Original Article

Speech Outcomes of Myanmar Children with Cleft Lip and Palate: Primary study

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Background: Multidisciplinary team approaches for cleft lip and palate (CLP), especially speech services are limited in Myanmar. Standard testing and speech services need to be established.

Objective: Our aim was to compare speech outcomes among Myanmarese children with CLP vs. normal children.

Materials and Methods: A descriptive 2-group study was conducted at Tachileik, Myanmar. Participants were recruited, including 10 Myanmarese children with CLP, who had undergone cheiloplasty and palatoplasty, and 10 normal children. The children were all students between 6 and 14 years of age in Grades 1 to 7. The standard Myanmar Articulation, Resonation, Nasal Emission and Nasal Turbulence Test and Articulation Screening Test were used for eliciting speech outcomes. Descriptive statistics were used for the demographic data, the Wilcoxon Sign Rank Test to determine articulation difference between children with and without CLP, and correlation to investigate the relationship between standard and screening tests.

Results: Ninety percent of children with CLP had functional articulation disorders. Phonological disorders, particularly voiceless for voice, was the most common type in children with CLP while 3 of the normal children had only 1 error sound. Children with CLP also had significantly more misarticulation sounds than typical children on the word, sentence, and connected speech level based on screening tests (median difference: MD = 5.5, 95% confidence interval (CI) = 4, 8; MD = 5, 95% CI = 5, 6; MD = 5.5, 95% CI = 4, 8 respectively). The Standard Burmese Articulation, Resonation, Nasal Emission, and Nasal Turbulence Test had a high correlation to the Screening Test at the word level (r = 0.81, 95% CI = 0.36, 0.95).

Conclusion: Children with CLP in Myanmar are at high risk of articulation errors and need critical speech services.

Keywords: Myanmar, Burmese, Speech outcome, Cleft palat

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Cleft lip and palate (CLP) is one of the most common birth defects. The worldwide incidence of CLP ranges between 0.11 and 1.00/1,000 live births[1-6]. In Thailand, Lao People's Democratic Republic (Lao PDR), and the Republic of the Union of Myanmar (Myanmar), the respective prevalence of CLP ranges between 0.58 and 2.4[7-9]; 0.02[10]; and, 1 and 1.25[11] per 1,000 live births.

The main treatment outcomes for CLP are good configuration, speech qualities, and quality of life (QoL). Surgical reconstruction is the 1st line of management for solving the stigma of defects. Speech qualities include articulation, resonation, intelligibility, understandability, acceptability, and voice quality. These are perceptual characteristics, which are usually compared to general or normal children in the respective society. QoL is the general well-being of individuals and societies, as outlined by the negative and positive features of the person's life. QoL— observed from life satisfaction— includes physical health, family, education, employment, wealth, religious beliefs, finance, and the environment.

The prevalence of speech abnormalities in children with CLP after CLP repair included: articulation errors (88.60%: 95% confident interval (CI) = 84.50 to 92.70); resonance disorders (43.3%: 95% CI = 36.60 to 50.00); voice disorders (12.50 to 19.10%: 95% CI = 14.26 to 24.82)[12,13]; and, delayed speech and language disorders (16.30%: 95% CI = 12.65 to 20.69).

Early diagnosis and early intervention are necessary for children with CLP. There is, however, a limited number of needed professionals for the multidisciplinary team in developing countries (i.e., plastic surgeons, orthodontists, nurse coordinators, and speech and language pathologist (SLP)). Crucially, assessment of speech and language defects in children with CLP is needed in beginning period of speech service, but speech services for children with CLP after configuration reconstruction are limited in Myanmar, just as

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in nearby Lao PDR, Vietnam, and Cambodia.

Since development of the Burmese Standard Articulation, Resonance, Nasal emission and Turbulence Test for children with CLP, speech outcomes should be assessed and a service plan provided. The objective of the current study was to explore speech outcomes in Myanmar children with CLP and to compare them to normal children.

Materials and Methods

Study design

This was a descriptive cross-sectional study conducted in Tachileik, Eastern Shan State of Myanmar. The study was reviewed and approved by the Khon Kaen University Ethics Committee for Human Research (HE 601372).

Participants

Participants for the primary study were selected by purposive sampling. We recruited 10 normal students and 10 with CLP (cleft palate with or without cleft lip) between 6- and 14 years of age, attending elementary school, living in or near Tachileik, Eastern Shan State of Myanmar. Parents provided written consent and students assent before being enrolled in the study.

Setting

The 10 children with CLP underwent speech outcome assessment at the Shwemahar Mahamuni Monastery while the 10 normal children underwent their assessments at the Golden Master Private School.

The assessment of speech outcomes was done using the Standard Burmese Articulation, Resonation, Nasal Emission and Nasal Turbulence Test. The test comprises 58 pictures including 31 Burmese sounds as words and 32 pictures as phrases or sentences. This test was developed by researchers for case validity in the final version.

For outcome measurements, each target sound test needed consensus among 4 investigators: 2 Thai speech and language pathologists with 30 years' experiences in CLP; 1 linguist expert in both Thai and Burmese with 15 years' experience in teaching and research in Burmese; and, 1 native Burmese professional with a Bachelor in Thai Language from Mae Fah Luang University fluent in both reading and writing Thai and Burmese. During the 15- to 30-minute testing period, speech outcomes were elicited from the children by naming pictures presented to them. If the children could not read or had no idea what pictures were, they were asked to repeat a reading by the Myanmar assistant. In case of any disagree on perceptual assessment among the four investigators, children were asked to name or repeat until a consensus was achieved among the investigators. These processes were done to avoid bias among the investigators.

Resonance based on standard tests was investigated and classified[14,15] as follows: within normal limits/none or normal resonance = 0; mild = 1; moderate = 2 and severe hypernasality = 3. Audible nasal air emission and/or nasal turbulence included: within normal limits/none or no deviation; intermittent or variable (i.e., some audible nasal air emission in high oral pressure consonants <4 target sounds); frequent or pervasive (i.e., audible nasal air emission in most of oral consonants ≥4 target sounds). For intelligibility, understandability, acceptability, and facial grimace, the outcomes were defined by universal parameters[14,15] as:

1) Intelligible conversational speech: Good intelligibility means others understand >75% of the speech; fair intelligibility means 50 to 75% of the conversation is understandable, and unintelligible means <50% of the speech could be understood).

2) Understandable conversational speech: 'Normal' speech is always easy to understand; 'Mild' deficit when speech is occasionally difficult to understand; 'Moderate' when speech is often difficult to understand; 'Severe' when speech is difficult to understand most or all of the time.

3) Acceptability of whole speech: 'Normal' when speech is most always acceptable; 'Mild' when speech deviates to a mild degree; 'Moderate' when speech deviates to a moderate degree; and 'Severe' when speech deviates to a severe degree.

Grimacing is an aberrant facial muscle movement and a subconscious attempt to inhibit abnormal nasal airflow by constricting the nares to reduce the air escape. The degree of grimace was assessed from the whole speech sample and was classified as: 'Normal' if within limits for normal configuration; ala = 1 – aberrant ala muscle movement; nasal bridge = 2 – aberrant bridge of nasal muscle movement; and, forehead = 3 – aberrant forehead muscle movement.

Descriptive analyses were performed for demographic data. The Wilcoxon Sign Rank Test was used to assess the median difference between the numbers of articulation errors. Correlation was used to evaluate the relationship among the tests used in this study.

Results

Both 10 typically and 10 children with CLP had general characteristics in Table 1. Each group comprised 4 females and 6 males. Children with CLP had an abnormality of the tongue and teeth, and malocclusion (Angle's Class Malocclusion). Normal participants had no abnormality of resonance while 60% of children with CLP had hypernasality. Voice and other speech perceptual outcomes are presented in Table 2. The prevalence of voice abnormality in children with CLP was 88.9% (8 of 9) while it was 60% in normal children (6 of 10). For other speech outcomes, normal children were within the normal limits while 22.2% (2 in 9) children with CLP had abnormalities from mild to severe vis-a-vis intelligibility, understandability, and degree of grimace. Only the children with CLP had audible nasal emission.

Our findings revealed that the most common type of speech disorders among children with CLP were functional articulation disorders (90% of children with CLP) followed by phonological articulation disorders, voiceless for voice sounds (50%), nasalized voice pressure consonant (30%), nasal consonant for oral pressure consonant (20%), and glottal sounds (20%). Thirty percent of the CLP children had nasal

Table 1. General characteristics of participants

No.	Gender		Age (year; month)		Language		Diagnosis		Occlusion Class**		Tongue		Teeth	
	CLP	N	CLP	N	CLP	N	CLP	N	CLP	N	CLP	N	CLP	N
1	Male	Female	11	12	Shan	Burmese	Bilat. CLP	N	III	III	N	N	Open bite	N
2	Male	Female	7	13	Shan	Burmese	CP	N	Normal	Normal	Mild tongue tie	N	Open bite	N
3	Female	Male	14	12	Lahu	Burmese	CP	N	II	III	N	N	N/A	Open bite
4	Female	Male	7	13	Burmese	Burmese	Lt. CLP	N	III	Normal	N	N	Missing#/ Open bite	Open bite
5*	Male	Female	6	10	Lahu	Burmese	Rt. CLP	N	III	III	Mild tongue tie	N	Missing#/	Open bite
6	Male	Male	11	10	Shan	Burmese	Bilat. CLP	N	III	I	Mild tongue tie	N	Open bite	N
7	Male	Female	12	8	Shan	Burmese	CP	N	II	Normal	N	N	Open bite	N
8	Male	Male	8	9	Mandarin	Burmese	Rt. CLP	N	III	Normal	N	N	Missing#/ Open bite	N
9	Female	Male	6	8	Burmese	Burmese	Lt. CLP	N	III	Normal	N	N	Missing#/ Open bite	N
10	Female	Male	10	7	Burmese	Burmese	Rt. CLP	N	N/A	Normal	N	N	Missing#	Missing#/ Open bite

CLP = Cleft lip and palate; N = Normal; Bilat. CLP = Bilateral cleft lip and palate; CP = Cleft palate; Rt. CLP = Right cleft lip and palate; Lt. CLP = Left cleft lip and palate
* Child with cleft lip and palate who had mother with right cleft lip; ** Angle's class malocclusion; # Missing upper front teeth

emission. Only 3 of the normal children had any articulation type of functional articulation disorder. The number of articulation errors are presented in Figure 1.

Comparing sentences in the Standard Burmese Articulation, Resonation, Nasal Emission and Nasal Turbulence Test and the screening test between children with CLP and normal children, children with CLP had significantly greater number of articulation errors (Table 3). The correlation between the Standard Myanmar Articulation, Resonation, Nasal Emission and Nasal Turbulence Test and the word and sentence levels and articulation screening test Spearman's rho (r) was 0.81 (0.36, 0.95); 0.44 (0.27, 0.83); and, 0.69 (0.12, 0.92), respectively.

Discussion

The speech outcomes of 10 children with CLP and 10 normal children were compared. The results revealed that children with CLP over against normal children had more abnormalities of occlusion, tongue, and teeth. Eight of nine children with CLP (88.9%) had malocclusion compared to 40% (4 in 10) in normal children. Being tongue-tied was 30% (3 in 10) in children with CLP compared to none

Table 2. Speech perceptual assessment in children with CLP

No.	Hypernasality		Voice**	Intelligibility*	Understandability*	Acceptability**	Grimace
	W	S					
C01	Moderate	Moderate	Ab	WNL	WNL	Mild	0
C02	Mild	Mild	Ab	If topic know	Mild	Moderate	0
C03	Moderate	Moderate	Ab	WNL	WNL	Mild	0
C04	WNL	WNL	Normal	Unintelligibility	Moderate	Severe	0
C05	WNL	WNL	Ab	WNL	WNL	Mild	N/A
C06	WNL	WNL	N/A	N/A	N/A	N/A	N/A
C07	Mild	Mild	Ab	WNL	WNL	Mild	N/A
C08	Mild	Mild	Ab	WNL	WNL	Mild	+2
C09	Mild	Mild	Ab	WNL	WNL	Moderate	+1
C10	Moderate	Moderate	Ab	WNL	WNL	Mild	0

W: Word; S: sentence; 0: within normal limit; WNL: Within normal limit; Ab: abnormal
* Assessment from conversational speech;** Assessment from whole speech sample; +1: ala; +2: nasal bridge N/A: Not available

No	Words		Sentences		Screening	
	Cleft lip and palate	Normal	Cleft lip and palate	Normal	Cleft lip and palate	Normal
1	w/hw, l/hl, t, l /θ, c/j, s/z, m/hm (6)	(0)	l/hl, t/θ, c/j, k/g, s/z (5)	(0)	θ/t; w/hw, m/b, l/d, c/j, s/z, p/b, k/g, l/hl, w/hw (10)	(0)
2	w/hw; A/-p; p,w/b; t/θ; l/d; c/j; k/g; s/ch; s/sh; s/hs; s/z (11)	(0)	l/y; p, w/b; t/θ; t/d; c/j; k/g; s/ch; s/sh; s/hs; s/z; u:i/ e (11)	(0)	c/j, s/sh, A/- ŋ; s/z; t^{s} s/ch; k/g; t/d o:i/o (8)	(0)
3	l,l/hl; t,l/θ; c/ɛ(3)	(0)	w/hw; t,l/θ; n/hn a/a:u; ʊa/ʊʊ e/ ɤ:i (5)	(0)	s/sh; w/hw, m/hm (a:/a:u) (4)	(0)
4	hw/w; n/l, hn/hl; m/b; p, m/p^{h}; ?/t; t, ?/ θ; ?/t^{h}; c/d; ɲ/c; ɲ/j; ?/g; ŋ/k; ŋ/k^{h}; ɲ/ch; ɲ/sh; t/s; ɲ,?/hs; ɲ/z (19) dental lisping	ŋ/hŋ (1)	hw/w; ɲ/y; n/l; hn/hl; m/b; m/b; p/p^{h}; t,c/θ; t,t?/t^{h}; c/d; c?/c; ɲ/j; ɲ/y; ŋ/g; ŋ/k; ŋ/k^{h}; c,ŋ/ch; c, ɲ/sh; ct/s; c?/hs; c?/z (21) dental lisping	(0)	ɲ/j,d,ch; ?/z, hs,sh; ?/s; p/t; t/t^{h}; y/ɲ; m/b; c/ch,; ŋ/k^{h}, p/p^{h}; n/hl; k/g (16) dental lisping	(0)
5	t/θ; c,cy/j; y/z, s/sh, (4)	(0)	cy/j; k/g, s/sh, o:u/o; e/ɤ:i (5)	(0)	y/j,z; s/z; s/sh; l/hl; k/g; l/y (6)	(0)
6	t/θ, c/j, k/g, s/z, m/hm (5)	p^{h}w/hw (1)	t/θ, c/j, k/g, ch/sh, s/z (5)	(0)	c/j, s/z, y/ɲ, k/g (4)	(0)
7	p^{h}w/hw, l/hl, p/b, t/θ, t/d, c/j, k/g, s/z (8)	(0)	t/θ, t/d, c/j, k/g, s/z (5)	(0)	s/z, c/j, m/hm, p/b, ɲ/hŋ, k/g, ɲ/y (7)	(0)
8	t/d, p/b, c/j, k/g, s/z, (5) dental lisping	(0)	l/hl, p/b, t/d, c/j, k/g, s/z (6) dental lisping	(0)	s/z, c/j, p/b, k/g (4) dental lisping	(0)
9	p/b; t/d; c/j; k/g, s/ch, s/sh, s/z (7) dental lisping	(0)	p/b, t/d, c/j, k/g, s/sh, s/z (6) dental lisping	(0)	c/j; s/sh; s/z; p/b; k/g (5) dental lisping	(0)
10	k^{h}/t^{h}, ?c/c, sh/ch (3)	t/d (1)	k^{h}/t^{h}, l/hl, ?c/c, sh/ch, s/hs (5)	(0)	(0)	(0)
Mean (SD) 95 % CI	7.10 (4.80)	0.3 0 (0.50)	7.40 (5.1)	0 (0)	6.40 (4.30)	(0)
Median (Min: Max) 95 % CI	5.50 (3:19)	0 (0:10)	5 (5:21)	-	5.5 (0:16)	-

Figure 1. Number of articulation errors.

Table 3. Median difference in number of articulation errors between children with CLP and normal children

Words tests		Sentence tests		Screening tests	
Median difference	95% confidence interval	Median difference	95% confidence interval	Median difference	95% confidence interval
5.5	4, 8	5	5, 6	5.5	4, 8

of the normal children. All of children with CLP had abnormal teeth compared to 40% (4 in 10) of the normal children (Table 1). So, even though normal children can have occlusion abnormalities, children with CLP had a higher risk[16,17].

The prevalence of resonance disorders in Burmese children with CLP both at the word and sentence levels was 70% (7 in 10) (Table 2), suggesting a high risk for velopharyngeal insufficiency (VPI) as has been reported elsewhere after palatoplasty (range, 30 to 100%)[13,18-22]. Audible nasal emission both at the word and sentence levels was 10% compared to the 16.7 to 100% reported in other studies[21, 23]; possibly because some Myanmarese phonetics have nasal emission (i.e., /hl/, /hs/, /hm/)[24].

Our findings suggest that voice disorders in Burmese children with CLP are high (88.9% or 8 in 9) as are rates in normal children (60%; 6 in 10). Both rates are higher than previously reported (viz., 5.5 to 43% in children with CLP[13,22,25-28] and 6 to 11% normal children)[29]. Both normal and those with CLP had higher prevalence of voice disorders than general voice disorders that found prevalence was 6 to 9%[30] in normal and 12 to 43% in CLP[27]. Possibly, Burmese children might be over use of voice from playing culture that normally talk very loud in playground, suggesting a need for attention to vocal hygiene. Further investigation is needed to clarify this finding and its cause.

When considering the relationship between resonance disorders and the characteristics of intelligibility, understandability, and acceptability (Table 2), three children with CLP had moderate hypernasality (No. 1, 3, and 10), which had a demonstrably negative effect on understandability and acceptability. Two of the children with CLP had mild hypernasality: No. 8 had a mild degree of deviation in acceptability and nasal bridge muscle contraction; and, the other (No. 9) had intermittent audible nasal emission, a moderate degree of deviation in acceptability, and ala muscle contraction. The two cases provide evidence that facial muscle contraction or grimacing is a compensatory mechanism for prevention air flow to nose[31].

There were many articulation errors in children with CLP and a high degree of hypernasality such as in child No. 2 and 4 (Figure 1). Most of the errors were compensatory articulation disorders (CAD), supporting the theory that compensatory articulation errors are caused by VPI, particularly in children with (a) hypernasality, (b) nasal air escape, (c) weak intraoral air pressure during oral sound production, and (d) typical location of production[32,33]. Relatedly, in regard to dialects, there were no voiced sounds in some languages (e.g., Thaiyai); so, children with CLP (No. 1, 2, 6, and 7), whose mother languages had no voiced sounds in the phonetic system had difficulty producing voiced sounds such as /z/, /g/, /j/. This phonological disorder was very common among Burmese children with CLP.

Children with CLP also had significantly more articulation errors than normal children (Table 3), implying that residual abnormalities of structure—especially VPI (No. 1, 3, 10) or resonance abnormality—was not directly related to the negative results in the number of articulation errors. Compensatory articulation disorders after palate repair are common problems in children with CLP and need prompt speech therapy. Several Speech Therapy Model should be considered for these children (e.g., Community-Based Speech Model[22,23,34-40] and speech summer camp[41-44]). There are not, however, sufficient speech services in Myanmar.

There was a high correlation between the Standard Burmese Articulation, Resonation, Nasal Emission and Nasal Turbulence Test and the Articulation Screening Test at the word level ($r = 0.81$). Articulation Screening Test can thus be used as an indicator of the Standard Test at the word level and so also appropriate for screening in CLP children in Myanmar. These findings represent a first study and further research is needed to confirm the results as the sample size limited.

Conclusion

This was the first report on children with CLP in Myanmar. Results indicate that after surgical treatment, speech abnormalities including articulation errors, resonance disorders, negative understandability, and acceptability remain, requiring speech services for better outcomes like prevention of negative sequelae (e.g., illiteracy, poor self-image, and poor QoL).

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What is already known on this topic?

The characteristics of speech defects in children with CLP after primary surgery have been studied, and speech abnormalities among children with CLP are common after palatoplasty. Speech services in Myanmar are lacking owing to insufficient numbers of professionals.

What this study adds?

After a primary palatoplasty, Myanmar children with CLP have speech defects including articulation errors, resonance disorders, and poor understandability and acceptability. Speech services are needed for the better speech outcomes.

Potential conflicts of interest

The authors declare no conflicts of interest.

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