

Speech Outcomes in Children with Cleft and Palate: Srinagarind Hospital, Khon Kaen University, Thailand

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Background: Speech defects in children with cleft lip and palate are common after cleft repair. Investigation of speech outcomes provides useful information for further intervention planning and protocol reviewing.

Objective: The aim of the present study was to determine speech outcomes in children with CLP, compared to normal children between 6 and 13.

Materials and Methods: This was a cross-sectional study. Fifteen children with cleft palate, with or without cleft lip (CLP) and 15 normal children between 6 and 13 were enrolled. Perceptual assessment via Thai Speech Parameters for Patients with Cleft Palate in a Universal Reporting System for identification and classification speech outcomes was performed by two senior speech and language pathologists by consensus. Descriptive analysis and Wilcoxon Sign Rank Test were used to present the data.

Results: The prevalence of speech abnormalities in primary school-aged children with CLP was high (viz., articulation errors (100%), abnormalities of resonance (hypernasality: 44%), audible nasal emission/turbulence (44 to 60%), voice disorders (27%), intelligibility (36%), less understandability (60%), deviation acceptability (87%). Children with CLP had more speech abnormalities of both types and numbers than normal children (median difference = 9; 95% confident interval = 7, 10).

Conclusion: Speech abnormalities in primary school-aged children with CLP were common, including: CAD, hypernasality, voice disorders, audible nasal emission/turbulence, less understandability, and deviation acceptability.

Keywords: Speech outcomes, Cleft lip and palate, Primary school, Speech abnormality

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Cleft lip and palate (CLP) or orofacial cleft are the most common congenital malformations. In Thailand, the prevalence of CLP ranges between 0.58 and 2.49 per 1,000 live births⁽¹⁻³⁾ compared to the worldwide incidence that ranges between 0.11 and 2.00⁽⁴⁻¹⁰⁾. The most common stigmas after surgery include various speech abnormalities⁽¹¹⁻¹⁴⁾, psychological disorders⁽¹⁰⁾, economic stress⁽¹⁵⁾, educational or literacy deficits^(10,16,17), and overall poor quality of life (QoL)⁽¹⁸⁻²²⁾.

After the primary surgery, the common speech defects among children with CLP include: articulation errors and phonological disorders (79.70 to 88.56%)^(11,13); resonance disorders (43 to 69%)^(11,13); voice disorders (12.5 to 19.13)^(13,23); and delayed speech and language disorders (16.33)⁽¹³⁾. Taken together, these errors and disorders result in reduced intelligibility⁽²⁴⁾ and poorer QoL⁽²⁵⁾.

The Center of Cleft lip and Cleft Palate and

Craniofacial Deformities, Khon Kaen University in association with the Tawanchai Project was established in 2006. The protocol for interdisciplinary approaches in children with CLP includes providing primary surgery for chieloplasty around the age of 3 months and palatoplasty at 1 year⁽²⁶⁾. Treatment outcomes are considered from various perspectives so as to achieve the best quality of care for each patient. An investigation of five-year speech outcomes and hearing in children with CLP at the Tawanchai Center has already been done^(14,27), so the aim of the current study was to determine speech outcomes in children with CLP compared to normal non-CLP children between 6 and 13.

Materials and Methods

Study design

This was a cross-sectional prospective study. The research protocol was reviewed and approved by the Central Research Ethics Committee (CREC Number: CREC038/58BPM, Certificate number: COA-CREC019/2526).

Setting

Speech Clinic, Srinagarind Hospital, Faculty of Medicine, and Nonmuang Primary School, Muang District, Khon Kaen.

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Participant selection

Participants were children with cleft palate, with or without cleft lip (CLP) registered in the Speech Clinic, Srinagarind Hospital, Faculty of Medicine, Khon Kaen University, and normal non-CLP children attending primary school in Khon Kaen province.

Inclusion criteria

For a comparison of speech outcomes, we included (a) children between 6 and 13 with CLP registered for treatment at the Speech Clinic, Srinagarind Hospital, Faculty of Medicine, Khon Kaen University; and, (b) normal non-CLP children between 6 and 13, in grades between 1 and 6 at Nonmuang Primary School, randomly selected by teachers.

Exclusion criteria

We excluded children with CLP and normal non-CLP children with health conditions that affected speech and/or language skills (i.e., delayed speech and language development, common cold, any syndrome that negatively affected development like Treacher Collins Syndrome, Global delayed development, Autism, or Attention Deficit Hyperactivity disorders).

Children between 6 and 13 years were enrolled (a) 15 with CLP and (b) 15 normal non-CLP.

After parents gave written consent and the children verbal assent, general characteristics and information were gathered through recorded interviews. An examination of the ears, nose, and throat was performed by an otorhinologist to screen for any diseases that might cause any deviation to normal speech and language outcomes. The Thai Early Language Milestone and the UTAH test for Language Development were used to screen for delayed speech and language development.

Tools for assessment of speech outcomes included the Standard Thai Articulation, Resonation, Nasal Emission and Nasal Turbulence Test. The test was developed by the principle investigators and their colleagues⁽²⁸⁾.

Each child was asked to name, read, or repeat to an investigator (the corresponding author)'s naming in case of she/he could not name or read pictures of target sounds, both words and sentences test by herself/himself. Two senior speech and language pathologists (SLP), who have experience in CLP more than 30 years, were investigators and independently did perceptual assessment and classified type of articulation errors for cleft speech types. If there was any disagree on perceptual assessment between 2 investigators, child was asked to name or read or repeat to an investigator (corresponding author)'s naming and discussed until having consensus between 2 investigators.

Resonance from the standard test was investigated and classified using universal criteria^(28,29) 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 were set as: within normal limits – none or no deviation; intermittent or variable – some audible nasal air emission in high oral pressure consonants <4 target

sounds; or frequent or pervasive – audible nasal air emission in most of oral consonants ≥ 4 target sounds. As for intelligibility, understandability, acceptability, and facial grimace, the outcomes were defined by universal parameters^(28,29) as follows: intelligibility from conversational speech – good when >75% of conversation understandable; fair – understandable when topic known; or, mean when 50 to 75% of conversation understandable; and, unintelligible when <50% of conversation understood. Speech understandability from conversational speech as follows: good = most always easy to understand; mild = occasionally difficult to understand; moderate = often difficult to understand; and, severe = difficult to understand most or all of the time. Speech Acceptability from whole speech sample: good = normal; mild = mild deviation; moderate = moderate deviation; severe = severe deviation.

Grimacing is an aberrant facial muscle movement and a subconscious attempt to inhibit abnormal nasal airflow by constricting the nares. Facial grimace from a whole speech sample—especially for those with high oral pressure sounds—within normal limits for normal configuration are: 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 presenting demographic data. The Wilcoxon Signed Rank Test was used to assess differences among medians vis-a-vis articulation errors. Correlation was used to analyze the relationship among tests.

Results

The general characteristics of the 15 typical and 15 children with CLP are presented in Table 1. The intra-group characteristics were comparable. Children with CLP had significant abnormalities of the tongue (tied), teeth, and bite (malocclusion) (Angle's Class Malocclusion) more than normal.

As for resonance disorders, normal children had no evidence of resonance abnormality. Children with CLP had both resonance disorders and audible nasal emission (Table 2). The incidence of hypernasality was 43%, most of whom had mild hypernasality (36%). Audible nasal emission was 60%, most of whom were of an intermittent level (50%).

Voice, intelligibility, understandability, acceptability, facial grimace, speech outcomes are presented in Table 2. Twenty-seven percent of children with CLP had voice abnormality; 36% had deviation of intelligibility or intelligibility if the topic of conversation were known; 60% had abnormality of understandability; and 97% of the whole speech sample was unacceptable. Face muscle constriction for facial grimace occurred in 33%. There was no speech abnormality related to voice, intelligibility, understandability, acceptability, or facial grimace among the typical non-CLP children.

The number of articulation errors was counted at both the word and sentence levels. In case there was any sound that had the same error at both the word and sentence

Table 1. Characteristics

Variables	Normal		Children with CLP	
	Number	%	Number	%
Sex				
Male	7	46.67	6	40
Female	8	53.33	9	60
Age				
Mean (SD)	8.33	(1.632)	9	(1.9639)
Cleft type				
Rt CLP	-	-	3	20
Bilateral CLP	-	-	6	40
Lt CLP	-	-	2	1.33
CP	-	-	4	26.67
Occlusion**				
Normal	13	92.85	-	-
Malocclusion class	-	-	-	-
Class I	-	-	-	-
Class II	1	6.67	-	-
Class III	-	-	13	100
N/A®	1	-	2	-
Home language				
Central or official	6	40	-	-
Northeast	6	40	15	100
Northeast & central or official	3	20	-	-
Tongue				
Normal	15	100	13	86.67
Tongue tied	-	-	2	13.33
Teeth				
Missing upper teeth	4	26.67	1	6.67
Open bite	3	20	3	20
Cross bite	1	6.67	-	-
Missing upper teeth & open bite	-	-	11	73.33
Normal	7	46.67	-	-

® Children with cleft lip and palate whose mother had right cleft lip, ** Angle's Class Malocclusion, # of missing upper front teeth
 Rt CLP = Right cleft lip and palate; Lt CLP = Left cleft lip and palate;
 Bilat. CLP = Bilateral cleft lip and palate

levels, it was counted as a articulation error. The types of articulation errors are presented in Figure 1. Children with CLP had more articulation defects than normal children.

The type of articulation errors in children with CLP and normal children were classified per standard guidelines^(28,29). Similar to the number of articulation errors, the types of articulation errors were classified as word and sentence levels. If a child had the same type of articulation error in both word and sentence levels, it was considered one type (e.g., if a child had [t^h], [k^h/t^h] at word level it was a velar type; vs. [t^h], [k^h/t^h] at sentence level it was a velar type. The type of articulation error was classified to be a velar type).

Classification of the types of articulation errors in children with CLP and normal children are presented in Table 3. The Velar and Trill error were the most common

patterns in both children with CLP and normal children. Two of the normal children had only an articulation type of functional articulation disorder: trill error.

Comparison of the number of articulation errors between children with CLP and normal children was done using the Wilcoxon Sign Rank Test. Children with CLP were poorer than normal children. Children with CLP had significantly more articulation errors than normal children (i.e., the relative proportion of articulation errors in children with CLP to normal children was 10 to 1 and the mean difference was 9; the 95% confidence interval was 7, 10).

Discussion

The children studied—those with CLP and normal children—had a similar background, sex, and age (Table 1). Two children (13.3%) were tongue tied. All of the children with CLP and a half of the normal children had an abnormality of teeth and malocclusion, which agrees with previous reports that children with CLP had a high risk for malocclusion and teeth abnormalities^(30,31).

Perceptual assessment for resonance (Table 2) revealed the prevalence of resonance disorders in children with CLP both at the word and sentence levels was 43% compared to none in normal children. This finding underscores the high risk for velopharyngeal insufficiency (VPI) and supports a previous reports that found the prevalence of hypernasality after palatoplasty in Thailand ranged between 37.5 and 43.3%^(13,32,33). Audible nasal emission prevalence (i.e., 60% at the word level and 45% at the sentence level) was higher than hypernasality. Audible nasal emission is the abnormal passing of oral air through a palatal cleft, or from some other type of velopharyngeal inadequacy (VPI). For example, during the production of a consonant a buildup of oral air pressure is required for proper pronunciation (e.g., /p/ or /s/). Nasal turbulence, also called a nasal rustle, is due to air pressure being forced through a small velopharyngeal opening. As the air pressure goes through, there is friction and bubbling of secretions above the opening. Our data show that audible nasal emission or nasal turbulence can occur without hypernasality.

For other speech outcomes (Table 2), voice disorders in CLP was 27% (3 in 11) compared to none in normal children. In previous studies, this rate ranged between 5.5 and 43%^(13,33-37) vs. none in normal children, supporting the theory that voice disorders or hoarseness is laryngeal compensation for abnormal velopharyngeal valving. VPI causes difficulty with articulation of specific consonants owing to the inability to build sufficient air pressure in the oral cavity. Compensatory mechanisms to increase air flow to correct articulation—in particular for stops, fricatives, sibilants, and affricates—is implicated in vocal cord abnormalities and voice disturbances. A more recent study, however, did not find any relationship between hoarseness in patients with cleft palate and VPI, calling into question the theory of laryngeal compensation as the source of hoarseness⁽³⁸⁾. There is no identified definitive cause of hoarseness, suggesting its cause is multifactorial or attributable

Table 2. Resonance and speech outcomes based on a perceptual assessment among CLP

No.	Hypernasality		Audible nasal emission Nasal turbulence		Voice**	Intelligibility*	Understandability*	Acceptability**	Facial grimace
	Sentences		Word						
	Words	Sentences	Word	Sentence					
1)	Mild	Mild	WNL	WNL	WNL	WNL	WNL	WNL	None
2)	Mild	Mild	Invasive	Invasive	Abnormal	If topic is know	Mild	Mild	Ala
3)	WNL	WNL	N/A	N/A	N/A	WNL	Mild	Mild	None
4)	WNL	WNL	N/A	N/A	Abnormal	If topic is know	Severe	Severe	None
5)	Moderate	Moderate	WNL	WNL	WNL	If topic is know	Moderate	Moderate	Nasal bridge
6)	WNL	WNL	Intermittent	Intermittent	N/A	N/A	Moderate	Moderate	None
7)	WNL	WNL	Intermittent	WNL	N/A	WNL	Mild	Mild	None
8)	Mild	Mild	N/A	Intermittent	WNL	If topic is know	Mild	Mild	None
9)	WNL	WNL	WNL	WNL	WNL	If topic is know	Mild	Mild	None
10)	Mild	Mild	Intermittent	Intermittent	WNL	WNL	Mild	Mild	Ala
11)	WNL	Mild	Intermittent	Intermittent	WNL	WNL	WNL	WNL	None
12)	WNL	WNL	WNL	WNL	WNL	WNL	WNL	Mild	None
13)	WNL	WNL	N/A	N/A	WNL	WNL	WNL	Mild	Nasal bridge
14)	Mild	WNL	Intermittent	Intermittent	N/A	WNL	WNL	Mild	Ala
15)	N/A	N/A	N/A	N/A	Abnormal	WNL	WNL	Mild	None
Total				Total					
Mild	5 (36%)	5 (36%)		WNL/None	8 (73%)	9 (64%)	6 (40%)	2 (13%)	10 (67%)
Moderate	1 (7%)	1 (7%)		Abnormality/ If topic known	3 (27%)	5 (36%)			
Intermittent					4	1			
Invasive				4 (36%)					
WNL	8 (57%)	8 (57%)	5 (50%)	1 (9%)			6 (40%)	10 (67%)	3 (20%)
			1 (10%)	-Mild/ala			2 (13%)	2 (13%)	2 (13%)
			4 (40%)	-Moderate/ nasal bridge					
N/A	1	1	5	4			1 (7%)	1 (7%)	-

WNL = Within normal limit; N/A = Not available

* Assessment from conversational speech; ** Assessment from whole speech sample

Patient Number	Children with CLP	Number of Articulation errors	Normal	Number of Articulation errors
1	[f,h/p ^h] [s,t ^h /t ^h] [t ^h /k ^h] [t ^h /s] [p/f] [p,p ^h /b] [t,?,t ^h /d] [t ^h ,/?/t] [k/t ^h] [l/r] [-p/-t]	11	[l/r]	1
2	[s/t ^h ,t ^h] [t ^h /k ^h ,s] [m,p ^h /b] [ŋ,n/d] [p,?,k,/p] [t ^h ,/?/k/t] [t ^h ,/?/t ^h] [/?/k] [ŋ/m,n] [l/r]	10	[l/r]	1
3	[t ^h /p ^h] [s/t ^h] [t ^h /s] [w/f] [p/b,k] [t/d,p] [k/t ^h] [d/n] [t ^h /j] [l/r] [-p/-t]	11	[l/r]	1
4	[t,k,k ^h /p ^h] [k ^h ,k/t ^h] [k ^h /t ^h] [k/s,d,t,t ^h] [kw,pk/f] [p/b] [ŋ/n] [ŋ,l,j/r] [-p/-t] [-k/-t] [-ŋ/-n] [-m/-ŋ]	12	[l/r]	1
5	[t ^h /k ^h] [m/b] [n/d,w] [p/?/p] [?,k/t] [/?/t ^h ,k] [ŋ/n] [l/r]	8	[l/r]	1
6	[p ^h ,k ^h /t ^h], [s/t ^h], [k ^h ,t ^h /k ^h], [k ^h /s], [p/f], [n/b] [t ^h ,k,k ^h /d], [k/t,t ^h], [ŋ/n] [n/l], [ŋ,d,ŋk/r], [-k/-t]	12	[l/r]	1
7	[k ^h /t ^h] [k/d] [k,tk/t] [b/m] [t ^h ,l/r] [-k/-t]	6	[l/r]	1
8	[t ^h /p ^h] [k ^h /t ^h ,t ^h] [k/s,d,t] [k,w/f] [k,kt ^h /t ^h] [ŋ/n,l,j] [l,ŋ/r] [-k/-t] [-ŋ/-n] [-n/-ŋ]	10	[l/r], [s/t ^h]	2
9	[?,b/p ^h] [k ^h ,/?/t ^h] [/?/k ^h] [k,l,t ^h ,t ^h /s] [w,p ^h /f] [p/b] [k,dk/d] [b/p] [/?/k] [ŋ/n] [l/r] [k/?] [O/-k,-n,-ŋ] [-k/-t]	14	[l/r]	1
10	[k ^h /p ^h ,t ^h] [t ^h ,k ^h /t ^h] [t ^h ,k/s] [k/f] [m/b] [n,k,k ^h /d] [/?/p] [k,/?/t] [?,d,j/t ^h] [?,k/?/k] [b/m] [ŋ/n] [/?w/w] [n,l/j] [l/r] [-m/-ŋ]	16	[l/r]	1
11	[k ^h /t ^h] [l/r]	2	[l/r]	1
12	[k ^h /t ^h],[s/t ^h],[t ^h /k ^h],[k ŋ/d],[t ^h ,/?/t],[/?/t ^h],[/?/k],[kd/l],[O/-k],[l/r],[-k/-p]	11	[l/r]	1
13	[l/t ^h],[l/s],[w/f],[/?/b],[k/d],[k/t],[ŋg/n],[l/r]	8	[l/r]	1
14	[s/t ^h],[t ^h /k ^h],[kd/d],[k,kt/d],[ŋ/n],[l/r]	6	[s/t ^h],[t ^h /s], [h,/?/p ^h], [k ^h /t ^h], [p/b], [l/r]	6
15	[p ^h ,k ^h /t ^h],[w/f],[ŋg/n],[ŋg/l],[l/r],[k ^h /h]	6	[l/r]	1

Figure 1. Number of articulation errors.

to some other under-recognized factor. Some patients with hoarseness and cleft palate are treated with voice therapy and anti-reflux treatment, the benefit of these interventions is also unknown⁽³⁶⁾.

Sixty percent of children with CLP had less understandability of conversations, and 87% of them had deviation in acceptability of whole speech. These data suggest that even with a high percentage of deviation of

Table 3. Type of articulation disorder

Type of articulation disorder	Children with CLP		Normal	
	Number	%	Number	%
Within normal limit	1	1.61	3	17.56
Pharyngeal	1	1.61	-	-
Glottal	5	8.06	-	-
Mid-dorsal palatal	0	0	-	-
Velar	14	22.58	-	-
Phoneme specific	-	-	-	-
Not Phoneme specific	-	-	-	-
Nasal consonant for oral pressure consonant	3	4.83	-	-
Nasalized pressure consonant	4	6.45	-	-
Weak oral pressure	-	-	-	-
Developmental articulation	1	1.61	-	-
Phonological error	-	-	-	-
Functional/other oral misarticulation	11	17.74	2	11.75
Dental lisping	-	-	-	-
Organic articulation disorder	-	-	-	-
Co-articulation	7	11.29	-	-
Trill	12	19.35	12	7.58
Gliding	2	3.23	-	-
Lateralize	1	1.61	-	-

speech acceptability, whole conversation understandability was fair; due to factors such as context, topic, and/or environment.

As for the number of articulation errors (Figure 1), children with CLP had significantly more articulation defects than normal children [median difference = 9; 95% confident interval = 7, 10;]. These results confirmed the occurrence of compensatory articulation disorders due to VPI or anatomical defects. Compensatory articulation disorders (CAD) are learned articulation errors. They are errors in place of articulation or direction of airflow; due to the inability to generate adequate intra-oral air pressure for consonant production, anterior structural anomalies, abnormal auditory-perceptual learning, or due to the anatomical inability to close the VP port, which can persist even after successful physical management of the VP mechanism or other factors. Regarding the type of articulation errors (Table 3), the velar type was the most common in children with CLP as was reported study⁽³⁹⁾. Besides functional articulation, other oral articulations and trill, our results are similar to a previous finding, which found glottal and pharyngeal productions were the latter common CAD. Meanwhile, the current study revealed that glottal, nasalized voice pressure consonants, and nasal for oral pressure consonants were common. These results also supported a previous study, which proposed that the productive phonological processes were consonant backing, final consonant deletion, gliding, and stopping⁽⁴⁰⁾. Interestingly, trill and functional articulation disorders were more prevalence in children with CLP as with previous studies^(13,33,39,41,42). They might reflect that Thai school might focus less on trill or flap. The current study also revealed that children with CLP had significantly more articulation

errors than normal children. This finding agrees with previous studies and the theory of the compensatory mechanism of air leakage into nasal cavity result in CAD and other speech abnormalities.

Taken together, the prevalence of speech abnormalities in children with CLP remains high including articulation error (100%), resonance disorders (hypernasality: 44%), audible nasal emission/turbulence (44 to 60%), voice disorders (27%), intelligibility (36%), less understandability (60%), and deviation acceptability (87%). This information might prove useful for further research and protocol review⁽¹⁴⁾.

Conclusion

Speech abnormalities—including CAD, hypernasality, audible nasal emission/ turbulence, less understandability, and deviation acceptability—remain common such that critical speech intervention and review protocols are needed for better speech outcomes and improved quality of life.

What is already known on this topic?

Speech abnormalities are common among children with CLP after chielolasty and palatoplasty. The prevalence of which various between centers, institutions, and nations. Studies on speech defects among children with CLP in primary school in Thailand need to be done.

What this study adds?

Primary school-aged children with CLP had high CAD, hypernasality, audible nasal emission/ turbulence, less understandability, and deviation acceptability.

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Potential conflicts of interest

The authors declare no conflicts of interest.

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