Speech Disorders in Operated Cleft Lip and Palate Children in Northeast Malaysia

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SUMMARY

The aim of this study was to determine the prevalence and association of speech disorders among operated cleft lip and palate children (CLP) in Northeast Malaysia. A comparative cross sectional study was performed on 98 operated CLP and 109 non-cleft subjects that aged between 3-12 years old. Data collection was done clinically and also by recording speech samples of each subject from both groups using a portable cassette recorder. Results showed that the prevalence of speech abnormality was 61.2% (95% CI: 51.41-71.04) and the risk of having speech abnormality was 174.5 times (95% CI: 23.04, 1320.67; P value<0.001) in CLP children compared to non-cleft children. Therefore it was found that children with appropriately repaired CLP in Northeast Malaysia failed to have normal speech.

KEY WORDS:
Cleft lip and palate, Speech disorders, Northeast Malaysia, hypernasality, hyponasality

INTRODUCTION

Cleft lip and palate (CLP) deformity has become a major public health problem affecting one in every 500 – 1000 births worldwide[1]. It is the fourth most common birth defect and the most common congenital defect of the craniofacial region. The incidence varies with racial background and it is usually quoted as one in every 750 live births[2]. Jensen *et al.* in Denmark noted a significant increased in cleft since 1942. He reported the study done by Fogh-Anderson in 1942 who noted the incidence of 1.5 per 1000 (1 in 667) live births, increasing to 1.75 per 1000 live births in 1971 and 1.89 per 1000 live births by 1981[3]. In Malaysia, results of The National Oral Health Surveys (NOHS) indicated an increased incidence of CLP, with varying occurrences from one in 1006 to one in 941 live birth[4, 5]. The number of studys registered at the Hospital USM Combined Cleft and Craniofacial Deformity Clinic (Combined clinic) from 1997 to 2000 was 760 patients[6].

Children with CLP frequently demonstrate speech and resonance disorders following primary surgical repair of the palatal cleft[7]. Speech is a motor component of our communication, which requires intact structures of lips, jaw, tongue, teeth and palate working in coordination with muscles of respiration and phonation. Indeed the four substructures of speech are respiration (our breathing), phonation (the sound that is made by the vocal folds), articulation (the production of sounds using the lips, teeth, tongue and jaw movements) and resonation (which are the quality of voice regulated by the integrity and the movement of the soft palate and surrounding structures).

Children born with CLP are at risk for resonance, articulation and expressive language problems that may impair communication permanently. The opening at the palate creates a communication between the nasal cavity and the oral cavity, thus sounds which must come out directly through the mouth may be greatly distorted or just impossible for the child to make. The impact of palatal cleft may be evident during early vocalizations of babies before surgical management and may persist long after an adequate oropharyngeal mechanism has been established. Various studies describing the early phonological development of children with cleft palate. Chapman and Hardin in their cross-sectional study on phonological process usage in 2, 3, 4 and 5 year old cleft and non cleft children showed mildly increased use of phonological processes in cleft children up to 4 years of age. However, at 5 years of age, there was no significant difference among the groups[7].

This current study provides information on speech disorders in CLP children and their non cleft peers in our population. Speech disorders are clinically important to ensure appropriate management of communication difficulties and awareness of the possible consequences of the disorder on literacy. Therefore, the objective of this study was to investigate the prevalence of speech disorders among the 3 to 12 years old CLP group and normal non cleft group in our children population looking into hypernasality, hyponasality, Cul-de-sac resonance, and articulation speech.

MATERIALS AND METHODS

This is a comparative cross sectional study involving non-syndromic CLP children as the study group while the control group comprised of non cleft children. The source population for study group was all registered CLP children who attended the Combined Cleft and Craniofacial deformity Clinic at Kota Bharu dental clinic while non cleft group were non cleft children attending out patient clinic at Kota Bharu dental clinic in six months duration. The inclusion criteria for study group was operated non-syndromic CLP children. The children with cleft lip only were excluded from this study.

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Patients in both study and control group were Kelantanese that aged between 3 and 12 years old. The Kelantanese children were selected because the speech assessment was based on the method used by Wan Zaharah *et. al.*, where the phonological assessment was designed specifically to cater for the Kelantanese children[8].

The CLP and non cleft children were examined by Otorhinolaryngologist and the hearing status were determined by using otoacoustic emmission and tympanometry. Only those who have normal hearing were included. The justification for exclusion was based on the study done by Schonweiler *et al.,* who stated that speech and language function in CLP patients were predominantly related to the hearing status[9]. The children in both groups who have other dental or maxillofacial abnormalities and other systemic diseases that can cause speech pathologies were also excluded from the study. The subjects in both groups were selected by simple random sampling.

The sample size was calculated using two proportion formula. Setting the power of 80% at alpha error of 0.05 with detectable difference between the two proportions of 15%, we obtained our sample size of 97 subjects per group. Thus, with anticipation of 20% non response rate, we have decided to take 120 subjects for each study and control group.

Clinical oral examination was carried out at the dental clinic with the child seated on a proper dental chair under good lighting using a mouth mirror. They were checked for type of cleft followed by speech assessment.

Speech assessment was conducted using selected sounds at one word level[8]. The phonological assessment was designed specifically for Kelantan-speaking children since to date there is still no standardized screening instrument to assess the phonological development in Malay language. It is a set of 28 picture cards that was designed to elicit 35 spontaneous one word response containing 'di' and 'trisyllabic words' which represent all possible initial and final consonants of Kelantan dialect. All 35 words depict objects and attributes are familiar to the children. All the words used for the assessment were among the earliest vocabulary acquired by the children in Kelantan. Data collection was done clinically by the author (Normastura AR) who was a native speaker of this dialect and thus familiar with their variants. The speech samples were also recorded for each subject using a portable cassette recorder in the cassette tape "Maxell" since it was claimed to be resistant to the background noise, clear and the sensitivity has been improved by 0.5dB in medium and high frequency ranges. The speech samples were taped for the purpose of confirmation and medical record only. There was no difference in terms of results between clinical examination and taped speech. Speech abnormality was assessed for hypernasality, hyponasality, Cul-de-sac resonance and articulation speech. This types are the most common speech problem that occur in CLP and found to have its clinical important.

SPSS version 12.0 (SPSS Inc, Chicago, IL, USA) statistical software was used for data entry and data analysis. Descriptive statistics such as means and standard deviation (SD) or median and interquartile range (IQR) for continuous variables, and frequency and percentages for categorical variables were calculated for each group. To determine the association between the study factor (CLP versus non-cleft) and categorical outcome (speech abnormality), simple logistic regression was used followed by multiple logistic regression analysis. Crude and adjusted odds ratios (adjusted for race, gender and age) were obtained from simple and multiple logistic regressions respectively. Ninety five percent Confidence Interval (CI) of the odds ratios and P value of likelihood-ratio (LR) tests were obtained in order to make inferences to the study population. In multiple logistic regression, the interactions between the study factor and each controlled variable were also checked by LR test. The model was tested for the fitness by using Hosmer-Lemeshow goodness-of-fit test. If the P value approached one, the model was perfect fit. The Receiver Operating Characteristic (ROC) curve for area under the curve and classification table for sensitivity, specificity and correctly classified were also obtained in order to evaluate the model fitness.

RESULTS

A total of 98 CLP (unilateral or bilateral) children and 109 non cleft children (control group) had agreed to participate in the study. Table I shows the socio-demographic characteristics of the 207 subjects. In the CLP group, 78 (79.6%) were unilateral cleft lip and palate (UCLP) and 20 (20.4%) were bilateral cleft lip and palate (BCLP).

The mean age for UCLP, BCLP and non cleft was 5.8 (SD 2.61), 7.3 (SD 3.06) and 7.5 (SD 2.60) years respectively. Males outnumbered females in UCLP and non cleft group, 56.4% and 51.4% respectively. However, they were equal in number in BCLP group. Malays were majority in all groups UCLP (96.2%), BCLP (90.0%) and non cleft (99.1%) which reflected the composition of Malay ethnic in Kelantan.

Table II shows the control of the distribution of speech abnormality in the CLP and non cleft children. Hypernasality occurred in 75% of BCLP and 57.7% of UCLP. Majority of the non cleft children (99.1%) has normal speech except for one (0.9%) has articulation speech. None of the children had hyponasality and Cul-de-sac resonance.

Table III shows control of the prevalence of speech abnormality at 95% Confidence Interval (CI) between CLP and non cleft children. CLP children shown to have higher prevalence of speech abnormality compared to non cleft children.

Table IV shows the summary results of simple logistic regression (SLR) analysis of association between CLP and speech abnormality. In the analysis, the UCLP and BCLP children were combined into one group (CLP group) to compare with the non cleft group. There was a significant association between CLP and speech abnormalities.

Table V shows that CLP was significantly associated with speech abnormality. The two way interactions were not significant. Hosmer-Lemeshow test for fitness of model was not significant (P value = 0.733 at df=8). Therefore, the model was fit. In this model, sensitivity was 93.4% and specificity was 76.0%. The area under the ROC curve was 0.886.

Table I: Socio-demographic characteristics of study samples (207subjects)

Characteristic	UCLP (n= 78)		BCLP (n= 20)		NON- CLEFT (n= 109)	
	Mean (SD)	Freq (%)	Mean (SD)	Freq (%)	Mean(SD)	Freq (%)
Age	5.8 (2.61)	-	7.3 (3.06)	-	7.5 (2.60)	-
Gender						
Male	-	44 (56.4)	-	10 (50.0)	-	56 (51.4)
Female	-	34 (43.6)	-	10 (50.0)	-	53 (48.6)
Race						
Malay	-	75 (96.2)	-	18 (90.0)	-	108 (99.1)
Chinese	-	3 (3.8)	-	1 (5.0)	-	1 (0.9)
Indian	-	-	-	-	-	-
Others	-	-	-	1 (5.0)	-	-

Table II: Distribution of the speech abnormality in CLP (UCLP and BCLP) and Non-Cleft children

Variables speech	UCLP (n= 78)		BCLP (n=20)		NON- CLEFT (n= 109)	
	Freq	%	Freq	%	Freq	%
Normal	33	42.3	5	25.0	108	99.1
Hypernasality	45	57.7	15	75.0	0	0
Hyponasality	0	0	0	0	0	0
Articulation speech	0	0	0	0	1	0.9
Cul-de-sac resonance	0	0	0	0	0	0

Table III: Prevalence of speech abnormality in CLP and Non-Cleft children

Variables	CLP (n= 98)		NON CLEFT (n= 109)	
	%	95% CI	%	95% CI
Speech	61.2	51.41, 71.04	0.9	-0.09, 2.74

Table IV: Univariate analysis for association between CLP and speech abnormality

Variable	Cleft Freq (%)	Non- cleft Freq (%)	Crude OR		LR statistic	P value
			OR	95% CI		
Speech						
Normal	38 (38.8)	108 (99.1)				
Abnormal	60 (61.2)	1 (0.9)	170.53	22.84, 1273.34	25.10	<0.001

Table V: Association between CLP and speech abnormality (outcome variable) adjusted for race, gender and age by using Multiple Logistic Regression*

Variable	Adjusted OR (95% CI)	LR statistic (df)	P value
CLP	**174.45 (23.04, 1320.67)**	**102.86 (1)**	**<0.001**
Non- cleft	**1.00**		
Race†			
Malay	2.77 (0.43, 17.92)	1.26(1)	0.269
Others	1.00		
Gender†			
Female	0.62 (0.27, 1.40)	1.30 (1)	0.256
male	1.00		
Age †	0.98 (0.85)	0.08 (1)	0.782

* the multiple logistic regression model is reasonably fit (Hosmer-Lemeshow goodness-of-fit: Chi square= 5.23 df= 8, P value= 0.733; correctly classified= 81.2%, sensitivity= 93.4 %, specificity= 76.0% ; area under ROC curve= 0.886).
†controlled variables: these variables included in the model to control their confounding effect.
† there is no significant interaction between CLP and each controlled variable.

DISCUSSION

The prevalence of speech abnormalities in CLP children was 61.2% which was very high compared to non cleft children (0.9%). Among the CLP children, speech abnormality was higher in BCLP (75.0%) compared to UCLP (57.7%). The only speech abnormality in BCLP and UCLP was hypernasality. Results showed that the risk of getting speech abnormality was 174.5 times more in CLP children compared to non cleft children. Study by Abdullah on Malaysian CLP patients in Hospital Universiti Kebangsaan Malaysia found that the percentage of BCLP patients with mild to severe hypernasality was 73.7%[10], which was almost similar to our finding (75.0%).

However, for UCLP patients, they got a contrary result where the prevalence of hypernasality was 74.3%.

High prevalence of hypernasality could be explained by the severe anatomic deformities among the BCLP patients. However, since both UCLP and BCLP patients had high prevalence of hypernasality, the timing of palate repair should also be taken into consideration. It is obvious that some surgeons advocate early palatal closure particularly for speech reason[11]. Karling *et al.* in their study found that the mean age for palate repair was 20 months for UCLP and 22 months for the BCLP patients. Therefore, they suggested that besides the more severe anatomic deformities among the BCLP patients, the rather late timing of palate repair may explain the greater need for speech therapy in the BCLP group[12]. In our group of children, we found that the width of cleft is wider and the palatal shelves are more vertical. Thus, the velopharyngeal incompetence (VPI) is much more severe and therefore leading to hypernasality.

None of UCLP and BCLP patients had hyponasality in this study. Karling *et al.* found that 24.0% of CLP patients had hyponasality that could be explained by insufficient nasal patency due to deviated nasal septum or too wide pharyngeal flaps[12]. The prevalence of hypernasality in their study was very low (36.0%) compared to ours but hypernasality was also found among their non cleft patient (5.0%). In our study, the only speech abnormality among the non cleft patient was articulation disorder. Even though in general, individuals with CLP are at high risk for disordered articulation, none of our CLP patients had it. CLP children could achieve their speech maturation as in non cleft children and there has been considerable improvement in treatment methods available. When cleft presents, we often believe as though the physical factors were the sole cause for speech problems in these children. We should appreciate that in the presence of cleft, learning factors and the strategies employed to compensate for the cleft may play an even more significant role in the acquisition of speech.

Therefore besides earlier palatal surgery, treatment plan for speech therapy should be reviewed and focus more on earlier parental information together with stimulation program. Both the parents and the children must be motivated since the successful speech therapy will depend on the consistent and continuous therapy. It was speculated that inconsistent team care and patient and family non-compliance or difficulty in following through with treatment recommendations might contribute to the unsuccessful speech rehabilitation among CLP patients[13].

The prevalence for speech abnormality was noted to be higher in CLP compared to non cleft children. It was higher compared to other studies and this may be related to the timing of palatal surgery or late speech therapy. However, in contrast with other studies, our CLP children only had hypernasality and none of them had hyponasality, articulation speech or cul-de-sac resonance. There was also a significant association between CLP and speech abnormalities. The risk of CLP children for having speech abnormalities is 174.5 times more compared to non cleft children. Abnormal speech is one of the unavoidable complications in the cleft child and it is the most difficult problem to restore in this commonest craniofacial deformity. In this study, the duration of speech therapy carried out for these CLP children cannot be assessed due to incomplete data. Before the year 2000, there was no Speech and Language Pathologist (SLP) in the state of Kelantan. The speech therapy was only conducted either by Physiotherapist or the patients were referred to the SLP in others states. These factors may contribute to the significant different of our results with other studies. Cleft children require support from dedicated speech and language pathologists for training and rehabilitation of speech. Education to parents and teachers who will guide these speech handicap children at home and at school is another important factor to consider. It is also important to realize that the success in speech rehabilitation efforts carried out by these groups is only attainable through a multidisciplinary team care approach that include the pediatric dentists, orthodontics, plastic surgeon, oral surgeon, the general dentists and maxillofacial technologists. This team of personnel will help to restore the dental and oral structure to the correct anatomical alignment so that the common pathological speech in cleft such as hypernasality may be minimized or eliminated.

CONCLUSION

The results indicate that the prevalence of speech abnormality was higher in CLP children compared to non cleft children. The high prevalence of speech abnormality among CLP patients was related to hypernasality. None of them had hyponasality, articulation speech or cul-de-sac resonance. There was a significant association between CLP and speech abnormalities. The risk in CLP child for having speech abnormalities is 174.5 times more compared to non cleft children. Therefore it was found that children with appropriately repaired cleft lip and or palate in Northeast Malaysia failed to have normal speech.

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REFERENCES

1. Hart TC, Marazita MC, Wright JC. The impact of molecular genetic on oral health paradigms. Critical Review Oral Biology Med 2000; 11: 25-56.
2. Kummer AW. Cleft of the lip and palate. Cleft Palate & Craniofacial Anomalies: Effect on speech and Resonance. Singular Thompson Learning, 2001; 52-62.
3. Jensen BL, Kreiborg S, Dahl E, Fogh-Anderson P. Cleft lip and Palate in Dernmark, 1976-1981: Epidemiology, Variability and Early Somatic Development. Cleft Palate Journal 1988; 25(3): 258-26.
4. Ministry of Health Malaysia. National Oral Health Survey Report 1988.
5. Ministry of Health Malaysia). National Oral Health Survey Report 1997.
6. Kelantan Health Department, Kelantan's Health, Technical Report 2001.
7. Chapman KL, Hardin MA. Phonetic and phonologic skills of two-year-olds with cleft palate. Cleft Palate-Craniofac J. 1992; 29(5): 435-43.
8. Wan Zaharah AW. Speech characteristics in Kelantan dialect. Department of Otorhinolaryngology. Universiti Sains Malaysia 2000 (Unpublished).

9. Schonweiler R, Lisson JA, Schonweiler B, Eckardt A, Ptok M, Trankmann J, *et al.* A retrospective study of hearing, speech and language function in children with clefts following palatoplasty and veloplasty procedures at 18-24 months of age. International J of Pediatr Otorhinolaryngol. 1999; 50(3): 205-17.
10. Abdullah S. A study of the results of speech language and hearing assessment of three groups of repaired cleft palate children and adults Annals Academy of Medicine 1988; 17(3): 189-91.
11. Lohmander-Agerskov A, Soderpalm E, Friede H, Lilja J. A longitudinal study of speech in 15 children with cleft lip and palate treated by late repair of the hard palate. Scand J Plast Reconstr Hand Surg. 1995; 29: 21-31.
12. Karling J, Larson O, Leanderson R, Henningsson G. Speech in unilateral and bilateral cleft palate patients from Stockholm. Cleft Palate-Craniofac J 1993; 30(1): 73-77.
13. Paterson-Folconer SJ, Hardin-Jones MA, Karnell MP. Communication Disorder associated with cleft palate. In: Cleft Palate Speech, 3rd edition. Mosby, 2001; 162-93.