Intensive speech therapy in Ugandan patients with cleft (lip and) palate: a pilot-study assessing long-term effectiveness

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Abstract

Background/aims: In resource-limited countries, traditional models for speech therapy delivery are not adequate to reach all patients in need. In those countries, intensive speech therapy might be a solution. Preliminary results of previous research demonstrated that intensive speech therapy can be effective in the short term for patients living in countries with limited access to speech therapy. Questions might arise whether or not intensive treatment results in long-term benefits for these patients. Hence, the present study investigated long-term effectiveness of intensive speech therapy provided to Ugandan patients born with a cleft palate with or without cleft lip (CP±L) in terms of different speech characteristics.

Methods: Five Ugandan patients with CP±L, who received intensive speech therapy in the past, were contacted to participate in this follow-up study. All patients agreed to participate. Perceptual and instrumental speech evaluations were performed identically to the assessments immediately before and after speech therapy, to allow for comparison. Additionally, the Cleft Evaluation Profile, investigating self-perceived satisfaction with cleft-related features was included to compare satisfaction before and after speech therapy.

*Result*s: Long-term improvement in percentage correct consonants was seen in four patients. Furthermore, after speech therapy, decreased presence of resonance disorders was observed in three of the included patients. Before speech therapy, all participants were dissatisfied with speech. Interestingly, after intensive speech therapy, satisfaction with speech was seen in every patient and this satisfaction remained in the long term.

Conclusion: In summary, speech improvements after speech therapy varied among the five patients. Nevertheless, present study provided encouraging results to further investigate effectiveness of intensive speech therapy in patients with CP±L.

Keywords: Cleft (lip and) palate, Intensive speech therapy, Uganda

1. Introduction

Clefts of the palate (with or without clefts of the lip) (CP±L) are frequently occurring birth defects and for Uganda, most recent incidence rates were estimated at 0.73 in 1000, resulting in the need for approximately 1800 cleft repairs per year [1]. Children born with CP±L often present with resonance (e.g. hypernasality) and nasal airflow (e.g. nasal emission) disorders [2]. Moreover, articulation errors can occur which are divided into passive and active cleft speech characteristics (CSCs) [3]. Passive CSCs are caused by abnormal structure, generally resulting in changes regarding the manner of articulation. Since the origin of passive CSCs is structural, surgical intervention is needed for correction [2]. Active CSCs include changes regarding the place of articulation [4] and are regarded as compensatory articulations to avoid the consequences of problems with oronasal coupling [5]. Given the impact of CSCs on speech intelligibility and/or speech acceptability, speech is considered one of the primary outcome measures of palatal repair [6]. However, despite advances in surgical management, children with CP±L often continue to demonstrate speech problems after palatal closure [7]. However, in Uganda, services for cleft care are scarce. Moreover, given long travel distances and other practical considerations (e.g. difficulties with public transportation), the traditional model for speech therapy delivery in more resourced countries (i.e. intervention with a frequency of 2 sessions per week and an intensity of approximately 21-30 minutes [8] is not adequate to reach all patients in need [9]. In this case, intensive speech therapy might possibly be a solution to eliminate active CSCs and thus, improve speech intelligibility and speech acceptability. Until now, research in the area of intensive speech therapy in developing countries mainly focused on the effectiveness of Community-Based Speech Therapy Models in Thailand [10-13] or speech summer camps in Mexico [14] (Table 1). In summary, these studies concluded that both Community-Based Speech Therapy Models and speech summer camps are reliable, efficient and cost-effective ways to provide speech therapy to patients with CP±L. In Uganda, Luyten, Bettens [9] were the first to assess short-term effectiveness of short, intensive speech therapy provided to patients with CP±L in terms of different speech characteristics (i.e. speech intelligibility, articulation, resonance and nasal airflow). Five patients received six hours of individual speech therapy in three to four days. Preliminary short-term results showed the effectiveness of short, intensive speech therapy for patients with CP±L in countries with limited access to speech therapy. However, one may wonder whether or not short, intensive speech therapy also has long-term benefits. Benjamas, Preeya [15] assessed the effectiveness of a speech camp and follow-up session in Thai children with CP±L. Intensive speech therapy (a four-day speech camp, including 18 hours of therapy) and a one-day long-term follow-up session (six months later) was provided to children between 3;6 – 13 years. A statistically significant decrease in articulation errors following both the speech camp and the follow-up session was found. However, it can be questioned whether the same long-term results would have been obtained without this follow-up session. The impact of intensive speech therapy delivery on articulation errors in Iranian patients with CP±L was studied by Derakhshandeh, Nikmaram [16]. Children (3-12 years) received 4 therapy sessions of 45 minutes per week during 10 weeks. Follow-up data were obtained 4 weeks after the end of the intervention phase. Results suggested that all of the patients were able to maintain the ability to articulate learned phonemes correctly after the 4-weeks break. However, to the best of our knowledge, only these two

Table 1. Overview of studies investigating the effectiveness of intensive speech summer camps and Community-Based Speech Therapy Models.

Authors	Participants	Methods	Results
Pamplona et al. (2005)	2 groups of children with CP: *1.* $n = 45$ children with repaired CP and CAD *2.* $n = 45$ age and gender matched children with repaired CP and CAD Age range: 3–10 years Context: Mexico City	Speech summer camp Group 1: - Speech summer camp for 3 weeks (4 h a day) - Content: camp activities concerning a specific topic Group 2: - Speech therapy during 12 months (1 h a day, twice per week) - Content: phonological principles and the whole-language model	Articulation: Both groups showed a significant decrease in the severity of the CAD after speech therapy Nasality: NA Satisfaction/Quality of Life: NA
Hanchanlert, Pramakhatay, Pradubwong, & Prathanee (2015)	Children with CLP ($n = 11$) Age range: 6–15 years Context: Thailand (Kosumphisai and Mueang Districts, Maha Sarakham)	Community-Based Speech Therapy Model SAs (i.e. one physiotherapist and one occupational therapist) and caregivers were trained by SLPs in a 3-years project Children were provided speech therapy - 3–4 times, 45 min by the SLP - 2 times, 30 min a month by the SAs (for 9 months) - 3–4 times, 20–30 min per week by the caregivers at home (for 9 months) - Content: The Manual of Speech Correction for Children with CP: Paraprofessionals and Caregivers[a] and Exercises for Articulation Correction[b]	Articulation: Significant decrease in number of articulation errors on both word and sentence levels after speech therapy Nasality: NA Satisfaction/Quality of Life: According to the SAs, two ($n = 2$) children showed an improvement in personality including social relationships and interactions
Makarabhirom, Prathanee, Suphawatjariyakul, & Yoodee (2015)	Children with CP ($n = 17$) Age range: 3; 4–14 years Context: Thailand (Chiang Rai and Phayao provinces)	Community-Based Speech Therapy Model SAs ($n = 8$) were trained by SLPs Children were provided speech therapy - by the SLP, accompanied by a SA and a caregiver of the child - 4-days intensive camp (6 h a day) - 5 follow-up camps (twice a month) - Content: hierarchical approach starting with nonsense syllables, words, phrases, sentences, reading and conversational levels. When a stable improvement of target sound was noticed at $> 90\%$, they would move up to higher level or other sounds.	Articulation: Significant improvement of compensatory misarticulations on both words and sentence levels after speech therapy Nasality: NA Satisfaction/Quality of Life: NA
Pumnum, Kum-ud, & Prathanee (2015)	Children with CLP ($n = 6$) Age range: 3; 6–15 years Context: Thailand (Borabue district)	*Hanchanlert, Pramakhatay, Pradubwong, & Prathanee (2015)*	Articulation: Significant decrease in articulation errors in three of the six children with CLP after speech therapy Nasality: NA Satisfaction/Quality of Life: NA
Sritacha, Pumnum, & Prathanee (2016)	Children with CLP ($n = 7$) Age range: 3–15 years Context: Thailand (Maha Sarakham province)	*Hanchanlert, Pramakhatay, Pradubwong, & Prathanee (2015)*	Articulation: Significant improvement in articulation in six of the seven children after speech therapy Nasality: NA Satisfaction/Quality of Life: NA

CAD: compensatory articulation disorders CP: cleft palate CLP: cleft lip and palate NA: not applicable SAs: speech assistants SLPs: speech-language pathologists.

[a] Dechongkit S, Prathanee B, Lorwatanapongsa P, Manochiopinig S, Makarabhirom K, Suphawatjariyakul R et al. Manual of speech and language therapy for children with cleft and palate. Khon Kaen: Klangnanawitaya Press; 2007. 35.

[b] Prathanee B. Exercises for articulation correction. KhonKaen: Department of Otorhinolaryngology, Faculty of Medicine, Khon Kaen University; 2010.

studies investigated long-term outcomes of intensive speech therapy in patients with CP±L, thus making it hard to draw overall conclusions.

Historically, outcome studies in patients with CP±L almost exclusively focused on the level of impairment [6]. However, according to the International Classification of Functioning, Disability and Health (ICF) presented by the World Health Organization (WHO) [17], outcome measures need to be broader, also including an individual's functional performance (i.e. activities) and the social consequences (i.e. participation). Nevertheless, almost no attention has been paid to psychosocial outcomes of speech therapy in patients with CP±L [6]. Luyten, Bettens [18] assessed parental satisfaction in Ugandan children with CP±L following synchronous lip and palatal repair. A survey based on the Cleft Evaluation Profile (CEP) was used to assess self-perceived satisfaction for individual features related to cleft care (i.e. speech, hearing, appearance of teeth, lip, nose and the face). Results showed that satisfaction varied: parents of children with CP±L were most satisfied with hearing and appearance of the lip and nose. For speech and appearance of the teeth, parents were moderately satisfied. Unfortunately, long-term data were not applicable and possible changes in this satisfaction following speech therapy were not determined. To the best of our knowledge, no study yet included any valid outcome measures (e.g. the CEP) to detect psychosocial changes before and after speech therapy.

Given the limited literature, the present study investigated long-term effectiveness of intensive speech therapy in Ugandan patients with CP±L. It was hypothesized that intensive speech therapy would decrease the presence of active articulation errors and increase self-perceived satisfaction with cleft-related features on the long term. Since treatment focused on correct production of consonants with attention to correct direction of airflow, it was hypothesized that indirectly, the presence of resonance disorders would decrease in the long term.

2. Methods

This study was approved by the Mildmay Uganda Research Ethics Committee (0611-2017) and the Uganda National Council for Science and Technology (HS 2448). All participants and their legal caregivers were informed about the study, both orally and by letter. In case of a language barrier, this information was translated to the local language (e.g. Luganda) by one of the health professionals of the CoRSU hospital (i.e. speech-language pathologists (SLPs), nurses or social workers). Informed consent was signed by the caregivers.

2.1. Design

A longitudinal cohort study was used, resulting in the acquisition of four data points (Figure 1).

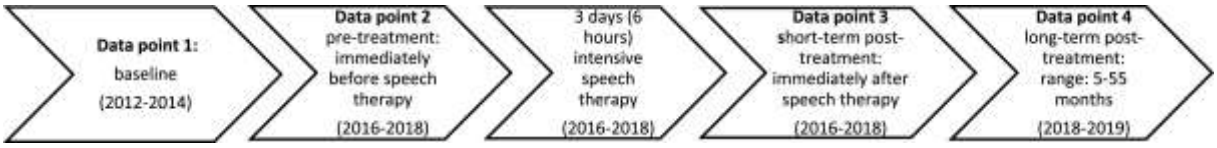

Fig. 1. longitudinal cohort study.Data was collected between March 2012 and March 2019.

Table 2. Demographic, cleft, and surgical details for the five patients.

	Patient 1	Patient 2	Patient 3	Patient 4	Patient 5
Demographic details					
Gender	Female	Female	Male	Male	Male
Age[a]	22; 1 years	11; 5 years	11; 4 years	11; 7 years	39; 10 years
Age speech therapy[b]	18; 0 years	8; 0 years	10; 4 years	11; 3 years	39; 6 years
Grade-level	Grade 6 at primary school	Grade 4 at primary school	Grade 4 at primary school	Grade 3 at primary school	NA
Employment mother	Cleaning lady	None	Teacher	NA	NA
Employment father	Driver	Graphic designer	NA	NA	NA
Use of English outside school	Yes, with friends	Yes, with friends	Yes, with friends	Yes, with friends	Yes
Employment	NA	NA	NA	NA	Driver
Mother language	Rutooro	Kakwa	Luganda	Luganda	Luganda
Literacy	Literate	Literate	Literate	Literate	Literate
Cleft and surgical details					
Cleft type	CP	UCLP (left)	UCLP (left)	Paramedian cleft	UCLP (left)
Age at lip closure	NA	2 months	3 months	5 months	22 years
Age at palatal closure	6 months	2 months	3 months	5 months	22 years
Palatal closure at CoRSU (performed by dr. A. H.)	No	Yes	Yes	Yes	No
Type of closure[c]	Early, one-stage closure	Early, one-stage closure	Early, one-stage closure	Early, one-stage closure	Late, one-stage closure
Secondary surgery	Yes: fistula repair (13 years), buccal flap (14 years)	Yes: lip re-repair (4 years)	No	Yes: buccal flap (5 years)	Yes: cleft lip and palate re-repair (35 years)
Alveolar bone graft	No	No	Yes (11; 0 years)	Yes (11 years; 6 years)	No

NA: not applicable CP: cleft palate UCLP: unilateral cleft lip and palate.

[a] Age at the moment of data point 4 (long-term follow-up).

[b] Age at start speech therapy.

[c] Early closure: prior to the age of 6 months.

2.2. Participants

Ugandan patients with CP±L *(n*=5*)* who received intensive speech therapy at CoRSU hospital between November 2016 and November 2018 were contacted by phone by one of the SLPs of the CoRSU hospital to obtain long-term follow-up data. Inclusion criteria for these patients were (1) repaired cleft palate whether or not in combination with repaired cleft lip, (2) sufficient English proficiency, and (3) followed 6 hours of short, intensive speech therapy provided by a non-Ugandan SLP between 2016 and 2018. All five patients agreed to participate in the present study and presented at CoRSU hospital between October 2018 and March 2019 in order to obtain the follow-up data. None of the patients suffered from acute ear, nose and/or throat diseases on the day of testing. Demographic, cleft, and surgical details for the participating patients are presented in Table 2.

2.3. Speech therapy

Speech therapy was provided between November 2016 and November 2018 (i.e. between data points 2 and 3) in a clinical room at CoRSU hospital by one of the non-Ugandan Flemish SLPs. Since English is one of the official languages in Uganda and all included patients were familiar with it, speech therapy was provided in English. The SLPs had a professional English proficiency, although they were native speakers of Flemish. Each patient received six hours of speech therapy during three consecutive days. Every session lasted one hour. Each participant received individualized speech therapy, hence target consonants differed between patients. Target sounds were selected using the following principles: (1) the SLP first focused on the consonants that influenced speech understandability the most and (2) if several speech sounds affected speech understandability in the same way, consonants that are normally acquired first during speech development, were addressed first [9]. Individual motor-phonetic articulation therapy [21] was provided. Articulation errors were treated in a phoneme-by-phoneme basis [21]. Distinctive features of the target consonant were identified using visual, tactile and/or auditory cues. This motor-phonetic approach was supplemented with linguistic-phonological principles supporting the establishment of the contrast between oral and nasal resonance and nasal airflow in consonants (i.e. the distinctive feature approach) [22, 23]. A more extensive description of the content of the speech therapy can be found in X [double blind review].

2.4. Assessments

2.4.1. Speech sample

To allow for comparison, the same protocol of speech assessments as used immediately before and after speech therapy (i.e. data points 1, 2 and 3) (X) was applied for long-term data collection (i.e. data point 4). The speech sample consisted of the repetition of English sentences of the MacKay-Kummer Simplified Nasometric Assessment Procedures (SNAP) test [24]. This test contains 15 sentences (12 oral and 3 nasal phrases), which were modeled by the SLP. Furthermore, automatic speech was recorded including counting from 1 to 10 and from 60 to 70 and naming the days of the week. Additionally, the standardized picture-naming test (i.e. Photo Articulation Test – Third Edition (PAT-3) [25] was carried out. The PAT-3 [25] contains 72 English words including

all English consonants in all permissible syllable positions as well as in common consonant clusters. While cued by colored pictures, participants were asked to repeat the 72 high frequency words read out by the SLP. All speech samples were both audio- and video-recorded. For all audio-recordings, a unidirectional condenser microphone (Samson, CO1U) was used. The microphone was placed in front of the patient, 10 cm from his/her mouth. For video-recordings, a Sony HDR-UX1 camera with a high quality built-in microphone was used.

2.4.2. Perceptual assessments

All speech samples were assessed by two SLPs (X and X) following internationally accepted protocols and guidelines [26-28]. Both SLPs have experience with cleft-related speech and have joined missions of our research unit to Uganda. Perceptual assessment of speech was undertaken using the Cleft Audit Protocol for Speech – Augmented (CAPS-A) [26, 27] with some modifications. Since Sell, John [27] cautioned against reporting speech intelligibility as this parameter is difficult to assess in a reliable way and given that Whitehill [29] recommended rating intelligibility and acceptability separately, both speech understandability (i.e.: within normal limits, mild, moderate, severe) [28, 30] and speech acceptability (i.e.: within normal limits, mild, moderate, severe) [28, 30] were assessed. To assess these two parameters, the two raters listened once to the audio-recordings of the automatic speech sample and the SNAP test [26, 27]. For perceptual evaluation of resonance (i.e. hypernasality and hyponasality) and nasal airflow (i.e. audible nasal emission and nasal turbulence), definitions of the CAPS-A protocol [26, 27] were applied. Firstly, the two raters listened to the audio-recordings of the automatic speech sample and the SNAP test. Subsequently, the raters listened to the video-recordings of the same speech samples to reconsider the parameters resonance and nasal airflow. Moreover, consonant errors were described as cleft speech characteristics (*CSCs*) (i.e. anterior, posterior, non-oral and passive CSCs) [26, 27]. Every category was assigned a score [27]. An “A-score” was assigned if no consonants were affected, a “B-score” if less than or equal to two consonants were affected, and a “C-score” if three or more consonants were affected. Although the CAPS-A protocol reports CSCs by severity, this study took the additional approach of calculating percentages of occurrence of the different CSCs to be able to present more detailed speech outcomes before and after intensive speech therapy [16]. Based on the ratings on the CAPS-A ordinal scales of the variables hypernasality, nasal emission, nasal turbulence, non-oral CSC’s and passive CSC’s, the Velopharyngeal composite score-summary CAPS-A (VPC-SUM) was determined. VPC-SUM scores were interpreted as follows: score 0-1: sufficient velopharyngeal function; score 2: borderline deficit and score 3-4: insufficient velopharyngeal function [31].

All video-recorded speech samples of the picture-naming test PAT-3 [25] and the 15 sentences of the SNAP test [24] were played back through over-ear headphones (Sennheiser EH150, Wedemark, Germany) and were phonetically transcribed by the two SLPs (X and X) using the International Phonetic Alphabet (IPA) [32] and the IPA extensions as well as additional symbols to describe specific cleft-related articulation errors [33]. Since Luyten, Bettens [9] reported that both Ugandans with and without clefts have difficulties with the correct pronunciation of the English fricatives /ɵ,ð,ʃ,ʒ/ and affricates /tʃ,dʒ/, these sounds were excluded from the analysis. Additionally, since the consonants /w/, /j/ and /r/ are not included as target consonants in the CAPS-A protocol [26, 27] these consonants were excluded in the assessment, resulting in 15 target consonants (/p/, /b/, /t/, /d/, /g/, /k/, /s/, /z/, /f/, /v/, /h/ /l/, /m/, /n/, and /ŋ/). In order to measure consonant proficiency, percentage

correct consonants (PCC), percentage correct places (PCP) and percentage correct manners (PCM) were calculated using guidelines described by [34]. All these speech samples were anonymized and randomized, hence the two raters were blinded to both the participants and the data points. For all parameters, patients' scores were determined by the score of the primary investigator (X), who did not provide speech therapy to any of the included patients and analyzed 100 % (20/20) of the speech samples, in order to avoid observer bias. The assessments of the second rater (X), who analyzed 60% (12/20) speech samples, were used to calculate inter-rater reliability. Furthermore, the first SLP (X) re-rated all speech samples again after 2 weeks in a different randomized order to determine intra-rater reliability. Both inter- and intra-rater reliability were calculated for all parameters by means of two-way random ICCs type consistency. For the inter-rater reliability both single and average ICC's measures were reported as a discrepancy was found between both values (Fleiss, 1979). ICC's were interpreted following the classification of Altman [35] (ICC < 0.20: poor, 0.21-0.40: fair, 0.41-0.60: moderate, 0.61-0.80: good, 0.81-1.00: very good).

2.4.3. Instrumental assessment

A KayPentax Nasometer (model II 6450) (NJ, Lincoln Park) was used to obtain objective nasalance values. At the beginning of each test session, the device was calibrated in a quiet room at CoRSU hospital following the instructions of the manufacturer's manual. Data were collected for the sentences of the SNAP test [24] and for the oral zoo passage and the oronasal rainbow passage [36]; The Nasality Severity Index 2.0 (NSI 2.0) was calculated using the nasalance scores for the vowel /u/ (%) and the oral zoo passage (%) as well as the voice low tone to high tone ratio (VLHR) of the vowel /i/ [37]. The VLHR was calculated by asking the patients to sustain the vowel /i/ for at least 2 seconds. This vowel was audio-recorded using PRAAT software version 5.4. [38]. The NSI 2.0 is a multiparametric index, calculated using a Praat script, with a positive value indicating the absence of hypernasality whereas a negative value indicates the presence of hypernasality [37].

2.4.4. Satisfaction with cleft-related features

The same questionnaire as utilized in previous missions of our research team to Uganda (X) was used to allow for comparison. Hence, the Cleft Evaluation Profile (CEP) [39] was applied to assess satisfaction with cleft-related features (i.e. (1) speech, (2) hearing, (3) appearance of the teeth, (4) appearance of the lip, (5) appearance of the nose and (6) appearance of the face). The original CEP uses a 7-point Likert scale (ranging from very satisfactory to very unsatisfactory) to rate satisfaction. Since [18] showed that some Ugandan patients were not familiar with a 7-point scale, this procedure was adapted and participants were asked if they were satisfied (i.e. happy) or dissatisfied (i.e. not happy). In other words, a 2-point scale (satisfied/not satisfied) was used. Questions were verbally asked in English, one of the official languages of Uganda, by a Dutch-speaking SLP. If needed, the questions were translated to the participants' mother language by one of the local health professionals (i.e. SLPs, nurses or social workers from the CoRSU hospital). If the patient was not able to answer the question him/herself, the questions were asked to the caregiver.

Table 3. Inter- and intra-rater reliability by means of a two-way mixed ICC (consistency).

	Inter-rater reliability						Intra-rater reliability		
	Single ICC consistency	95% CI single consistency	Inter-pretation of single ICC**	Average ICC consistency	95% CI average ICC consistency	Inter-pretation of average ICC**	Single ICC consistency	95% singles ICC consistency	Interpretation of the single ICC**
Speech understandability	0.684	0.237–0.989	good	0.938	0.684–0.998	very good	0.806	0.411–0.994	very good
Speech acceptability	0.649	0.220–0.987	good	0.937	0.693–0.998	very good	0.757	0.332–0.992	good
Hypernasality	0.879	0.583–0.997	very good	0.983	0.918–1.000	very good	0.670	0.241–0.988	moderate
Hyponasality	-*	–	–	–	–	–	–	–	–
Audible nasal emission	0.582	0.166–0.984	moderate	0.921	0.615–0.998	very good	0.820	0.420–0.955	very good
Nasal turbulence	0.457	0.069–0.974	moderate	0.871	0.372–0.997	very good	0.797	0.016–0.995	good
Anterior oral CSCs	0.366	0.019–0.965	fair	0.822	0.136–0.995	very good	0.429	0.052–0.972	moderate
Posterior oral CSCs	0.500	0.045–0.979	moderate	0.857	0.221–0.996	very good	0.597	0.171–0.984	moderate
Non-oral CSCs	0.457	0.069–0.974	moderate	0.871	0.372–0.997	very good	0.495	0.093–0.978	moderate
Passive CSCs	0.557	0.138–0.982	moderate	0.910	0.561–0.998	very good	0.593	0.168–0.984	moderate

CI: confidence interval, *reliability was not computed as there was no variance in the data set for this parameter, **based on Altman (1990): ICC $<$ 0.20: poor, 0.21–0.40: fair, 0.41–0.60: moderate, 0.61–0.80: good, 0.81–1.00: very good.

3. Results

3.1. Reliability of perceptual assessments

Results regarding inter- and intra-rater reliability are presented in Table 3. Inter-rater reliability, based on average measures ICC's, was very good for all parameters. Observing the single measures ICC's, good to very good ICC values were found for the parameters "speech understandability", "speech acceptability" and "hypernasality". However, fair to moderate ICC values were found for the parameters "nasal emission", "nasal turbulence", "anterior oral CSCs", "posterior oral CSCs", "non-oral CSCs" and "passive CSCs". Regarding intra-rater reliability, moderate ICC values were found for the parameters "hypernasality", "anterior oral CSCs", "posterior oral CSCs", "non-oral CSCs" and "passive CSCs". However, good to very good single ICC's were found for the parameters "speech acceptability", "nasal turbulence", "speech understandability" and "audible nasal emission".

Results for the perceptual and instrumental assessments and satisfaction with cleft-related features are respectively provided in Table 4 and 5. Table 6 provides a synthetic overview of the evolution of some of the most notable speech characteristics.

3.2. Patient 1

3.2.1. Speech outcomes

Prior to speech therapy (data points 1 and 2), patient 1's speech was characterized by severe hypernasality and occasional audible nasal emission. Speech understandability and speech acceptability were both mildly disturbed on data point 1 and respectively severely and moderately disturbed on data point 2. In accordance, increased nasalance values were observed for the bilabial, alveolar, velar and sibilant sentences and the oral and oronasal passages when compared to normative data [40]. Moreover, on both data points, a strongly negative NSI 2.0 value was obtained. On data point 1, PCC was 88.64% on the word level and 72.04% on the sentence level. PCP and PCM scores on this data point were respectively 92.04% and 92.60% on the word level and 79.49% and 94.62% on the sentence level. A trend towards lower PCC and PCP scores was seen on the word level on data point 2: PCC scores decreased from 88.64% (data point 1) to 78.40% (data point 2). Furthermore, PCP scores on the word level decreased from 92.04% (data point 1) to 78.40% (data point 2). Regarding the cleft type characteristics, (inter-)dentalization of the apico-alveolar consonants /s/ and /z/ occurred in 2.40% of the words on data point 1 and in 7.23% of the words on data point 2, thus indicating a B-score for anterior oral CSCs. Moreover, the fricative consonants /s/, /z/, /f/ and /v/ (i.e. C-score for non-oral CSCs) were mostly produced as active nasal fricatives (22.50% on data point 1, 72.50% on data point 2).

Speech therapy in this patient focused on the elimination of these active nasal fricatives, especially the correct production of the consonants /s/ and /z/ in isolation as well as in syllables, words and sentences, with special attention to the correct direction of airflow. Time between data point 1 (i.e. baseline measurement) and data point 2 (i.e. pre-treatment measurements) was 23 months. Post-treatment short-term data were obtained immediately after speech therapy. Time between data point 3 (i.e. short-term post-treatment measurements) and data point 4 (i.e. long-term post-treatment measurements) was 55 months.

Table 4. Results for the perceptual (including consonant proficiency) and instrumental assessments).

	Patient 1				Patient 1				Patient 1			
	Data point 1 (baseline)	Data point 2 (pre-treatment)	Data point 3 (post-treatment)	Data point 4 (long-term post-treatment)	Data point 1 (baseline)	Data point 2 (pre-treatment)	Data point 3 (post-treatment)	Data point 4 (long-term post-treatment)	Data point 1 (baseline)	Data point 2 (pre-treatment)	Data point 3 (post-treatment)	Data point 4 (long-term post-treatment)
Speech understandability	mild	moderate	mild	moderate	mild	mild	WNL	WNL	severe	severe	severe	severe
Speech acceptability	mild	moderate	mild	moderate	mild	mild	WNL	WNL	severe	severe	severe	severe
Hypernasality	severe	severe	moderate	moderate	normal	normal	normal	normal	severe	severe	severe	severe
Hyponasality	absent	absent	absent	absent	normal	normal	normal	normal	absent	absent	absent	absent
Audible nasal emission	occasional	occasional	occasional	occasional	absent	absent	absent	absent	absent	absent	absent	absent
Nasal turbulence	absent	occasional	occasional	occasional	absent	occasional	absent	absent	absent	absent	absent	absent
VPC-SUM	insufficient VPF	borderline deficit	borderline deficit	insufficient VPF	sufficient VPF	sufficient VPF	sufficient VPF	sufficient VPF	insufficient VPF	insufficient VPF	insufficient VPF	insufficient VPF
Consonant proficiency	**W S**	**W S**	**W S**	**W S**	**W S**	**W S**	**W S**	**W S**	**W S**	**W S**	**W S**	**W S**
Percentage correct consonants (PCC)	88.64 72.04	78.40 74.19	90.91 95.70	78.90 75.30	79.54 74.20	81.81 78.49	90.90 90.30	94.90 95.70	34.40 25.80	42.61 32.26	46.59 37.63	51.10 54.80
Percentage correct places (PCP)	92.04 78.49	78.40 70.97	92.61 96.77	77.80 73.12	77.84 76.34	75.57 78.49	74.50 79.50	80.10 84.90	42.04 31.11	43.18 33.33	52.77 35.58	56.80 54.80
Percentage correct manners (PCM)	92.60 94.62	93.75 93.55	92.61 94.62	95.50 97.80	97.72 98.82	93.55 100.00	92.60 91.40	94.90 95.60	86.36 88.17	94.89 88.17	92.04 83.87	89.80 95.70
Instrumental assessment												
Bilabial sentences (%)	69	69	53	60	22	18	8	8	45	70	60	57
Alveolar sentences (%)	62	70	67	75	33	19	13	8	31	73	64	54
Velar sentences (%)	56	69	52	74	28	17	12	10	52	70	67	63
Sibilant sentences (%)	67	78	68	81	63	79	20	17	48	79	74	65
Nasal sentences (%)	70	74	65	80	73	84	68	66	54	76	74	63
Rainbow passage (%)	69	72	72	76	45	54	34	33	NA	67	69	59
Zoo passage (%)	60	70	65	74	38	30	14	11	NA	71	71	58
VLHR/i/ 4,47*FOHz (dB)	23.46	24.40	22.11	22.64	17.91	19,97	12.50	21.50	NA	23.46	NA	27.03
NSI 2.0	−11.79	−14.78	−12.02	−15.06	−2.71	−2.21	5.05	2.50	NA	−14.55	NA	−13.28

	Patient 4				Patient 5			
	Data point 1 (baseline)	Data point 2 (pre-treatment)	Data point 3 (post-treatment)	Data point 4 (long-term post-treatment)	Data point 1 (baseline)	Data point 2 (pre-treatment)	Data point 3 (post-treatment)	Data point 4 (long-term post-treatment)
Speech understandability	Severe	Severe	Moderate	Moderate	Moderate	Moderate	Mild	Mild
Speech acceptability	Severe	Severe	Moderate	Moderate	Moderate	Moderate	Mild	Mild
Hypernasality	Severe	Severe	Severe	Severe	Moderate	Moderate	Mild	Mild
Hyponasality	Absent	Absent	Absent	Absent	Absent	Absent	Absent	Absent
Audible nasal emission	Occasional	Occasional	Occasional	Occasional	Absent	Absent	Absent	Absent
Nasal turbulence	Absent	Absent	Absent	Absent	Absent	Absent	Absent	Absent
VPC-SUM	Insufficient VPF	Insufficient VPF	Insufficient VPF	Insufficient VPF	Sufficient VPF	Sufficient VPF	Sufficient VPF	Sufficient VPF
Consonant proficiency	**W S**	**W S**	**W S**	**W S**	**W S**	**W S**	**W S**	**W S**
Percentage correct consonants (PCC)	35.40 42.92	38.10 43.61	73.86 48.40	77.27 68.80	64.15 63.04	62.15 65.67	82.95 83.97	80.11 75.26
Percentage correct places (PCP)	51.89 50.36	52.80 49.40	82.90 68.80	77.27 68.80	69.74 70.65	62.15 65.67	82.95 83.97	80.11 75.26
Percentage correct manners (PCM)	87.10 88.35	86.90 87.10	88.60 76.30	85.79 77.40	98.65 93.36	100.00 100.00	100.00 100.00	100.00 100.00
Instrumental assessment								
Bilabial sentences (%)	57	49	61	56	40	39	32	25
Alveolar sentences (%)	49	40	57	50	34	36	28	27
Velar sentences (%)	65	62	58	53	47	46	36	31
Sibilant sentences (%)	71	63	69	53	45	44	40	34

	Patient 4				Patient 5			
	Data point 1 (baseline)	Data point 2 (pre-treatment)	Data point 3 (post-treatment)	Data point 4 (long-term post-treatment)	Data point 1 (baseline)	Data point 2 (pre-treatment)	Data point 3 (post-treatment)	Data point 4 (long-term post-treatment)
Nasal sentences (%)	69	55	73	62	43	47	41	38
Rainbow passage (%)	56	55	65	57	35	34	29	24
Zoo passage (%)	60	59	60	53	42	39	35	25
VLHR/i/ 4,47*FOHz (dB)	24.47	25.31	19.31	27.19	15.04	14.72	14.68	27.61
NSI 2.0	−13.57	−13.05	−13.74	−11.86	−5.57	−5.12	−2.50	−2.78

WNL: within normal limits, VPF: velopharyngeal function, W: word level, S: sentence level, NA: not applicable.

Table 5. Results for the different features of the Cleft Evaluation Profile (CEP).

		Rater	Speech	Hearing	Appearance of the teeth	Appearance of the lip	Appearance of the nose	Appearance of the face
Patient 1	Data point 1 (baseline)	Mother	Satisfied	Satisfied	Satisfied	Satisfied	Satisfied	Satisfied
	Data point 2 (pre-treatment)	Patient	Dissatisfied	Satisfied	Satisfied	Satisfied	Satisfied	Satisfied
	Data point 3 (post-treatment)	Patient	Satisfied	Satisfied	Satisfied	Satisfied	Satisfied	Satisfied
	Data point 4 (long-term post-treatment)	Patient	Satisfied	Satisfied	Satisfied	Satisfied	Satisfied	Satisfied
Patient 2	Data point 1 (baseline)	Mother	Dissatisfied	Dissatisfied	Satisfied	Satisfied	Satisfied	Satisfied
	Data point 2 (pre-treatment)	Mother	Dissatisfied	Dissatisfied	Dissatisfied	Dissatisfied	Dissatisfied	Satisfied
	Data point 3 (post-treatment)	Mother	Satisfied	Dissatsified	Dissatisfied	Satisfied	Satisfied	Satisfied
	Data point 4 (long-term post-treatment)	Mother	Satisfied	Dissatisfied	Satisfied	Satisfied	Satisfied	Satisfied
Patient 3	Data point 1 (baseline)	Mother	Dissatisfied	Satisfied	Dissatisfied	Satisfied	Satisfied	Satisfied
	Data point 2 (pre-treatment)	Mother	Dissatisfied	Dissatisfied	Dissatisfied	Satisfied	Satisfied	Satisfied
	Data point 3 (post-treatment)	Mother	Satisfied	Satisfied	Dissatisfied	Satisfied	Satisfied	Satisfied
	Data point 4 (long-term post-treatment)	Mother	Satisfied	Dissatisfied	Satisfied	Satisfied	Satisfied	Satisfied
Patient 4	Data point 1 (baseline)	Grandmother	Dissatisfied	Satisfied	Dissatisfied	Satisfied	Dissatisfied	Dissatisfied
	Data point 2 (pre-treatment)	Grandmother	Dissatisfied	Satisfied	Dissatisfied	Satisfied	Dissatisfied	Dissatisfied
	Data point 3 (post-treatment)	Grandmother	Satisfied	Satisfied	Dissatisfied	Satisfied	Dissatisfied	Dissatisfied
	Data point 4 (long-term post-treatment)	Grandmother	Satisfied	Satisfied	Satisfied	Satisfied	Dissatisfied	Dissatisfied
Patient 5	Data point 1 (baseline)	Patient	Dissatisfied	Satisfied	Satisfied	Satisfied	Satisfied	Satisfied
	Data point 2 (pre-treatment)	Patient	Dissatisfied	Satisfied	Satisfied	Satisfied	Satisfied	Satisfied
	Data point 3 (post-treatment)	Patient	Satisfied	Satisfied	Satisfied	Satisfied	Satisfied	Satisfied
	Data point 4 (long-term post-treatment)	Patient	Satisfied**	Satisfied	Satisfied	Satisfied	Satisfied	Satisfied

*Rationale according to the patient: she goes to school and has many friends.
**Patient reported that he was satisfied with his speech, except for the sound/k/.

Table 6. Summary of the evolution of speech characteristics in the short and long term.

Speech characteristic	Patient 1		Patient 2		Patient 3		Patient 4		Patient 5	
	Short term (data point 3)	Long term (data point 4)	Short term (data point 3)	Long term (data point 4)	Short term (data point 3)	Long term (data point 4)	Short term (data point 3)	Long term (data point 4)	Short term (data point 3)	Long term (data point 4)
Understandability, acceptability and consonant proficiency										
Speech understandability and speech acceptability	↑	↓	↑	↑	=	=	↑	=	↑	=
Percentage correct consonants (PCC)	↑	↓	↑	↑	↑	↑	↑	↑	↑	=
Nasality										
Perceptual assessment of hypernasality	↑	=	=	=	=	=	=	=	↑	=
NSI 2.0	↑	↓	↑	= [a]	NA	= [a]	=	= [a]	↑	= [a]

↑ improvement compared to previous data point, ↓ deterioration compared to previous data point, = parameter is stable compared to previous data point.

[a] Value lying within 95% confidence interval: [NSI 2.0 ± 2.69] [36].

Immediately after speech therapy (data point 3), perceptual evaluation showed moderate hypernasality and mildly disturbed speech understandability and speech acceptability. A negative NSI 2.0 value was observed. However, a genuine decrease (i.e. ≥5% [41]) was noticed for the nasalance values of the bilabial, velar and sibilant sentences and the oral text. Furthermore, PCC scores increased from 78.40% (word level) and 74.19% (sentence level) on data point 2 to 90.91% (word level) and 95.70% (sentence level) on data point 3. In accordance, a decrease in active nasal fricatives (72.50% on data point 2 to 12.50% on data point 3) was observed.

On the long term (data point 4), patient 1 still presented with resonance disorders which was reflected by moderate hypernasality and a negative NSI 2.0 value. Furthermore, the parameters "speech understandability" and "speech acceptability" were assessed as moderately disturbed. Occurrence of active nasal fricatives increased from 12.50% (data point 3) to 70% (data point 4). This increase in occurrence of active nasal fricatives was also seen in the decrease of the PCC and PCP scores on data point 4. PCC and PCP scores on the word level respectively decreased from 90.91% and 92.61% (data point 3) to 78.90% and 77.80% (data point 4).

3.2.2. Satisfaction with cleft-related features

On data point 1, the mother of patient 1 answered the questions. She was satisfied with all cleft-related features. On data point 2, patient 1 answered the questions herself. Results showed that she was dissatisfied with speech and satisfied with all other cleft-related features. After speech therapy, patient 1 obtained satisfaction with speech and this remained in the long term.

3.3. Patient 2

3.3.1. Speech outcomes

Prior to speech therapy (data points 1 and 2), patient 2's speech understandability and acceptability were categorized as mildly disturbed. Despite the fact that the parameter "hypernasality" was perceptually assessed as "normal", increased nasalance values for the sentences of the SNAP-test were observed. Speech understandability was mostly influenced by the occurrence of active nasal fricatives (data point 1: 75.00%, data point 2: 67.50%, resulting in respectively a C- and B-score for the non-oral CSCs). Furthermore, inter-dental production of the apico-alveolar consonants (/s/, /z/, /t/, /d/, /n/ and /l/) was observed (data point 2: 14.45%). This resulted in PCC scores of 79.54% and 74.20% on data point 1 and 81.81% and 78.49% on data point 2, respectively on the word and sentence level.

Therapy was focusing on the elimination of active nasal fricatives (/s/ and /z/) and correct production of the other apico-alveolar consonants in isolation as well as in syllables, words, sentences, texts and transfer to spontaneous speech. The importance of oral airflow was particularly emphasized. Time between data point 1 (i.e. baseline measurement) and data point 2 (i.e. pre-treatment measurements) was 22 months. Post-treatment short-term data were obtained immediately after speech therapy. Time between data point 3 (i.e. short-term post-treatment measurements) and data point 4 (i.e. long-term post-treatment measurements) was 50 months.

Immediately after speech therapy (i.e. data point 3), no resonance disorders or airflow deviation errors were observed perceptually. Moreover, speech understandability and acceptability were assessed as normal. These

perceptual findings were confirmed by the objective nasalance values, which were within normal limits (Luyten et al., 2012), and the positive NSI 2.0 value. Regarding articulation, the occurrence of active nasal fricatives decreased from 67.50% (data point 2) to 0.00% (data point 3), resulting in an A-score for non-oral CSCs. This decrease in occurrence of active nasal fricatives resulted in higher PCC scores when comparing data points 2 and 3: PCC increased from 81.81% to 90.90% on the word level and from 78.49% to 90.30% on the sentence level. However, inter-dental production of the apico-alveolar consonants (/s/, /z/, /t/, /d/, /n/ and /l/) (i.e. a C-score for anterior CSCs) was still present which resulted in PCP scores of 74.50% (word level, data point 3) and 79.50% (sentence level, data point 3).

On the long term (i.e. data point 4), patient 2 presented with normal speech understandability and speech acceptability and no hypernasality or other resonance disorders, which was again reflected by normal objective nasalance values and a positive NSI 2.0 value. Despite the absence of active nasal fricatives (0.00%), patient 2 presented with inter-dental articulation of the apico-alveolar consonants /s/, /z/, /t/, /d/, /n/ and /l/ (38.50%), which was reflected in the PCP scores on this data point (word level: 80.10%, sentence level: 84.90%).

3.3.2. Satisfaction with cleft-related features

On all data points, the mother of patient 2 answered the questions. On data point 1, the mother reported that she was dissatisfied with speech and hearing. Before therapy (i.e. data point 2) the mother indicated that she was dissatisfied with speech, hearing, and the appearance of the teeth, lip and nose of patient 2. After speech therapy, the mother reported that she was satisfied with patient 2's speech and this satisfaction remained in the long term. Dissatisfaction regarding the feature "hearing" remained on both short term and long term.

3.4. Patient 3

3.4.1. Speech outcomes

Prior to speech therapy (i.e. data points 1 and 2) patient 3's speech was characterized by severe hypernasality. Both speech understandability and acceptability were severely disturbed. In accordance, increased nasalance values of the sentences of the SNAP-test and the oral and oronasal passages were observed. Regarding articulation, speech was mainly characterized by non-oral CSCs (resulting in a C-score): glottal articulation (data point 1: 32.74%, data point 2: 46.90%) and glottal reinforcement (data point 1: 15.04%, data point 2: 23.01%), affecting both PCC and PCP scores. For data point 1, PCC was 34.40% on the word level and 25.80% on the sentence level. PCP was 42.04% and 31.11% on respectively the word and sentence levels.

Intervention focused mainly on the elimination of glottal articulation and glottal reinforcements. A glottal stop/reinforcement was visualized using the picture of a lion. Time between data point 1 (i.e. baseline measurement) and data point 2 (i.e. pre-treatment measurements) was 44 months. Post-treatment short-term data were obtained immediately after speech therapy. Time between data point 3 (i.e. short-term post-treatment measurements) and data point 4 (i.e. long-term post-treatment measurements) was 10 months.

After speech therapy (i.e. data point 3), patient 3 still presented with severely impaired speech understandability and speech acceptability. In accordance, hypernasality was severely disturbed, reflected by the high nasalance values for the sentences of the SNAP-test and both the oral and oronasal passages. Regarding the non-oral CSCs,

occurrence of glottal articulation decreased from 46.90% to 34.74% and glottal reinforcement decreased from 23.01% to 19.47% after speech therapy. Furthermore, active nasal fricatives (5.00%) and pharyngeal articulation (4.42%) were observed immediately after speech therapy, resulting in a C-score for the non-oral CSCs.

Long-term results (i.e. data point 4) were in line with short-term findings. Presence of glottal (19.47%) and pharyngeal articulation (25.66%) was observed. Furthermore, the occurrence of glottal reinforcements decreased from 19.47% on data point 3 to 1.80% on data point 4.

3.4.2. Satisfaction with cleft-related features

On all data points, the mother of patient 3 answered the questions. On data point 1, the mother reported that she was dissatisfied with speech and appearance of the teeth. Before therapy (i.e. data point 2) the mother indicated that she was dissatisfied with speech, hearing and appearance of the teeth. After speech therapy the mother reported that she was satisfied with patient 3's speech and this satisfaction remained on the long term. Dissatisfaction regarding the feature "hearing" remained both on short term and long term.

3.5. Patient 4

3.5.1. Speech outcomes

Prior to speech therapy (i.e. data points 1 and 2) patient 4's speech was characterized by severe hypernasality. Both speech understandability and acceptability were severely disturbed. In accordance, increased nasalance values of the sentences of the SNAP-test and the oral and oronasal passages were observed. Concerning articulation, anterior CSCs (inter-dentalization of all apico-alveolar consonants), non-oral CSCs (glottal articulation and glottal reinforcement of the /k/, /t/ and /g/) and passive CSCs (passive nasal fricatives) were observed resulting in PCC scores of 35.40% and 42.92% (data point 1, word and sentence level respectively) and 38.10% and 43.61% (data point 2, word and sentence level respectively).

Intervention focused on the elimination of the anterior and non-oral CSCs (i.e. glottal articulation and glottal reinforcements of the /k/, /t/ and /g/ and inter-dentalization of all apico-alveolar consonants). Time between data point 1 (i.e. baseline measurement) and data point 2 (i.e. pre-treatment measurements) was 40 months. Short-term post-treatment data were obtained immediately after speech therapy. Time between data point 3 (i.e. short-term post-treatment measurements) and data point 4 (i.e. long-term post-treatment measurements) was 5 months.

After speech therapy (i.e. data point 3), patient 4 still presented with moderately impaired speech understandability and speech acceptability. Nevertheless, hypernasality was still severely disturbed, reflected by high nasalance values for the sentences of the SNAP-test and both the oral and oronasal passages. Considering articulation, PCC score increased from 38.10% to 73.86% on the word level. Moreover, glottal articulation and glottal reinforcement decreased from 45.78% and 38.13% before speech therapy to 15.36% and 20.78% after speech therapy, respectively. Long-term findings (i.e. data point 4) were in line with these short-term results. Moreover, PCC scores increased from 73.86% to 77.27% on the word level and from 48.40% to 68.80% on the sentence level.

3.5.2. Satisfaction with cleft-related features

On all data points, the grandmother of patient 4 answered the questions. Before speech therapy, she was dissatisfied with speech, appearance of the teeth, appearance of the nose and appearance of the face. After speech therapy, the grandmother became satisfied with patient 4's speech and this satisfaction remained in the long term. Nevertheless, she was still dissatisfied with the appearance of the nose and the face.

3.6. Patient 5

3.6.1. Speech outcomes

At data points 1 and 2, patient 5's speech was characterized by moderate hypernasality and moderately disturbed speech understandability and acceptability. Non-oral CSCs were observed in patient 5's speech. More specifically, pharyngeal articulation of /g/ and /k/ (data point 1: 25.67% and data point 2: 26.78%, both C-scores) and glottal reinforcement of /k/ and /t/ (data point 1: 10.18% and data point 2: 11.07%, both C-scores).

Intervention focused on elimination of the non-oral CSCs. Time between data point 1 (i.e. baseline measurement) and data point 2 (i.e. pre-treatment measurements) was 40 months. Short-term post-treatment data were obtained immediately after speech therapy. Time between data point 3 (i.e. short-term post-treatment measurements) and data point 4 (i.e. long-term post-treatment measurements) was 5 months.

After speech therapy (i.e. data point 3), patient 5 presented with mildly disturbed speech understandability and speech acceptability. In accordance, hypernasality was mildly disturbed. After intervention, PCC scores increased from 62.15% and 65.67% (data point 2, word and sentence level respectively) to 82.95% and 83.97% (data point 3, word and sentence level respectively). Long-term results (i.e. data point 4) were in line with short-term findings. Interestingly, after treatment, presence of pharyngeal articulation was observed merely for the /k/-sound in word initial positions (B-score).

3.6.2. Satisfaction with cleft-related features

Patient 5 answered all questions himself. Before therapy (i.e. data point 2), he was dissatisfied with speech and satisfied with all other cleft-related features. After treatment, the patient reported that he was satisfied with his speech. Interestingly, this satisfaction remained on the long term. Nevertheless, he highlighted that he was not completely satisfied with the production of the /k/.

4. Discussion

Literature regarding the effectiveness of speech therapy in patients with CP±L is scarce [6]. More specifically, there is a lack of studies investigating long-term effectiveness. Hence, present study investigated the impact of intensive speech therapy in Ugandan patients with CP±L. It was hypothesized that intensive speech therapy would decrease presence of active articulation errors and increase self-perceived satisfaction with cleft-related features in the long term. Since treatment focused on the correct production of consonants with attention to correct direction of airflow, it was hypothesized that indirectly, the presence of resonance disorders would decrease in the long term.

4.1. Speech understandability, acceptability and consonant proficiency

Short-term improvement in speech understandability and speech acceptability was found for patients 1, 2, 4 and 5 (Table 6). In patient 1, speech therapy focused on the elimination of active nasal fricatives. Hence, the increased speech understandability and speech acceptability are likely related to the successful decrease in the occurrence of active nasal fricatives, which was also evidenced by the increased PCC scores (Table 4 and 6). However, in the long term, occurrence of active nasal fricatives increased from 12.50% (short term, data point 3) to 70% (long term, data point 4). In other words, long-term findings for patient 1 showed the re-occurrence of speech disorders that required additional speech therapy, thus suggesting no long-term benefits of speech therapy in this patient. A possible explanation for these long-term findings was the relatively old age of patient 1 at the moment of speech therapy (i.e. 18 years) whereas patients with CP±L (especially in more developed countries) usually start speech therapy at the age of 2.5-3 years [42]. Different authors suggested an early start of speech intervention since therapy delay has different undesirable consequences including fixation of articulation errors, resistance to treatment, inappropriate psychological, mental and social effects on the child, and negative effects on learning [16, 33]. However, early speech intervention is not always evident in Uganda due to different practical and socio-economic considerations (e.g. limited access to cleft care, financial factors, prevailing taboo). Benjamas, Preeya [15] assessed the outcomes of an intensive speech camp and one follow-up therapy session (after six months) in children with CP±L and found a decrease in articulation errors both after the speech camp and the follow-up session. Moreover, findings showed that this was a cost-effective way to provide speech therapy to children with CP±L in Thailand. In light of these ideas, patient 1 might have benefited from an intensive follow-up therapy session. Provision of follow-up speech camps to boost articulation after intensive therapy might consequently be a solution for such patients. Practically, the CoRSU hospital provides a hostel for the patients and their relatives and thereby, overnight stays during rehabilitations are possible to bridge the long travel distances. Unlike patient 1, relatively stable PCC, PCP and PCM scores on the long term were seen in patients 2, 4 and 5 (Table 4). Interestingly, in patient 2, speech understandability and acceptability were assessed as normal after treatment (both on the short and long term) (Table 4). Moreover, after speech therapy, the fricative consonants /s/ and /z/ were no longer produced as active nasal fricatives. Possible factors contributing to these results were the fact that patient 2 (1) originally presented with a limited amount of affected consonants, (2) had a young age at the start of the speech therapy (<12 years) (3) had undergone early palatal closure (≤12 months) [18] and (4) had a very good proficiency in English [9]. In contrast to the encouraging results for patients

1, 2, 4 and 5, patient 3 still presented with severely impaired speech understandability and speech intelligibility after speech therapy (Table 4 and 6). Nevertheless, a decrease in glottal stops and glottal reinforcements was seen immediately after speech therapy, which was reflected by increased PCC scores (Table 6). Moreover, this trend continued in the long term, thus suggesting some long-term benefits of speech therapy in patient 3. The decrease in glottal articulation could possibly be explained by the shift that was seen for the consonants /s/ and /z/: glottal production of these fricatives was substituted by pharyngeal fricatives (4.42%) or active nasal fricatives (5.00%) after speech therapy. These substitutions may also explain the relatively stable PCC, PCP and PCM scores before and after speech therapy (Table 4 and 6). Just as the other included patients, patient 3 received only six hours of speech therapy, which is a limited amount of time compared to previous speech therapy studies (e.g. 60 hours of speech therapy [14], 18 hours of speech therapy [15]). Hence, not all affected consonants were treated. This limited therapy period, combined with the high amount of affected consonants before speech therapy might be an important explanation for the relatively poor results in this patient.

4.2. Nasality

Immediately after speech therapy, a better perceptual assessment of hypernasality was found in patients 1 and 5 and this finding remained stable in the long term (Table 4 and 6). However, still a strongly negative NSI 2.0 value was observed after speech therapy in these two patients (Table 4). In patient 1, a distinction between perceptual and instrumental assessments of hypernasality was seen (Table 4). Even though there was a better perceptual rating of hypernasality on data point 4 (i.e. moderately disturbed) compared to data points 1 and 2 (i.e. severely disturbed), the NSI 2.0 index on data point 4 had a lower value (-15.06) when compared to data points 1 (-11.79), 2 (-14.78). It should be mentioned that Bettens, Wuyts [43] reported that the interval of NSI 2.0 ± 2.68 for children defines the 95% confidence interval. Hence, if a new obtained NSI 2.0 value lies within this interval for a specific patient, the observed change is not considered to be a result of physiological changes. Moreover, despite the fact that the NSI 2.0 correlates significantly with perceived hypernasality [43], Watterson, Lewis [44] pointed out that nasalance scores may be increased in children with audible nasal airflow problems, thus causing inconsistency with raters' judgments. Given that on data point 4, audible nasal emission was frequently heard on pressure consonants in patient 1 (Table 4), this might be a plausible explanation for present findings. Present contradiction highlights the importance of combining perceptual and instrumental measurements in the assessment of nasality. In patient 2, positive NSI 2.0 values were found in the short and long term, thus indicating the absence of hypernasality after intensive speech therapy (Table 4 and 6). In patients 3 and 4, no evolution in perceptual assessment of hypernasality was found when comparing the different data points (Table 4 and 6). Moreover, strongly negative NSI 2.0 values were found for the different data points (Table 4), hence indicating the presence of severe hypernasality. Present finding for these patients were not in line with the hypothesis namely that the presence of resonance disorders would (indirectly) decrease after intensive speech therapy. Results for patients 3 and 4 seem reasonable taking into account the VPC-sum score for these participants, suggesting insufficient velopharyngeal function (Table 4).

4.3. Satisfaction with cleft-related features

All patients were satisfied with their speech after therapy and this satisfaction remained on the long term in every patient (Table 5). Interestingly, this finding suggests psychosocial advantages of intensive treatment for patients with CP±L.

Regarding satisfaction, the mother of patient 1 was satisfied with all cleft-related features on data point 1 (Table 5). This finding was in line with Luyten [18] who found overall high levels of satisfaction for all features of the CEP in patients with CP±L. However, on data point 2, the patient herself answered the questions and results showed that she was dissatisfied with speech (Table 4). This dissatisfaction with speech was most likely related to the high occurrence of active nasal fricatives (72.50%) before speech therapy, which was severely affecting patient 1's speech understandability and acceptability (Table 4). Interestingly, after speech therapy, patient 1 reported that she was satisfied with speech, which was probably associated with the decreased occurrence of active nasal fricatives. Despite the fact that the occurrence of nasal fricatives increased in the long term, patient 1 remained satisfied with her speech. A possible explanation for these findings is the fact that patient 1 reported that she was socially accepted in her community (Table 5). Since social acceptance is predominantly determined by speech [18], the reported satisfaction might not be very surprising. Another explanation may be the fact that these questions were asked by a Flemish colleague of the treating Flemish SLP. Despite the fact that the patient was told that answers would have no influence on the relationship between her and the SLPs, it is possible that the patient was reluctant to tell that she was not happy with her speech resulting in socially desirable answers. The mother of patient 2 reported dissatisfaction with hearing on all data points (Table 5). Since patients with CP±L often present with hearing difficulties [45], it is important to investigate this specific matter. Therefore, multidisciplinary teams (including an audiologist) are necessary in order to provide better cleft care in resource-limited countries in the future. Exchange of knowledge with local health professionals will be important in order to explore local practices and perceptions concerning problems with hearing in these patients. Possible presence of hearing difficulties might have biased results for patient 2. However, since patient 2 responded well on the provided therapy and given that varying results were found for the other parameters of the CEP (e.g. for appearance of the lip, dissatisfaction was reported on data point 2 while satisfaction was reported on data point 3) (Table 5), it is plausible that changes in the answer resulted, to some extent, from a language barrier. Moreover, today there still exists a taboo regarding clefts in the Ugandan society [18]. From our own experience, parents of children with CP±L find questions, especially related to satisfaction, funny and don't always take it too seriously, thus the reliability of the answers might be questioned. Moreover, the fact that parents sometimes made fun of these questions might possibly result from a defense mechanism since they are not familiar with such questions, thus making them feel uncomfortable. Regarding the feature "speech", the mother of patient 2 indicated that she was dissatisfied with this feature before speech therapy and satisfied with it after therapy. Satisfaction with speech remained in the long term and is most likely related to absence of production of nasal fricatives and absence of resonance disorders (Table 4 and 6). Despite the fact that there was a relatively high occurrence of inter-dental production of all apico-alveolar consonants after speech therapy, both speech understandability and acceptability were assessed as normal, possibly explaining the mothers' satisfaction with

speech. Despite the fact that articulation and resonance disorders were still present in the short and long term in patient 3, the mother indicated that she became satisfied with speech after therapy. Furthermore, satisfaction with this feature remained in the long term (Table 5). In general, it was previously suggested that respect and gratitude towards (foreign) health professionals might influence answers of the parents and patients in a positive way [18, 39]. Regarding appearance of the teeth, the mother indicated that she was dissatisfied with this feature on data points 1, 2 and 3. On data point 4, she reported satisfaction with this feature. This finding can possibly be explained by the fact that an alveolar bone graft was performed between data points 3 and 4. Interestingly, a shift from dissatisfaction to satisfaction with appearance of the teeth after performance of an alveolar bone graft was also seen in patient 4.

4.4. Strengths and limitations

To the best of our knowledge, this was the first study investigating long-term outcomes of intensive speech therapy in a resource-limited country. An extensive speech analysis, with good inter- and intra-rater reliability, was performed in accordance to internationally accepted definitions and protocols (e.g. definitions of the CAPS-A protocol [26, 27], consonant proficiency [34]). Moreover, this was the first study including psychosocial outcomes after speech therapy using the Cleft Evaluation Profile [39] to assess satisfaction with cleft-related features. In the future, the adapted version of the VELO questionnaire [19] can be used to compare health-related quality of life before and after speech therapy. However, as suggested by Bruneel [19], further adaptations of this questionnaire and a translation to Luganda will be necessary. A drawback of using standardized questionnaires is that possible perspectives of patients that are not included in the questionnaire, will be missed. Therefore, the use of a qualitative design (e.g. semi-structured interviews and/or focus group discussions) might be a good solution to investigate psychosocial outcomes after receiving intensive speech treatment. However, abovementioned studies used a self-report questionnaire. These standardized questionnaires assume that there is already a clear idea of what parents' experience and expect in relation to speech therapy. Thereby, possible perspectives and expectations of parents that are not included in the questionnaire will be missed, thus highlighting the importance of qualitative research on this specific matter.

The small sample size (*n*=5) prevented statistical testing so that stronger evidence for the improvements could not be obtained. Due to organizational reasons, time intervals between the several data points were not equal for all patients. Furthermore, time between data point 1 (baseline) and data point 2 (pre-treatment) was on average 29 months, possibly resulting in effects related to maturation. Unfortunately, the acquisition of more data points before speech therapy was not possible due to time-constrictions on site. In future research, a single subject experimental design can be used so that each patient can serve as his or her own control [46]. In addition, it should be noted that there was no information regarding the velopharyngeal closing pattern of the patients due to practical considerations (no imaging equipment such as naso-endoscopy or videofluoroscopy was available). For some patients (e.g. patient 1 and 3) insufficient velopharyngeal functioning might be an explanation for the limited progress during speech therapy, which was also suggested by the VPC-sum scores (Table 4). Nevertheless, this study provides important information since it was the first to use several data points to assess outcomes of speech therapy in patients with CP±L living in a resource-limited country. In the future,

different challenges will need to be faced. Firstly, in order to allow comparison, it will be necessary to collect normative PCC, PCP and PCM data in both Ugandan children with and without CP±L. Secondly, there is still no international consensus regarding the content of speech therapy in patients with CP±L. In the present study, speech therapy was provided using a combined phonetic-phonological approach [21-23]. However, it is known that active CSCs may initially occur as a consequence of CP±L, producing a phonetic articulation disorder. Over time, the error becomes part of the child's rule system, producing a phonological error [47]. Whether or not a phonological approach may have been more effective than a phonetic approach for some of the Ugandan patients, is subject for further research.

5. Conclusion

Present study investigated effectiveness of intensive speech therapy in Ugandan patients with CP±L. Short- and long-term improvement in percentage correct consonants was seen in four patients, thus supporting the hypothesis that intensive speech therapy would reduce presence of active articulation errors. Furthermore, intensive speech therapy indirectly decreased the presence of resonance disorders in some patients (*n*=5). Interestingly, all parents/patients were satisfied with their speech after intensive speech therapy and this satisfaction remained in the long term. In general, present study provided encouraging results to further investigate the effectiveness of intensive speech therapy in patients with CP±L. Further research investigating outcomes of intensive speech therapy in larger patient groups is necessary.

Funding

The first author was funded by a PhD grant of the Research Fund Flanders (1120919N).

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