Comparison of motor-phonetic versus phonetic-phonological speech therapy approaches in patients with a cleft (lip and) palate: a study in Uganda

Cassandra Alighieri^{a,*}, Kim Bettens^a, Laura Bruneel^a, Daniel Sseremba^a, Duncan Musasizi^b, Isaac Ojok^b, Kristiane Van Lierde^{a,c}

- ^a Department of Rehabilitation Sciences, Ghent University, Corneel Heymanslaan 10, 2P1, 9000, Gent, Belgium
- ^b Comprehensive Rehabilitation Services in Uganda (CoRSU), P.O. Box 46, Kisubi, Uganda
- ^c Faculty of Humanities, Department of Speech-Language Pathology and Audiology, University of Pretoria, Lynnwood Road Hillcrest, Pretoria, South Africa
- *Corresponding author. Corneel Heymanslaan 10, 2P1, 9000, Gent, Belgium. E-mail address: Cassandra.Alighieri@UGent.be (C. Alighieri).

Abstract

Introduction: At present, there is growing interest in combined phonetic-phonological approaches to treat active speech errors in children with a cleft (lip and) palate ($CP \pm L$). Unfortunately, evidence for these type of speech interventions in this population is lacking. Therefore, the present study investigated the effectiveness of speech intervention in Ugandan patients with $CP \pm L$. Moreover, a comparison was made between a motor-phonetic and a phonetic-phonological speech intervention.

Methods: Eight patients (median age: 11.26y) with an isolated CP \pm L were assigned into a group receiving motor-phonetic treatment (n = 4) or a group receiving combined phonetic-phonological treatment (n = 4). The participants received 6h of individual speech therapy. In both groups, perceptual and instrumental speech evaluations were performed to evaluate the patients' speech before and after the intervention.

Results: Speech therapy (irrespective of the used approach) was found to be effective in increasing consonant proficiency and in decreasing the occurrence of non-oral and passive CSCs. No statistically significant differences in outcome variables were found when comparing the two groups pre- and post-treatment. The descriptive results, however, revealed a larger increase in % correctly produced consonants, places and manners after the intervention in the group receiving a combined phonetic-phonological treatment compared to the group receiving a motor-phonetic treatment.

Conclusion: This study took a first step in providing evidence concerning the effectiveness of different speech therapy approaches in children with $CP \pm L$. The present study holds some important implications for clinical practice suggesting that an additional phonological approach may be beneficial for the patients with $CP \pm L$. Further research including randomized controlled trials with larger sample sizes is necessary to provide further evidence.

1. Introduction

Patients with a cleft of the palate with or without a cleft of the lip (CP±L) often present with speech sound disorders related to the structural anomalies in the oral, nasal and/or pharyngeal cavities. When the deviant anatomical structure causes distortion of the speech sounds, passive (i.e. obligatory) errors occur [1, 2]. Examples of passive speech errors are hypernasality, nasal emission and/or nasal turbulence, and weak or nasalized production of pressure consonants. Passive errors can only be eliminated through correction of the structure and thus, speech therapy is not indicated in most cases [1, 2]. Active (i.e. compensatory) errors on the other hand require speech intervention

since articulatory placement is incorrect (e.g. backing to a palatal or velar place of articulation, glottal stops, active nasal fricatives) [1, 2]. Various authors described active errors as phonetic disorders since the patient lacks the physical or motor ability to produce the target sound, resulting in an alteration of the place (and sometimes the manner) of articulation [1, 3]. Historically, these errors were eliminated by providing speech therapy using traditional articulatory (i.e. motor-phonetic) approaches [4]. Although there is no straightforward description of traditional articulation therapy, Van Riper [5] is considered to be the developer of this approach [4, 6]. Motor-phonetic approaches treat articulatory errors on a phoneme-by-phoneme basis, emphasizing phonetic placement and shaping techniques [5]. The goal of treatment is to correct articulatory placement and/or manner of production [1]. Since the early 1980s, description of articulatory processes in patients with CP±L moved from a phonetic orientation to a phonological orientation [7]. In other words, it is assumed that during childhood, the active (phonetic) speech disorders become gradually incorporated into the child's developing rule system of the language, producing a phonologically-based error [8]. Phonological disorders are linguistically-based, representing difficulties with organization and representation of the sound system of a language [9, 10]. Moreover, emphasis was put on higher processes of speech, including the speaker's knowledge and perception of the sound system and error patterns [7]. With the development of this mindset, linguistic-phonological approaches were gradually applied in the intervention of active speech errors in patients with CP±L [4, 11]. In this approach, different articulation errors are treated simultaneously targeting multiple errors by focusing on the phonological rules that are active in the child's rule system. The use of cognitivelinguistic activities is highlighted, including the development of sound contrasts and the provision of meaningful language contexts [8, 12]. The aim of phonologically-based treatment is cognitive reorganization of the rule-system of the language that the child speaks. To date, the effectiveness of motor-phonetic and linguistic-phonological approaches has been barely demonstrated. A systematic review by Bessell et al. [4] identified seven studies [3, 12-17] investigating effectiveness of motorphonetic approaches and three studies [16, 18, 19] investigating effectiveness of linguisticphonological approaches when treating children with CP±L. Unfortunately, a lack of evidence to support any approach was found since most studies were dated, used different, dubious outcome measures and lacked detailed description concerning treatment provider, setting, and included patients [4]. As a result, selection of particular approaches is often based on general opinions, clinical experience, or strategies described in papers [4]. In general, literature suggests that traditional motor-phonetic (i.e. articulation) treatment results in overall improvement of speech [3, 12-17]. Detailed description of the treatment approach often lacked and the specific nature of the change was seldomly reported [4]. Concerning the linguistic-phonological approach, Hodson et al. [20] investigated the effectiveness of a phonological-based remediation program in order to increase intelligibility in one child, aged 5 years, with repaired cleft palate. After 13 months of treatment (36 weekly 60-90 minute sessions and twice weekly 20 minute sessions + home program including 65 hours over 13 weeks), the participant showed improved articulatory production (i.e. decreased production of glottal stops and elimination of nasal additions/replacements). In accordance, Van Demark and Harding [16] showed that a 6-weeks treatment program consisting of four 1-hour sessions per day for 26 days, targeting multiple sounds at one time, resulted in a decreased percentage of errors immediately after therapy. To the best of our knowledge, only one study [12] compared the effectiveness of conventional motor-phonetic (i.e. articulation) treatment to linguisticphonological treatment. In this study, the time of speech therapy necessary for total elimination of active articulation disorders was investigated. Patients with CP±L aged 3 to 7 years were randomly assigned into a motor-phonetic speech therapy group (n=15) or a linguistic-phonological speech therapy group (n=14). Both groups received 1-hour treatment sessions twice weekly until normal articulation was achieved. The findings showed that the total time of speech intervention was

significantly reduced when providing a linguistic-phonological intervention. Unfortunately, no comparison of speech outcomes between the two groups was provided and study findings not were confirmed in either the same or other centers, making generalization difficult [4]. Recently, Derakhshandeh et al. [21] investigated the effectiveness of combined phonetic-phonological treatment on articulation errors in five children with CP±L and velopharyngeal insufficiency. Participants received 2.5 months of articulation therapy including four sessions per week with a duration of 45 minutes per session. Treatment resulted in decreased presence of non-oral cleft speech characteristics (CSCs) in all five patients. These findings suggested that combined phoneticphonological treatment in patients with CP±L and VPI can result in an improvement of oral placements and passive CSCs. In accordance, two studies [22, 23] investigated the short- and longterm effectiveness of an intensive combined phonetic-phonological speech therapy approach in Ugandan patients with CP±L. Patients received six 1-hour speech therapy sessions during 3 consecutive days. These studies demonstrated that this short and intensive approach can be effective for patients with CP±L in countries with limited access to speech therapy. However, it was concluded that there an urgent need to compare the effectiveness of this combined approach to a pure motor-phonetic approach, in order to provide the most effective, efficient and sustainable care for patients with CP±L.

To respond to the limited literature, the present pilot study investigated the effectiveness of speech intervention in Ugandan patients with CP±L. Moreover, a comparison was made between a motor-phonetic and a phonetic-phonological speech intervention. It was hypothesized that, irrespective of the used approach, speech intervention would be beneficial in reducing active articulation errors and thereby, in increasing consonant proficiency. Furthermore, given the combined phonetic-phonological nature of active speech errors, the phonetic-phonological approach was hypothesized to be the most effective treatment technique in terms of eliminating active speech errors.

2. Materials and methods

This pilot study was approved by the Mildmay Uganda Research Ethics Committee (0611-2017) and the Uganda National Council for Science and Technology (HS 2448). All the 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 (i.e. Luganda) by one of the health professionals of the X hospital (i.e. speech-language pathologists (SLPs), nurses or social workers).

2.1. Participants

Ugandan patients with an isolated CP±L and presence of at least one active speech error who presented at X hospital were asked to participate in the present study. Participants with (1) moderate or severe hearing loss (subjectively reported by the caregiver or patient), (2) neurological disorders, (3) cognitive impairments and (4) insufficient proficiency in English and/or illiteracy were excluded. Patients who were not able to stay in the hospital for one week (to receive treatment) or who simultaneously followed speech therapy provided by a local speech-language pathologist (SLP) were also excluded to participate (*n*=4). All information was obtained from the patient's files and by questioning the caregivers or the patients. Unfortunately, the presence or absence of velopharyngeal insufficiency (VPI) could not be taken into account for the selection of patients, since no visualization of the velopharyngeal mechanism by means of nasoendoscopy or videofluoroscopy was available at this hospital.

2.2. Sample size calculation and study design

Based on a pilot study that investigated the short-term effect of speech therapy in five Ugandan patients with CP±L [22], a sample size calculation was performed based on the percentage correct consonants (PCCs) before and after intervention. "SAS power and sample size calculation" [28] was used to calculate sample size using an alfa-level of 0.05 and an estimated power of 0.80. To receive a power of 0.82, a sample size of at least 10 participants per group was needed. According to Connelly [44], a pilot study sample should include 10% of the calculated sample resulting in minimal 2 participants per group for the present pilot study.

Participants were seen for speech assessment on three separate data points: baseline measurement (22-44 months before speech therapy) (T1), pre-treatment measurement (immediately before speech therapy) (T2), and post-treatment measurement (immediately after the ending of the speech intervention period) (T3). Taking into account the limited access to speech therapy services for Ugandan patients, principles of a pragmatic randomized controlled trial (PRCT) were used to assess effectiveness of motor-phonetic versus phonetic-phonological speech therapy approaches [24]. A PRECIS-2 (Pragmatic Explanatory Continuum Indicator Summary-2) tool assessment was performed [25]. This tool has nine dimensions for assessing the level of pragmatism in a clinical trial on the explanatory–pragmatic continuum (i.e. (1) eligibility criteria, (2) recruitment path, (3) setting, (4) organization: expertise and resources needed to deliver the intervention, (5) flexibility in delivery of the intervention, (6) flexibility of adherence of participants to intervention, (7) follow-up of participants, (8) primary outcome and its relevance to the participants, and (9) primary analysis: how much of the data are included). Each of these domains is scored on a scale of 1–5 from highly explanatory to highly pragmatic [24, 25]. Using this tool, the study was found to be rather pragmatic.

2.3. Speech therapy

Patients were assigned to two groups (i.e. group 1: motor-phonetic approach, group 2: combined phonetic-phonological approach), taking into account the principles of PRCTs [24, 25]. All patients received six hours of individual speech therapy during three consecutive days (i.e. 2 hours per day: one hour in the morning, one hour in the afternoon) in a clinical room at X hospital. Speech therapy was provided in English, since this language is one of the official languages in Uganda. Intervention was provided by non-Ugandan SLPs (A.L./K.B./C.A.) with professional English proficiency. All speech therapists had experience with cleft-related speech (A.L.: 10 years, K.B.: 8 years, C.A.: 2 years).

2.4. Group 1: motor-phonetic approach (MPA)

The patients in group 1 received six hours individual motor-phonetic articulation therapy [5], thus target consonants differed between the patients. Articulation errors were treated in a phoneme-by-phoneme basis following the guidelines described by Van Riper [5]. The motor-phonetic treatment consisted of four different stages on six different levels: (1) identification of the target sound, (2) discrimination of the target sound, (3) variation and correction of the target sound and (4) stabilization and transfer of the target sound in isolation, in syllables, in words, in sentences, in texts and in spontaneous speech [5]. Features of the target consonant were identified using visual, tactile and/or auditory cues. External devices such as mirrors or wooden tongue blades were used for visual and tactile feedback, respectively. Subsequently, auditory discrimination was practiced by differentiating between correct and error consonants, produced by the SLP. If the patient was able to discriminate between the SLP's production, he/she was encouraged to compare his/her own productions with those of the SLP to detect differences. After achieving the step of auditory discrimination, correct production of the target consonants was elicited using progressive approximation, direct auditory stimulation/imitation, articulatory placement and/or modification

from another known sound [5]. Stabilization of the correct production of the target consonants was elicited by repeating and prolonging the sound and varying loudness levels [5]. Therapy focused successively on the production in syllables, words, sentences, texts and transfer to spontaneous speech. A next level was introduced when the patient was able to produce the target consonant in approximately 90% of the time with minimal cues from the SLP [5].

2.5. Group 2: combined phonetic-phonological approach (PPA)

The patients in group 2 received six hours individual combined phonetic-phonological speech therapy. In accordance with the patients of the MPA group, a phonetic approach, described by Van Riper [5], was used to elicit target consonants. Additionally, phonological methods were applied to establish contrast between the target sounds. More specifically, the distinctive feature approach was used [26, 27]. An important principle of this approach is that distinctive features are the underlying parameters of sounds and that phonetic errors are the surface realization of inappropriately learned underlying (phonological) features [27, 28]. Since features are the parameters of phonemes, features must be taught within the context of phonemes. Within the purpose of this study, features were trained by systematically contrasting two sounds that differ in the presence or absence of that specific feature. The intervention began with the production of the phoneme containing the target feature. The child was asked to imitate the target sound. If the production of the target was not correct after imitation, the traditional elicitation techniques described by Van Riper [5] were used. If the patient was able to produce the feature in isolation, a feature contrast training was implemented in which two sounds with one contrastive feature were elicited. The features were gradually trained in isolation, in syllables, in words, in sentences, in texts and during spontaneous speech production. On the word and the sentence level, different minimal pairs were used. The SLP provided the child feedback on the adequacy of the spoken message stimulating the child's (meta)phonological awareness and pragmatic skills. It has been recognized that working with minimal pairs, by providing feedback on the spoken message, enhances the generalization to other sounds that share the same feature as the targeted feature [26 – 28].

2.6. Speech sample

An identical speech sample was used to assess speech at the baseline, pre- and post-treatment data points. The speech sample included (1) repetition of the English sentences of the MacKay-Kummer Simplified Nasometric Assessment Procedures (SNAP) test [29], (2) repetition of the English words of the standardized Photo Articulation Test — Third Edition (PAT-3) [30], and (3) automatic speech (i.e. counting from 1 to 10 and from 60 to 70 and naming the days of the week). All assessments were simultaneously audio- and video-recorded using an unidirectional condenser microphone (Samson, CO1U) and a Sony HDR-UX1 camera.

2.7. Speech assessment

2.7.1. Perceptual assessment

Hypernasality, hyponasality, nasal emission and nasal turbulence were perceptually assessed using the scales of the CAPS-A outcome tool [31, 32]. Speech understandability and speech acceptability were evaluated using the guidelines described by Henningsson et al. [33]. Firstly, all variables were scored using the audio-recordings of the automatic speech sample and repetition of the sentences of the SNAP-test [29]. Secondly, variables were re-assessed based on video-recordings of the same speech sample. For articulatory analysis, all video-recorded speech samples of the PAT-3 test [30] and the SNAP test [29] were played back through over-ear headphones (Sennheiser EH150, Wedemark, Germany) and were phonetically transcribed by two SLPs (C.A. and K.B.) using the International Phonetic Alphabet (IPA) [34] and the IPA extensions as well as additional symbols to

describe specific cleft-related articulation errors [35]. The articulation assessment focused on 15 consonants (i.e. /p/, /b/, /t/, /d/, /g/, /k/, /s/, /z/, /f/, /v/, /h/ /l/, /m/, /n/, and /ŋ/) found in English as well as in Luganda [22]. Articulatory errors were categorized in different cleft speech characteristics (CSCs): anterior CSCs, posterior CSCs, non-oral CSCs and passive CSCs [31, 32]. The present study calculated percentages of occurrence of the different CSCs in order to compare changes before and after speech therapy more precisely [21, 23]. In addition, consonant proficiency, on both word and sentence level, was calculated using percentage correct consonants (PCC), percentage correct places (PCP) and percentage correct manners (PCM) [36].

All these speech samples were anonymized and randomized, hence the two raters were blinded to both the participants and the data points. The first rater (C.A.) analyzed 100% (24/24) of the speech samples. In order to calculate inter-rater reliability, the second rater (K.B.) analyzed 37.5% (9/24) of the speech samples. Moreover, the first SLP re-rated 100% (24/24) of the speech samples after 4 weeks in a different randomized order to determine intra-rater reliability.

Based on the ratings of the assessment of the variables hypernasality, nasal emission, nasal turbulence, non-oral CSCs and passive CSCs, the Velopharyngeal composite score-summary CAPS-A (VPC-SUM) was determined [37]. VPC-SUM scores were interpreted as follows: score 0-1: sufficient velopharyngeal function; score 2: borderline deficit and score 3-4: insufficient velopharyngeal function [37].

2.7.2. Instrumental assessment

Nasalance values for the sentences of the SNAP test [29] and for the oral zoo passage and oronasal rainbow passage [38] were obtained using a KayPentax Nasometer (model II 6450) (NJ, Lincoln Park). At the beginning of each assessment procedure, the device was calibrated in a quiet room at X hospital following the instructions of the manufacturer's manual. Moreover, 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/ [39]. 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. [40]. 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 [39].

2.7.3. Statistical analysis

IBM SPSS Statistics software version 25.0 (IBM Corp., Armonk, NY) was used for the statistical analysis of the data. The significance level was set at $p \le 0.05$. Both inter- and intra-rater reliability were calculated for all parameters by means of two-way random ICCs type consistency (single measures). ICCs were interpreted following the classification of Altman [41] (ICC < 0.20: poor, 0.21-0.40: fair, 0.41-0.60: moderate, 0.61-0.80: good, 0.81-33 1.00: very good). To investigate the effectiveness of speech intervention for the whole group (irrespective of the used approach), withingroup comparisons over time were made (T1-T2, T1-T3, T2-T3) using a Wilcoxon Signed Rank Test. To compare the two groups over time (i.e., T1, T2, and T3), between-group comparisons were made using the Mann Whitney U-test and the Fisher's exact test for continuous and categorical variables, respectively. P-values for the Wilcoxon Signed Rank Test and Mann Whitney U-test were adjusted using the Benjamini-Hochberg correction with the false discovery rate set at 0.1 [42]. Each individual P-value was compared to its Benjamini-Hochberg critical value (calculated using the formula "(i/m)Q", where i corresponds to the rank, m to the total number of tests, and Q to the false discovery rate (i.e., 0.1)). The largest P-value that had P-(i/m)Q was considered to be significant, and all of the P-values smaller than this value were also considered significant [42]. Moreover, effect

size estimates (r) were calculated and interpreted following the classification of Cohen [43] (r=0.10: small effect, r=0.30: medium effect, r=0.50 large effect).

3. Results

3.1. Participants

Eight patients with CP±L (MPA group: n=2 men, n=2 females, PPA group: n=2 men, n=2 females) presented at X hospital and fulfilled the inclusion criteria. Median age of the participants was 11.26 years (minimum-maximum (min-max): 6.87y-39.50y) and 16.19 years (min-max: 10.63y-17.43y) for the MPA group and the PPA group respectively. The Mann Whitney U-test indicated no statistically significant difference in age between the two groups (W= 8.0, p=1.000). One patient from the PPA group had already followed speech therapy in the past (2 hours/day during 6 weeks). None of the other included patients had followed speech therapy. Demographic and cleft characteristics are provided in Table 1.

3.2. Reliability analysis

Results regarding inter- and intra-rater reliability are provided in Table 2. Inter-rater reliability was good to very good for all parameters. Regarding intra-rater reliability, moderate ICC values were found for the parameters "audible nasal emission" and "passive CSCs", However, good to very good single ICC's were found for the other assessed variables.

3.3. Effectiveness of speech intervention irrespective of the used approach

Results for the evolution of the whole group are presented in Table 3. No statistically significant differences in pre-treatment measurements (i.e. T1-T2) were found when comparing the measures for the whole group (Wilcoxon Signed Rank Test, p>0.05). According to the Benjamini-Hochberg procedure (BHP), the PCCs and PCPs scores on the word and sentence level were statistically significant higher on T3 compared to T1 and T2 (P-value<(i/m)Q, r>0.50) (Table 3). Considering the PCM scores on the word level, the BHP revealed statistically significant higher scores on T3 compared to T1 (P-value<(i/m)Q, r=0.84) (Table 3). Comparing the occurrence of CSCs, a statistically significant lower occurrence of non-oral CSCs was found after performance of the BHP procedure on T3 when compared to T1 and T2 (*P-value*<(*i/m*)*Q*, *r*=-0.89). The BHP also demonstrated a statistically significant lower occurrence of passive CSCs on T3 compared to T1 and T2 (P-value< (i/m)Q, r=-0.89). For the anterior and posterior CSCs, the BHP revealed no statistically significant differences when comparing T1-T3, T1-T2 and T2-T3 (P-value>(i/m)Q). Within the whole group (n=8), a statistically significant lower NSI 2.0 value was found on T3 when compared to T2 and T1 after performance of the BHP (P-value < (i/m)Q) (Table 3). In accordance, statistically significant lower nasalance values for the oral and oronasal passages were found on T3 when compared to T2 and T1 (P-value<(i/m)Q) (Table 3).

3.4. Effectiveness of motor-phonetic versus linguistic-phonological speech therapy approach

Results for the comparison of the motor-phonetic and linguistic-phonological approaches are presented in Table 4. The BHP revealed that there were no statistically significant differences between the two groups for none of the data points (*P-values>(i/m)Q)* (Table 4). When considering the descriptive statistics, however, a greater increase in median PCCs, PCPs and PCMs scores on the word and sentence level was seen when comparing the pre- and post-treatment measurements (i.e. T2 and T3) in the PPA group and in the MPA group (Table 4). The median PCC scores on the word level increased 49.45% in the PPA group compared to 24.4% in the MPA group. In accordance, the median PCC scores on the sentence level increased 29.7% and 3.88% in the PPA and MPA group,

Table 1. Demographic and cleft characteristics for the eight included patients.

Patient	Motor-phonetic grou	p (MPA) (n	- 4)		Phonetic-phonological group (PPA) $(n = 4)$				
	1	2	3	4	1	2	3	4	
Age	6;9y	10;4y	39;5y	19;7y	17;4y	11;2y	10;6y	16;1y	
Gender	F	M	M	F	F	M	F	M	
Native language	Kakwa	Luganda	Luganda	Luganda	Rutooro	Luganda	Luganda	Luganda	
Cleft type	UCLP	UCLP	UCLP	UCLP	CP	MCLP	UCLP	CP	
Age of palatal closure	2 m	5 m	22y	Зу	6 m	5 m	11 m	13y	
Type of closure	one-stage ^b	one-stage	one-stage	one-stage	one-stage	one-stage	one-stage	one-stage	
Secondary surgery	revision cleft lip (4;1y)	/	cleft lip and palate re-repair (35;0y)	revision cleft lip (18;3y) fistula repair (18;3y) palatal re-repair (19;0y)	fistula repair (14;0y) buccal flap (14;6y) division buccal flap (15;0y)	buccal flap (5;0y)	buccal flap (8;5y) alveolar bone graft (9;0y)	/	
	fistula repair (4;1y) nasal correction with open rhinoplasty								
Speech therapy	no	по	по	no	yes	по	no	no	

years (y); months (m); male (M); female (F); cleft palate (CP); unilateral cleft lip and palate (UCLP); median cleft lip and palate (MCLP); cleft palate (CP).

aAge at moment of speech intervention.

b simultaneous lip and palatal closure.

Table 2. Inter- and intra-rater reliability by means of a two-way mixed ICC (consistency).

	Inter-rater reliabi	lity		Intra-rater reliability			
	Single ICC consistency	95% CI single consistency	Interpretation of single ICC**	Single ICC consistency	95% single ICC consistency	Interpretation of the single ICC**	
Speech under- standability	0.933	0.631-0.998	very good	0.700	0.382-0.918	good	
Speech acceptability	0.933	0.631-0.998	very good	0.652	0.318-0.902	good	
Hypernasality	0.982	0.878-1.000	very good	0.858	0.652-0.965	very good	
Hyponasality	.*	-	-	-	-		
Audible nasal emission	0.700	-0.090-0.991	good	0.556	0.203-0.865	moderate	
Nasal turbulence	1.000	1.000-1.000	very good	1.000	1.000-1.000	very good	
Anterior oral CSCs	0.722	0.125-0.991	good	0.784	0.513-0.944	good	
Posterior oral CSCs	1.000	1.000-1.000	very good	0.618	0.196-0.896	good	
Non-oral CSCs	0.900	0.506-0.997	very good	0.635	0.296-0.896	good	
Passive CSCs	0.722	0.125-0.991	good	0.505	0.151-0.844	moderate	

confidence interval (CI), *reliability was not computed as there was no variance in the data set for this parameter **based on Altman [41]: 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.

Table 3. Effectiveness of speech intervention irrespective of the used approach (n = 8).

	Descriptive statistics: median (min-max)			Within-group comparison $(n = 8)$				
	T1	T2	Т3	T1-T2	T1-T3	T2-T3		
				p-value	p-value	p-value		
PCC word level PCC sentence level PCP word level PCP sentence level PCM word level PCM sentence level NSI 2.0 Oral passage Oronasal passage Anterior CSCs	39.85 (13.20-51.20) 61.17 (25.50-70.13) 66.82 (34.40-85.47) 61.84 (31.11-89.00) 86.30 (67.72-98.65) 84.15 (63.40-94.62) -10.87 (-15.25;-2.71) 58.00 (38.00-66.00) 57.00 (35.00-70.00) 21.79 (0.00-56.10)	40.13 (18.61-51.81) 63.30 (32.26-70.96) 65.64 (32.61-86.93) 61.80 (33.33-89.20) 87.20 (63.55-100.00) 87.64 (62.36-100.00) -11.65 (-14.78;-2.21) 60.00 (30.00-71.00) 59.00 (39.00-72.00) 20.46 (0.00-55.42)	79.88 (36.58–95.91) 78.82 (37.36–98.90) 78.70 (42.77–94.80) 73.05 (55.58–100.00) 92.33 (72.60–100.00) 92.31 (67.40–100.00) -8.61 (-14.80;-4.10) 46.00 (13.00–71.00) 47.50 (28.00–69.00) 2.41 (0.00–53.00)	$\begin{array}{lll} W = 25.00, p = 0.663, r = 0.66 \\ W = 22.00, p = 0.293, r = 0.89 \\ W = 11.00, p = 0.327, r = -0.35 \\ W = 21.00, p = 0.237, r = 0.42 \\ W = 25.50, p = 0.293, r = 0.37 \\ W = 19.00, p = 0.398, r = 0.30 \\ W = 21.00, p = 0.237, r = 0.42 \\ W = 9.00, p = 0.752, r = -0.12 \\ W = 21.00, p = 0.293, r = 0.78 \\ W = 2.50, p = 0.176, r = -0.48 \\ \end{array}$	$\begin{array}{llll} W &=& 36.00, p = 0.012^{+*}, r = 0.89 \\ W &=& 36.00, p = 0.012^{+*}, r = 0.89 \\ W &=& 34.00, p = 0.025^{+*}, r = 0.79 \\ W &=& 36.00, p = 0.012^{+*}, r = 0.84 \\ W &=& 36.00, p = 0.012^{+*}, r = 0.84 \\ W &=& 21.00, p = 0.237, r = 0.42 \\ W &=& 25.00, p = 0.063^{+*}, r = 0.66 \\ W &=& 2.50, p = 0.050^{+*}, r = -0.69 \\ W &=& 2.00, p = 0.075^{+*}, r = -0.63 \\ W &=& 3.00, p = 0.116, r = -0.55 \\ \end{array}$	$\begin{array}{llll} W &=& 36.00, p = 0.012^{++}, r = 0.89 \\ W &=& 36.00, p = 0.012^{++}, r = 0.89 \\ W &=& 35.00, p = 0.017^{++}, r = 0.84 \\ W &=& 34.00, p = 0.025^{++}, r = 0.79 \\ W &=& 23.00, p = 0.128, r = 0.54 \\ W &=& 17.00, p = 0.612, r = 0.18 \\ W &=& 33.00, p = 0.036^{++}, r = 0.74 \\ W &=& 0.00, p = 0.018^{++}, r = -0.84 \\ W &=& 4.50, p = 0.058^{++}, r = -0.67 \\ W &=& 4.00, p = 0.173, r = -0.48 \\ \end{array}$		
Posterior CSCs Non-oral CSCs Passive CSCs	0.00 (0.00-0.00) 47.55 (10.19-88.60) 2.50 (0.00-33.97)	0.00 (0.00-0.67) 47.18 (10.19-87.50) 4.67 (0.00-33.97)	0.00 (0.00-0.00) 7.60 (0.00-67.50) 0.00 (0.00-26.70)	W = 1.000, p = 0.317, r = 0.35 W = 6.00, p = 0.345, r = -0.33 W = 7.00, p = 0.465, r = -0.33	W = 0.000, p = 1.000, r = -0.35 W = 0.00, p = 0.012**, r = -0.89 W = 0.00, p = 0.068**, r = -0.89	W = 0.000, p = 0.317, r = -0.35 W = 0.00, p = 0.012**, r = -0.89 W = 0.00, p = 0.028**, r = -0.89		

minimum-maximum (min-max); percentage consonants correct (PCC); baseline measurement (22–44 months before speech therapy) (T1); pre-treatment measurement (immediately before speech therapy) (T2); post-treatment measurement (immediately after speech therapy) (T3); **P-value considered to be statistically significant according to the Wilcoxon Signed-Rank test with post-hoc Benjamini-Hochberg correction (i.e., P< (i/m)Q) [42].

Table 4. Effectiveness of motor-phonetic versus phonetic-phonological speech therapy approach (n=4 participants per group).

	Descriptive statistics: median (min-max)						Between-group comparison			
	MPA group (n = 4)			PPA group $(n - 4)$			T1 T2	T2	Т3	
	T1	T2	Т3	T1	T2	Т3	p-value	p-value	p-value	
PCC word level	38.10	39.58 (18.61-51.81)	63.98 (36.58-78.90)	40.95 (37.35-43.77)	40.75 (37.16-44.70)	90.20 (83.86-95.91)	U = 10.00, z = 0.58,	U = 10.00, z = 0.58,	U = 16.00, z = 2.31,	
	(13.20-51.20)						p = 0.686, r = 0.20	p = 0.686, r = 0.20	p = 0.029, r = 0.82	
PCC sentence level	61.62	63.39 (32.26-68.49)	67.27 (37.36-83.97)	61.17 (42.92-70.13)	63.30 (43.31-70.79)	93.00 (73.66-98.90)	U = 9.00, z = 0.30,	U = 9.00, z = 0.30,	U = 15.00, z = 2.021,	
	(25.50-64.20)						p = 0.886, r = 0.10	p = 0.886, r = 0.10	p = 0.043, r = 0.71	
PCP word level	64.98	63.86 (32.61-65.70)	64.61 (42.77-74.50)	70.95 (51.89-85.47)	70.55 (52.80-86.93)	90.36 (82.90-94.80)	U = 13.00, z = 1.44,	U = 13.00, z = 1.44,	U = 16.00, z = 2.31,	
	(34.40-67.84)						p = 0.149, r = 0.51	p = 0.149, r = 0.51	p = 0.021, r = 0.82	
PCP sentence level	61.84	63.44 (33.33-68.49)	66.69 (55.58-76.60)	64.81 (45.50-89.00)	61.69 (49.40-89.20)	95.69	U = 10.00, z = 0.58,	U = 8.00, z = 0.00,	U = 14.00, z = 1.73,	
	(31.11-66.34)					(69.80-100.00)	p = 0.564, r = 0.20	p = 1.000, r = 0.00	p = 0.083, r = 0.62	
PCM word level	84.83	89.60	88.92	86.67 (85.00-92.60)	87.20 (84.00-93.75)	92.91 (88.60-99.40)	U = 10.00, z = 0.58,	U = 7.00, z = -0.29,	U - 11.00, z - 0.87	
	(67.72-98.65)	(63.55-100.00)	(72.60-100.00)				p = 0.686, r = 0.20	p = 0.886, r = -0.10	p = 0.386, r = 0.31	
PCM sentence	78.50	94.09	91.94	84.24 (67.15-94.62)	84.40 (67.70-93.55)	92.31 (76.30-98.90)	U = 10.00, z = 0.58,	U = 5.00, z = -0.87,	U = 7.00, z = -0.29,	
level	(63.40-93.36)	(62.36-100.00)	(67.40-100.00)				p = 0.564, r = 0.20	p = 0.384, r = -0.14	p = 0.772, r = -0.10	
NSI 2.0	-5.57 (-10.87;	-7.68	-2.73 (-12,37;	-12.68	-13.68	-12.88	U = 2.00, z = -1.41,	U = 5.00, z = -0.87,	U = 5.00, z = -0.87,	
	-2.71)	(-14,55;-2.21)	3.24)	(-15,24;-3.53)	(-14.78; -3.50)	(14,80;4,10)	p = 0.157, r = -0.50	p = 0.386, r = -0.31	p = 0.386, r = -0.31	
Oral passage	42.00	47.50 (30.00-71.00)	33.00 (14.00-71.00)	60.00 (43.00-66.00)	65.50 (40.00-70.00)	37.75 (14.00-71.00)	U = 11.00, z = 1.78,	U = 11.00, z = 0.87,	U = 9.00, z = 0.29,	
	(38.00-58.00)						p = 0.074, r = 0.63	p = 0.386, r = 0.31	p = 0.773, r = 0.10	
Oronasal passage	45.00	56.50 (39.00-67.00)	35.00 (34.00-69.00)	63.00 (41.00-70.00)	65.00 (43.00-72.00)	62.50 (28.00-65.00)	U = 9.00, z = 1.06,	U = 11.50, z = 1.06,	U = 9.00, z = 0.29,	
	(35.00-59.00)						p = 0.289, r = 0.37	p = 0.309, r = 0.37	p = 0.770, r = 0.10	
Anterior CSCs	16.46 (2.24-40.25)	16.96 (1.24-36.14)	2.41 (0.00-53.00)	21.79 (0.00-56.10)	20.46 (0.00-55.42)	4.82 (0.00-15.66)	U = 8.00, z = 0.00,	U = 8.00, z = 0.00,	U = 7.00, z = -0.30,	
	,	, ,		1	, ,		p = 1.000, r = 0.00	p = 1.000, r = 0.00	p = 0.886, r = -0.10	
Posterior CSCs	0.00 (0.00-0.00)	0.00 (0.00-0.00)	0.00 (0.00-0.00)	0.00 (0.00-0.00)	0.00 (0.00-0.67)	0.00 (0.00-0.00)	U = 8.00, z = 0.00,	U - 10.00, z - 1.00,	U - 8.00, z - 0.00,	
	()	(,	(,	()	(,	,	p = 1.000, r = 0.00	p = 0.686, r = 0.35	p = 1.000, r = 0.00	
Non-oral CSCs	44.18	42.68 (10.19-84.89)	7.60 (0.00-67.50)	47.55 (24.10-88.60)	47.18 (23.89-87.50)	9.500 (2.50-45.00)	U = 10.00, z = 0.58,	U = 10.00, z = 0.58,	U = 8.00, z = 0.00,	
The second second	(10.19-83.24)	(10.11 0 1.03)	(0.00 07.00)	(21110 00100)	(20102 07100)		p = 0.564, r = 0.20	p = 0.564, r = 0.20	p = 1.000, r = 0.00	
Passive CSCs	2.56 (0.00-7.99)	4.96 (1.71-7.40)	0.00 (0.00-1.14)	2.50 (0.00-33.97)	2.28 (0.00-33.97)	0.57 (0.00-26.70)	U = 8.00, z = 0.00,	U = 5.00, z = -0.87,	U = 10.00, z = 0.66,	
	(0.00 7.00)					(0.00 20.00)	p = 1.000, r = 0.00	p = 0.384, r = -0.31	p = 0.564, r = 0.23	

minimum-maximum (min-max); percentage consonants correct (PCC); baseline measurement (22–44 months before speech therapy) (T1); pre-treatment measurement (immediately before speech therapy) (T2); post-treatment measurement (immediately after speech therapy) (T3); **P-value considered to be statistically significant according to the Mann-Whitney U test with post-hoc Benjamini-Hochberg correction (i.e., P < (i/m)Q) (42).

Table 5. Results for the categorical outcome measures in the MPA (n=4) and PPA groups (n=4). Statistical analysis based on Fisher's Exact Test (between group-comparison) with significance level set on p < 0.05.

		T1		T2		Т3	
		MPA group	PPA group	MPA group	PPA group	MPA group	PPA group
Speech intelligibility	Within normal limits	-	_	-	_	n = 2 (50%)	n = 1 (25%)
	Mild	n = 1 (25%)	_	n = 1 (25%)	_	n = 1 (25%)	n = 3 (75%)
	Moderate	n = 1 (25%)	n = 2 (50%)	n = 1 (25%)	n = 2 (50%)	-	-
	Severe	n = 2 (50%)	n = 2 (50%)	n = 2 (50%)	n = 2 (50%)	n = 1 (25%)	_
Significance		p = 1.000	, ,	p = 1.000	, ,	p = 0.486	
		MPA group	PPA group	MPA group	PPA group	MPA group	PPA group
Speech acceptability	Within normal limits	-	-		-	n = 1 (25%)	n = 1 (25%)
	Mild	n = 3 (75%)	-	n = 3 (75%)	-	n = 2 (50%)	n = 3 (75%)
	Moderate	-	n = 3 (75%)	-	n = 3 (75%)	-	-
	Severe	n = 1 (25%)	n = 1 (25%)	n = 1 (25%)	n = 1 (25%)	n = 1 (25%)	-
Significance		p = 0.057		p = 0.057		p = 1.000	
		MPA group	PPA group	MPA group	PPA group	MPA group	PPA group
Hypernasality	Absent	n = 1 (25%)		n = 1 (25%)		n = 2 (50%)	n = 1 (25%)
	Borderline/minimal	-	-	-	-	n = 1 (25%)	-
	Mild	-	n = 1 (25%)	-	n = 1 (25%)	-	n = 2 (50%)
	Moderate	n = 2 (50%)	n = 1 (25%)	n = 2 (50%)	n = 1 (25%)	-	n = 1 (25%)
	Severe	n = 1 (25%)	n = 2 (50%)	n = 1 (25%)	n = 2 (50%)	n = 1 (25%)	-
Significance		p = 1.000		p = 1.000		p = 0.486	
		MPA group	PPA group	MPA group	PPA group	MPA group	PPA group
Audible nasal emission	Absent	n = 3 (75%)	n = 1 (25%)	n = 3 (75%)	n = 1 (25%)	n = 4 (100%)	n = 2 (50%)
	Occasional	n = 1 (25%)	n = 2 (50%)	n = 1 (25%)	n = 2 (50%)	-	n = 2 (50%)
	Frequent	-	n = 1 (25%)	-	n = 1 (25%)	-	-
Significance		p = 0.486		p = 0.486		p = 0.492	
		MPA group	PPA group	MPA group	PPA group	MPA group	PPA group
Nasal turbulence	Absent	n = 4 (100%)	n = 3 (75%)	n = 4 (100%)	n = 3 (75%)	n = 4 (100%)	n = 4 (100%)
	Occasional	-	n = 1 (25%)	-	n = 1 (25%)	-	-
	Frequent	-	-	-	-	-	-
Significance		p - 1.000		p - 1.000		p - 1.000	
		MPA group	PPA group	MPA group	PPA group	MPA group	PPA group
VPC-sum	Sufficient velopharyngeal function	n = 2 (50%)	-	n = 2 (50%)	-	n = 3 (75%)	n = 2 (50%)
	Borderline deficit	n - 1 (25%)	n = 1 (25%)	n = 1 (25%)	n = 2 (50%)	-	n = 1 (25%)
	Insufficient velopharyngeal function	n = 1 (25%)	n = 3 (75%)	n = 1 (25%)	n = 2 (50%)	n = 1 (25%)	n = 1 (25%)
Significance		p = 0.657		p = 0.486		p = 1.000	

percentage consonants correct (PCC); baseline measurement (22–44 months before speech therapy) (T1); pre-treatment measurement (immediately before speech therapy) (T2); post-treatment measurement (immediately after speech therapy) (T3); Note: hyponasality was not presented since this parameter was assessed as absent in all patients.

respectively. An increase of 19.81% for the PPA group and 0.75% of the MPA group was found for the PCP scores on the word level. The median PCP scores on the sentence level increased 3.25% for the MPA group and 34.00% for the PPA group. A negative trend for the median PCM scores on the word and sentence level was seen in the MPA group: the PCMs decreased 0.68% and 2.15%, respectively. This was in contrast with the PPA group where an increase in median PCM scores on the word and sentence level was found after the intervention. For the PPA group, the median PCM scores increased 5.71% and 7.91% on the word and sentence level, respectively. Considering the categorical outcome measures (i.e. speech intelligibility, speech acceptability, hypernasality, hyponasality, audible nasal emission, nasal turbulence, and VPC-sum score), the Fisher's exact test showed no statically significant differences between the MPA group and the PPA group at all data points (p>0.05) (Table 5).

4. Discussion

Over the past 35 years, few studies investigated the outcomes of speech intervention in patients with CP±L [4]. The small sample sizes, methodological flaws and non-specific description of the treatment setting, frequency, provider, and included population made the generalization of the results difficult. To respond to the limited literature, this study investigated the effectiveness of speech therapy in Ugandan patients with CP±L. More specifically, a comparison between a motor-phonetic versus a phonetic-phonological approach was made. It was hypothesized that, irrespective of the used approach, the speech intervention would be beneficial in reducing active articulation errors and thereby, in increasing consonant proficiency. Furthermore, given the combined phonetic-phonological nature of active speech errors, the phonetic-phonological approach was hypothesized to be the most effective treatment technique.

4.1. Effectiveness of speech therapy (irrespective of the used approach)

Concerning the evolution of the whole group irrespective of the used approach, no statistically significant difference in speech outcomes was found when comparing the data points before provision of speech therapy, indicating stable measurements before the intervention (Table 3). For the whole group, consonant proficiency in terms of PCCs, PCPs and PCMs was significantly higher after the provision of speech therapy (Table 3). Interestingly, large effect size estimates were found for these significant results (r>0.50) (Table 3). Considering the CSCs, lower occurrence of non-oral and passive CSCs was found after speech therapy (Table 3). These results were in line with Derakhshandeh et al. [21] who found that speech therapy may decrease the presence of these types of CSCs in patients with velopharyngeal insufficiency (VPI). If there is the potential for velopharyngeal closure or near closure, the child might possible learn to articulate oral pressure consonants during therapy [21]. Unfortunately, the present study could not provide any information about the velopharyngeal functioning in the included patients since no visualization equipment was available at CoRSU hospital. The VPC-sum scores, however, indicated that six of the included patients (75%) presented with either borderline (n=1) or sufficient (n=5) velopharyngeal function after speech therapy (Table 5) which might explain the findings for the passive CSCs.

No evolution in anterior nor in posterior CSCs was found when comparing results for the whole group over time. Posterior CSCs were absent in seven of the eight included patients explaining the absence of significant differences when comparing pre- and post-treatment conditions. Despite these non-significant results, a positive trend towards lower occurrence percentages of anterior CSCs was seen after speech therapy (Table 3). Most likely, the small sample size (n=8) prevented obtaining significant results.

Regarding the instrumental assessment, significantly lower NSI 2.0 values and nasalance values for the oral and oronasal passages were found after speech therapy. In accordance, Luyten et al. [22] and Alighieri et al. [23] found that speech therapy can possibly decrease the presence of resonance disorders indirectly.

4.2. Effectiveness of motor-phonetic versus linguistic-phonological speech therapy approach

The Mann Whitney U-test with post-hoc performance of the BHP revealed that there were no statistically significant differences between the two groups when comparing the pre- and posttreatment measurements (Table 4). Nevertheless, the descriptive statistics revealed a larger increase in PCP, PCP and PCM scores after the intervention in the PPA group compared to the MPA group (Table 4). In the PPA group, the distinctive feature approach [26, 27] was used in addition to the motor-phonetic approach [5]. Using this technique, distinctive features were trained by systematically contrasting two sounds that differ in the presence or absence of that specific feature. Additionally, greater emphasis was put on features such as place and manner of articulation. Interestingly, a decrease in PCM scores on the word and sentence level was found for the MPA group after the intervention. This was in contrast with the PPA group were an increase in PCM scores was found after the intervention. In the PPA group, phonological methods were applied to support the establishment of contrasts between oral and nasal resonance and nasal airflow [26, 27] paying greater attention to the manner of articulation. This difference in focus of the two approaches applied in the two groups might explain these findings. It might also be possible that languagebased exercises in natural communicative contexts as used in the PPA group are easier to implement and establish than the motor-based exercises in the MPA group.

Considering the ideas behind the distinctive feature approach, it is questionable why no statistically significant results were found when comparing the presence of CSCs between the two groups. In accordance, descriptive results showed decreasing trends that were similar in both groups (Table 4). The present study was one of the first to investigate the effectiveness of a motor-phonetic versus a phonetic-phonological approach, hence it is difficult to compare present findings to existing literature. A plausible explanation for these non-significant results when comparing the groups is the small and heterogeneous sample (n=8). Pamplona et al. [12] compared a phonetic-based intervention with an exclusively phonological-based intervention. Results showed that the phonological approach significantly reduced the total intervention time necessary for complete elimination of active articulation errors in children with CP±L. Nevertheless, time needed for complete correction of the active CSCs was 14.50 months in the phonological group and 30.07 months in the phonetic group, which is considerably longer than the 6 hours of intervention provided in the present study. Moreover, comparison of results is hampered since Pamplona et al. [12] used a pure phonologically-based intervention whereas the present study used a combined phoneticphonological intervention. As argued before, it can be questioned whether a strictly phonologicalbased approach is appropriate for children with CP±L. Children with CP±L have disruptions at some level of the articulatory processes, resulting in active errors and consequently, wrong articulatory placement. Hence, intervention must somehow address the elimination of faulty placement of the articulators by diminishing the motor speech program of the active error and establishing new motor routines for the targeted sounds. Nevertheless, Hoffman [9] suggested that phonological principles are capable of adding new dimensions to traditional motor-phonetic speech intervention.

4.3. Strengths and limitations

To the best of our knowledge, this was one of the first studies comparing the effectiveness of a motor-phonetic versus a phonetic-phonological speech intervention in patients with CP±L in a

resource-limited country such as Uganda. An important limitation is the small and heterogeneous sample (n=8) that most likely prevented obtaining more statistical significant results. Future studies should consider this limitation since the results can vary with different sample characteristics. The heterogeneity of this sample was reflected in the inclusion of 2 children versus one adolescent and one adult in the MPA group and 2 children versus 2 adolescents in the PPA group (Table 1). In other words, there were no exclusion criteria related to the age of the participants resulting in a very large age range (min-max: 6.87-39.50 years). In Uganda, several financial and practical (e.g. transport) considerations often prevent people to seek help. Especially in these contexts, it is highly essential that the research question is valid and relevant to the population. Patients that met the other criteria (i.e. patients with isolated CP±L and good proficiency in English presenting with at least 1 active speech error and absence of moderate/severe hearing loss, neurological disorders and/or cognitive impairments) were included so that every patient was offered the opportunity to follow speech therapy. Using the PRCT, the age of the participants was considered when assigning them to either the MPA or the PPA group. The patient aged 39.50 years, for example, was assigned to the motorphonetic group considering that phonological awareness develops during early childhood. Moreover, mastering the sound system depends on individual experiences, speech-producing capabilities, insights and perception into the structure of the language, and several other learning factors [7]. Taking into account the effect of age on these influencing factors, future research should limit age range to avoid this selection bias. Despite that the PPA group included patients with more heterogeneous cleft types compared to the MPA group (Table 1), the results of the present study were in favor of the PPA group suggesting the benefits of an additional phonological approach. The speech therapy was provided by three different non-Ugandan SLPs of which two assessed the speech samples. Despite randomization and anonymization of the speech samples and good to very good inter- and intra-rater reliability, this might have resulted in trainer bias influencing the findings to some extent.

Doing research in resource-limited countries goes hand in hand with barriers and confounding variables explaining the difficulties in establishing sufficient baseline data points. To establish larger sample sizes, future studies could for example organize intensive speech camps during the holiday periods. For younger children, it might also be interesting to implement parental training programs to train parents how to address speech disorders at home. It has been reported before that parents can be trained to provide speech therapy at home [45]. To which extent this type of intervention is applicable in resource-limited countries remains a topic for further research. Despite these limitations, this pilot study provides preliminary evidence concerning the effectiveness of speech therapy approaches in this unique population and holds some important implications for clinical practice. The descriptive statistics suggested that an additional phonological approach may be beneficial for patients with CP±L, especially in terms of consonant proficiency. Nevertheless, further research including randomized-controlled trials with larger and more homogenous samples, more data points and psychosocial outcome measures will be necessary to provide stronger evidence.

5. Conclusion

At present, there is growing interest in the combined phonetic-phonological dimension of active speech disorders in children with CP±L and its consequences for speech intervention in this population. Unfortunately, evidence for these speech therapy approaches is lacking. Speech therapy (irrespective of the used approach) was found to be effective in increasing consonant proficiency and in decreasing occurrence of non-oral and passive CSCs. The descriptive results revealed that the PPA group showed a greater increase in the PCC, PCP and PCM scores on the word and sentence after the intervention compared to the MPA. Further research including randomized-controlled trials

with larger sample sizes, more data points and psychosocial outcome measures will be necessary to provide stronger evidence for the use of phonological approaches in this population.

Funding

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

References

- 1. Kummer, A.W. *Speech therapy for errors secondary to cleft palate and velopharyngeal dysfunction*. in *Seminars in speech and language*. 2011. © Thieme Medical Publishers.
- 2. Harding, A., P.J.I.j.o.l. Grunwell, and c. disorders, *Active versus passive cleft-type speech characteristics*. 1998. **33**(3): p. 329-352.
- 3. Van Demark, D. *Some results of speech therapy for children with cleft palate.* 1974. **11**(1): p. 41-49.
- 4. Bessell, A., et al., *Speech and Language Therapy Interventions for Children with Cleft Palate: A Systematic Review.* The Cleft Palate-Craniofacial Journal, 2013. **50**(1): p. 1-17.
- 5. Van Riper, C., *Speech correction*. 1972: Prentice-Hall.
- 6. Herreras Mercado, R., Simpson, K. and Bellom-Rohrbacher, K. *Effect of Prompts for Restructuring Oral Muscular Phonetic Targets (PROMPT) on Compensatory Articulation in Children With Cleft Palate/Lip.* 2019. **6**: p. 2333794X19851417.
- 7. Kuehn, D.P. and Moller, K. *Speech and language issues in the cleft palate population: the state of the art.* 2000. **37**(4): p. 1-35.
- 8. Chapman, K. *Phonologic processes in children with cleft palate.* 1993. **30**(1): p. 64-72.
- 9. Hoffman, P. Synergistic development of phonetic skill. 1992. 23(3): p. 254-260.
- 10. Norris, J. Whole language in theory and practice: Implications for language intervention. 1990. **21**(4): p. 212-220.
- 11. Grunwell, P., and Russell, J. *Phonological development in children with cleft lip and palate.* 1988. **2**(2): p. 75-95.
- 12. Pamplona, M.C., Ysunza A, and Espinosa J., A comparative trial of two modalities of speech intervention for compensatory articulation in cleft palate children, phonologic approach versus articulatory approach. 1999. **49**(1): p. 21-26.
- 13. Chen, R. Wang, G. and Sun, Y. *A new speech training method for patients following cleft palate repair.* 1996. **31**(4): p. 220-223.
- 14. Hardin-Jones, M., Chapman, K. *The impact of early intervention on speech and lexical development for toddlers with cleft palate: a retrospective look at outcome.* 2008.
- 15. Ma, L., et al., *A preliminary report of speech training following the cleft palate surgery and its effect.* 1990. **25**: p. 86-89.
- 16. Van Demark, D. and Hardin-Jones, M. *Effectiveness of intensive articulation therapy for children with cleft palate.* 1986. **23**(3): p. 215-224.
- 17. Yang, X., N. Li, and Bu, L. *The analysis of formant characteristics of vowels in the speech of patient with cleft palate.* 2003. **21**(6): p. 451-3, 462.
- 18. Pamplona, M. C., et al., *Speech summer camp for treating articulation disorders in cleft palate patients.* 2005. **69**(3): p. 351-359.
- 19. Pamplona, M.C., A. Ysunza, and Ramírez, P. *Naturalistic intervention in cleft palate children.* 2004. **68**(1): p. 75-81.
- 20. Hodson, B.W., et al., *Phonological evaluation and remediation of speech deviations of a child with a repaired cleft palate: A case study.* 1983. **48**(1): p. 93-98.
- 21. Derakhshandeh, F., et al., Speech characteristics after articulation therapy in children with cleft palate and velopharyngeal dysfunction—A single case experimental design. 2016. **86**: p. 104-113.

- 22. Luyten, A., et al., Short-term effect of short, intensive speech therapy on articulation and resonance in Ugandan patients with cleft (lip and) palate. 2016. **61**: p. 71-82.
- 23. Alighieri, C., et al., *Intensive speech therapy in Ugandan patients with cleft (lip and) palate: a pilot-study assessing long-term effectiveness.* International journal of pediatric otorhinolaryngology, 2019. **123**: p. 156-167.
- 24. Dal-Ré, R., P. Janiaud, and Ioannidis, J. *Real-world evidence: How pragmatic are randomized controlled trials labeled as pragmatic?* 2018. **16**(1): p. 49.
- 25. Loudon, K., et al., *The PRECIS-2 tool: designing trials that are fit for purpose.* 2015. **350**: p. h2147.
- 26. McReynolds, L.V., Bennett, S. *Distinctive feature generalization in articulation training.* 1972. **37**(4): p. 462-470.
- 27. Costello, J. and Onstine J. *The modification of multiple articulation errors based on distinctive feature theory.* 1976. **41**(2): p. 199-215.
- 28. Metz, D.E., Card S., and Spector, P. A distinctive-features approach to the remediation of voicing errors produced by hearing-impaired adults. 1980. **13**(3): p. 231-237.
- 29. Kummer, A. Kay Elemetrics, *Simplified Nasometric Assessment Procedures (SNAP): Nasometer Test-Revised.* 2005.
- 30. Lippke, B.A., et al., PAT-3: Photo Articulation Test. 1997: Pro-Ed.
- 31. John, A., et al., *The cleft audit protocol for speech—augmented: A validated and reliable measure for auditing cleft speech.* 2006. **43**(3): p. 272-288.
- 32. Sell, D., et al., Cleft Audit Protocol for Speech (CAPS-A): a comprehensive training package for speech analysis. 2009. **44**(4): p. 529-548.
- 33. Henningsson, G., et al., *Universal parameters for reporting speech outcomes in individuals with cleft palate*. 2008. **45**(1): p. 1-17.
- 34. International Phonetic Association. *Handbook of the International Phonetic Association: A guide to the use of the International Phonetic Alphabet*. 1999: Cambridge University Press.
- 35. Peterson-Falzone, S., et al., *The clinician's guide to treating cleft palate speech. Mosby.* 2006, Elsevier.
- 36. Klintö, K., et al., *The impact of speech material on speech judgement in children with and without cleft palate.* 2015: p. 1-13.
- 37. Pereira, V.J., et al., *Effect of maxillary osteotomy on speech in cleft lip and palate: perceptual outcomes of velopharyngeal function.* 2013. **48**(6): p. 640-650.
- 38. Fletcher, S. Contingencies for bioelectronic modification of nasality. 1972. **37**(3): p. 329-346.
- 39. Bettens, K., et al., *The Nasality Severity Index 2.0: revision of an objective multiparametric approach to hypernasality.* 2016. **53**(3): p. 60-70.
- 40. Boersma, P. and D.J.A.e. Weenink, *Praat: doing phonetics by computer, 5.3. 62.* 2014. **4**.
- 41. Altman, D.G., *Practical statistics for medical research*. 1990: CRC press.
- 42. Benjamini, Y. and Hochberg, Y. *Controlling the false discovery rate: a practical and powerful approach to multiple testing.* 1995. **57**(1): p. 289-300.
- 43. Cohen, J. A power primer. 1992. **112**(1): p. 155.
- 44. Connelly, L.M.J.M.N., *Pilot studies*. 2008. **17**(6): p. 411.
- 45. Sell, D., et al., 1 Cleft palate speech: a mixed methods evaluation of parent led, therapist supervised articulation therapy (PLAT) supported by telemedicine. 2019, BMJ Publishing Group Ltd.