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Speech disorders in operated cleft lip and palate children in Northeast Malaysia

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ABSTRACT

The Objective of this study was to investigate the prevalence of speech disorders among operated cleft lip and or palate children in Northeast Malaysia. A comparative cross sectional study was done on 98 operated cleft lip and or palate children attending the combined cleft palate and craniofacial deformity clinic in Hospital Universiti Sains Malaysia (USM). A control group of 109 subjects were selected from healthy non cleft children attending the outpatient dental clinic in Hospital USM. Patients in both groups were inhabitants in the region aged between 3-12 years old. Those with systemic diseases and with hearing problems were excluded from the study. Data collection was done by recording speech samples of each subject from both groups using a portable cassette recorder. The cassette tape was then sent to a Speech and Language Pathologist for interpretation. Our results showed that hypernasality occurred in 75% of bilateral and 57.7% of unilateral cleft lip and palate children. Majority of the non-cleft children (99.1%) in the control group has normal speech. None of the children in both groups had hyponasality and Cul-de-sac resonance. In conclusion, children with operated cleft lip and or palate were shown to have higher prevalence of speech abnormality compared to non-cleft children and there was a significant association between operated cleft lip and palate and speech abnormalities. Therefore it was found that children with appropriately repaired cleft lip and or palate in Northeast Malaysia failed to have normal speech.
Keywords: Cleft lip and palate, speech disorders.

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 cases registered at the Hospital USM Combined Cleft and Craniofacial Deformity Clinic (Combined clinic) from 1997 to 2000 was 760 patients[6].

Children with cleft lip and palate 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].

The current study provided information on speech disorders in cleft lip and palate children and their non cleft peers in our population. Speech disorders is clinically important to ensure appropriate management of communication difficulties and awareness of the possible consequences of the disorder on literacy. Therefore, the aim of this study was to investigate the prevalence of speech disorders among the 3 to 12 years old cleft lip and palate group and normal non cleft group in our children population looking

MATERIALS AND METHODS

This is a comparative cross sectional study involving non-syndromic cleft lip and palate children as the case group while the comparison group comprised of non-cleft children. The source population for cases were all registered cleft lip and palate 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 6 months duration. The inclusion criteria for cases were operated non-syndromic CLP children. Patients in both cases and comparison 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].

All the children with hearing problem were excluded in both groups. The justification for exclusion was based on the study done by Schonweiler *et al.*, who found that speech and language function in CLP patients were predominantly related to the hearing status[9]. Those with systemic diseases were also excluded from both groups. 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 group.

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 by recording speech samples of each subject using a portable cassette recorder by the author who was a native speaker of this dialect and thus familiar with their variants. This method of speech assessment is routinely practised by the Speech and Language Pathologist for medical record and also for the purpose of re-evaluation. The data for speech were recorded 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 cassette tape was then sent to a Speech and Language Pathologist for interpretation. Speech abnormality was assessed for hypernasality, hyponasality, Cul-de-sac resonance and articulation speech. SPSS version 11.0 (SPSS Inc., 1999) statistical software was used for data entry and data analysis. Descriptive

RESULTS

A total of 98 CLP (unilateral or bilateral) children and 109 non-cleft children (comparison 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 and 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 comparison 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 1 (0.9%) has articulation speech. None of the children had hyponasality and Cul-de-sac resonance.

Table III shows comparison 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.

DISCUSSIONS

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 HUKM 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 complication in the cleft child and it is the most difficult problem to restore in this commonest craniofacial deformity. Cleft children require support from dedicated speech and language pathologists for trainning and rehabilitation of speech. Education to parents and teachers who will guide this 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. These team of personnel will help to

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

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[b]			
Malay	2.77 (0.43, 17.92)	1.26(1)	0.269
Others	1.00		
Gender[b]			
Female	0.62 (0.27, 1.40)	1.30 (1)	0.256
male	1.00		
Age [b]	0.98 (0.85)	0.08 (1)	0.782

[a] 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).

[b] controlled variables: these variables included in the model to control their confounding effect.

[b] there is no significant interaction between CLP and each controlled variable.