, /z/ Weak/nasalized consonants: /p/, /s/, /b/ Nasal realization of plosives: /b/, /g/	Glottal articulation: /p/, /k/ Glottal reinforcement: /t/, /s/, /z/ Weak/nasalized consonants: /b/, /d/ Nasal realization of plosives: /b/	yes	yes	no
9	Active nasal fricative: /s/, /z/	Dentalization/interdentalization: /l/, /s/, /z/	yes	yes	yes
10	Lateral articulation: /s/, /z/	Lateral articulation: /s/, /z/ Palatal articulation: /t/	no	no	yes

More specifically, all children without CP±L reached acceptable speech before the age of 5 years in contrast to children with CP±L.

The improvements in speech understandability and speech acceptability in children with CP±L may be related to the improvements found for the variables related to compensatory articulation. A significant decrease in non-oral articulation errors was found at T2 in the group with CP±L. Additionally, no more significant differences were found at T2 for the variable 'non-oral CSCs' between the group with and without CP±L. Even when the two children with CP±L who received speech therapy between T1 and T2 were removed from the analysis, this result remained non-significant. This indicates that compensatory articulation improved over time in some children even without speech therapy intervention. It was unexpected that the presence of non-oral CSCs significantly decreased in children who were not enrolled in speech therapy. No other reports are available about the improvement of compensatory articulation over time without speech therapy and we do not have an explanation for this finding. At T2, 30% of the children with CP±L was still in need of speech therapy to remediate compensatory articulation errors. This proves that time alone is often not enough to eliminate cleft speech characteristics.

Our results are comparable with the results reported by Nyberg et al. (2014) who also found a decrease in compensatory articulation in children with CP±L between the age of 5 and 10 years old who received their palatal closure at 13 months. At the age of 5 years, 26% showed posterior CSCs and 22% showed non-oral CSCs; at the age of 10 years this was reduced to 5% and 2% respectively. A difference with the current study is that 61% of the children in the study by Nyberg et al. (2014) was enrolled in speech therapy compared to 20% in the current study. A side note here is that all children who were considered in need of speech therapy in the study by Nyberg received therapy, in contrast to only 2/8 children in the current study. Moreover, 43% of the children in the study by Nyberg et al. (2014) received pharyngeal flap surgery between T1 and T2, resulting in a decrease of hypernasality, audible nasal airflow and the presence of weak pressure consonants. In the current study, none of the children with CP±L received speech improving surgery between T1 and T2. As a result, no significant improvements were found between T1 and T2 for variables related to hypernasality, audible nasal airflow and passive CSCs. In contrast, more children presented with hypernasality and audible nasal airflow at T2, although this decline was not statistically significant. Moreover, velopharyngeal insufficiency and passive CSCs were still present in some children at the age of 10, suggesting the need for additional speech improving surgery. Three of the four children who received palatal re-repair before T1 (including the child who received additional buccal flap surgery) still showed mild to moderate hypernasality, audible nasal airflow and passive CSCs suggesting the presence of velopharyngeal insufficiency despite additional secondary surgery. It is unclear what caused this high

occurrence of passive errors. Progressive reduction of the adenoid sagittal thickness and growth of the nasopharynx has been reported in children without cleft palate between 4 and 13 years old (Park et al., 2016; Vilella et al., 2006). As a result, velopharyngeal closure may become more challenging for children with CP±L when they become older. This could have contributed to the increase in hypernasality and audible nasal airflow. Moreover, a high prevalence of fistulas was noted which may be related to the high presence of audible nasal airflow (Karling et al., 1993).

The high prevalence of fistulas (60%) and early palatal re-repair due to the presence of dehiscence after primary palatal closure (30%) may suggest that primary palatal closure may have been complex. A study by Katusabe et al. (2018), including 54 children with cleft palate that received their primary palatal closure at CoRSU hospital in Uganda, showed that malnutrition is one of the influencing factors in wound healing and fistula formation. Thirty-six percent of the infants who received palatal closure prior to 6 months suffered from a fistula within 6 months post-surgery in the study by Katusabe et al. (2018). Although age at palatal closure was not related to fistula formation, underweight in these very young children is often an important issue. Since 2005, all children with CP±L who present at CoRSU hospital undergo a one-stage cleft lip and palate repair if they are deemed fit for anesthesia (Hodges, 2010). This resulted in the inclusion of children whose palate was closed at a very young age in the current study (40% had an age < 3 months old at the time of surgery). There are two main reasons to follow this procedure at CoRSU hospital. First, closure of the palate is done to optimize the feeding process in these children. The inability to breastfeed adequately, including problems with suckling due to the oral-nasal coupling, has been described as a great predictor for malnutrition in children with CP±L before palatal closure (Tungotyo et al., 2017). Indeed, a study by Tungotyo et al. (2017) at CoRSU hospital showed that 68.2% of the 44 included children with CP±L between 0 and 10 months (median age 1 months, IQR 1-3m) who presented at CoRSU hospital were malnourished, of which 57% had moderate-to-severe malnutrition. These children are referred to the nutrition unit of the hospital. However, some children with malnutrition already receive closure of the cleft lip and palate because caregivers are often not able to afford feeding supplements (Katusabe et al., 2018). Second, the driving factor for parents to present their infants for surgery is the appearance of the cleft lip (Hodges, 2010). If the most common surgical procedure to close a cleft lip and palate would be followed (i.e., closure of the lip at 3 months and closure of the palate at 12 months, Shaw et al. (2019)), it was expected that many children would not return for the palatal closure. This can be related to the difficulty to travel (long distances) given the economic pressure on families in Uganda. For example, in the study by (Hodges, 2010), of the ten children who underwent only anterior vomer flap and cleft lip repair due to the width of their cleft palate, only one (10%) returned for the palatal repair. Although it has not yet been demonstrated that surgery in underweight children with cleft palate actually

improves their nutrition status and malnutrition has a significant impact on fistula formation (Katusabe et al., 2018), CoRSU hospital did not have the resources to monitor children who present from across the country to improve their nutrition prior to surgery. Therefore, the surgeons chose to operate earlier (Hodges, 2010).

Unfortunately, no information was available about the presence of malnutrition for the children included in the current study. Additionally, cleft width and tissue deficiency were also found to be influencing variables in fistula formation (Katusabe et al., 2018). When a cleft is too wide to close in a single stage, a vomer flap is used in first stage and soft palate repair is performed 3 months later in a second stage at CoRSU hospital. As a two-stage palatal closure was one of the exclusion criteria in the current study, children with very wide clefts were probably all excluded in the current study. However, no information about cleft width was available for the children included in this study. Although Katusabe et al. (2018) reported no relation between timing of palatal closure and fistula development, Eliason et al. (2020) found a significant increase in the rate of fistula formation in 108 children who underwent palatal closure prior to 6 months of age (40.9%) compared to 108 children who underwent palatal closure after 6 months of age (20.0%). Moreover, 50% of the 60 children whose palate was closed at the age of 0 to 3.9 months developed a fistula. Child gender, cleft type and associate syndrome were not significantly associated. However, surgeries were performed by different surgeons with a variety of training backgrounds using different techniques in both groups. No more information about these influencing variables was provided.

Analyzing complications related to very early palatal closure was not the aim of the current study. Based on the limited study sample and lack of anatomical and medical information before palatal closure, no conclusions can be made. However, it seems necessary to take these possible influencing factors into account when thinking about applying very early palatal closure in clinical practice or research. Whether malnutrition, decreased wound healing and additional surgeries have an impact on velopharyngeal functioning and speech is also question for further research.

Whether a one-stage palatal closure prior to the age of 6 months is more favorable for speech outcomes compared to one-stage palatal closure at 12 months is still not clear. At a mean age of 5 years, posterior and non-oral compensatory articulation errors were present in 70% of the included children with CP±L. Comparable with the results reported by Luyten et al. (2013), significant differences were found for the presence of compensatory articulation between the group with and without CP±L at this age in the current study. This is in contrast with the hypothesis that palatal closure before the onset of canonical babbling should reduce the anchoring of wrong patterns in the child's developing phonological system and, as a result, should minimize the occurrence of a compensatory

articulation pattern (Dorf & Curtin, 1982). However, compensatory articulation may result from a complex interplay of several influencing factors, of which timing of palatal closure may be one, but also anatomy and physiology of the velopharyngeal area, hearing, speech input and other unknown variables need to be considered (Lancaster et al., 2020; Lieberman et al., 2019; Lohmander et al., 2021). Although most children catch up over time, even without being enrolled in speech therapy, the presence of compensatory articulation and a decreased speech understandability and acceptability during childhood may have an impact on the child's psychosocial functioning (Murray et al., 2010). Therefore, speech therapy is still of utmost importance although providing speech therapy in resource-limited countries remains a challenge. It is difficult to draw conclusions from this first study that investigated the evolution of speech after early palatal closure in depth. A comparison with the longitudinal results in children who received palatal closure after the age of 6 months in a larger study sample is needed before any strong conclusions can be made. Additionally, prospective studies including the analysis of babbling and consonant inventory in young children who received very early palatal closure and follow-up during speech development would be interesting.

This study was conducted in a unique setting where, due to a limited availability of health care professionals, participants could not be treated by a cleft team. Several attempts are made at CoRSU hospital to provide the best possible care to children with CP±L, including high surgical and anesthesia standards, which are required to perform save interventions in such young babies (Hodges, 2010). However, children are only treated by a single surgeon and an SLP and/or nutritionist at times. No ENT specialist, nor audiologist is yet available. As a result, the hearing of the children cannot be evaluated and hearing problems are often not treated. This could have had a negative influence on the reported speech outcomes in the current study. Another possible influencing factor is the use of the children's second language, English, during the speech assessments. Despite excluding non-native consonants from the articulation analysis and including a control group with the same cultural and language background, possible influence on the results could not be fully excluded. Another limitation related to the context is the restricted response rate. Only 29% from the eligible children with CP±L participated in the current study. Other studies in countries with limited healthcare access also mentioned this limitation (Balasubramaniyan et al., 2017; de Buys Roessingh et al., 2012), which can be related to the difficulty to travel (long distances) for follow-up given the economic pressure on families. As a result, especially those children with CP±L and their caregivers who still perceived difficulties with speech or post-operative complications (e.g., fistula) may have shown up. As mentioned above, 60% of the included children received additional surgery (i.e., fistula repair, palatal re-repair, buccal flap). These children were already enrolled in a follow-up trajectory which could have lowered the threshold to participate in the present study and/or could have resulted in the inclusion

of those children with a worse outcome. Due to large variability and the mentioned influencing factors, caution is required in generalizing these results to the entire Ugandan population with CP±L.

Overall, good to excellent intra- and interrater reliability agreement was found for the different perceptual speech variables. For some variables, poor to fair intra- or inter-rater reliability was found based on ICCs. When interpreting ICCs, the heterogeneity of the results has to be taken into account as a homogeneous group can induce lower ICC values compared to a more heterogeneous group despite similar levels of agreement (Costa-Santos et al., 2011; Hallgren, 2012). Therefore, percentages of absolute agreement were calculated to complement the ICCs. However, low reliability was found for the variables 'anterior oral CSCs' and 'non-cleft speech errors'. Other authors also reported low reliability results for these parameters (Bruneel et al., 2020; Chapman et al., 2016; Sell et al., 2009). Most anterior CSCs included dental/interdental articulation. It has been mentioned that the distinction between dental/interdental articulation as an anterior oral CSC or a developmental error in children with CP±L is difficult (Sell et al., 2009). Other reasons for low reliability of these variables that are mentioned in the literature are limited training and overlap between some cleft and non-cleft speech immaturities/errors (Sell et al., 2009). A different interpretation by both raters may explain the low inter-rater reliability of these variables. In future research, a training session including consensus listening may result in higher agreement for these variables.

5. Conclusion

Although the speech of Ugandan children who were born with CP±L and received palatal closure prior to the age of 6 months generally improved over time, the effect of very early palatal closure on speech was below expectations. At the age of 5 years, 70% of the children with CP±L showed compensatory articulation errors affecting speech understandability and acceptability. Differences were still present compared to the speech of children without CP±L at the age of 10 years, especially regarding understandability and the presence of passive speech errors such as hypernasality. It is difficult to draw strong conclusions based on the small study sample and the complex interaction of several influencing factors. The limited response rate, complicated wound healing probably due to malnutrition, and limited access to speech therapy, speech improving surgery and hearing interventions need to be taken into account when interpreting the results of this study. These influencing variables should also be considered when thinking about applying very early palatal closure in clinical practice or research. The unique context of Uganda resulted in a new approach regarding timing of palatal closure with specific challenges. Whether palatal closure prior to the age of 6 months is transferable to other countries is subject for further research, including both longitudinal and prospective designs with larger samples.

Author statement

Title
Better speech outcomes after very early palatal repair? – A longitudinal case-control study in Ugandan children with cleft palate

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