

# An exploratory study on speech and hearing outcomes in children with cleft lip and palate

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## ABSTRACT

**Introduction:** Little is known about the treatment outcomes of children with cleft lip and/or palate (CLP) receiving surgical care for primary lip and palate closure in Malaysia. **Objectives:** This study examined the speech and hearing status of Malay-speaking children with CLP residing in Kuala Lumpur.

**Methods:** Parents whose children were between the age of 5 and 7 years were recruited via the Cleft Lip and Palate Association of Malaysia (CLAPAM) registry. Parents completed a survey and the children completed a speech and hearing assessment at the Audiology and Speech Sciences Clinic, Universiti Kebangsaan Malaysia.

**Outcomes:** Speech measures include nasality rating, nasalance scores, articulation errors and speech intelligibility rating, while hearing measures include hearing thresholds and tympanometry results for each child.

**Results:** Out of 118 registered members who fulfilled the inclusion criteria, 21 agreed to participate in the study. The overall speech and hearing status of children in this sample were poor. Only four (19%) participants had normal speech intelligibility rating and normal hearing bilaterally. In terms of overall cleft management, only four (19%) participants were seen by a cleft team while seven (33%) had never had their hearing tested prior to this study.

**Conclusion:** Participants in this sample had poor outcomes in speech and hearing and received uncoordinated and fragmented cleft care. This finding calls for further large scale research and collaborative efforts into improving and providing centralised, multidisciplinary care for children born with CLP.

## KEY WORDS:

*Speech; hearing; cleft lip and palate*

## INTRODUCTION

Cleft lip and/or palate (CLP) are among the most common congenital abnormalities, with an overall worldwide prevalence of 9.92 per 10,000 births.<sup>1</sup> The management of

CLP is long term, beginning from birth and continuing into early adulthood.<sup>2</sup> The complex nature of CLP requires a variety of treatment and management interventions beginning with feeding, primary surgery, orthodontic, hearing, speech and language and dental care. Treatment protocols for the management of CLP is diverse and it is essential for health professionals to continually monitor and evaluate treatment outcomes in order to provide the best quality care for children with CLP.<sup>3</sup> Measures of success in the treatment of CLP include one or more of these parameters; attaining favourable speech, hearing, feeding, facial appearance and psychological wellbeing.<sup>4</sup> Nevertheless attaining normal speech and hearing remain one of the key outcome measures of primary palatal surgery. Primary surgical closure of the lip and palate is typically the first step and is crucial in determining further treatment and management. However, large scale studies looking at outcomes of primary cleft surgery are few and difficult to compare because of the heterogeneous nature of CLP population, diversity in the management of cleft as well as methodological construct of studies.<sup>5</sup>

One factor that remains consistent for improved outcomes in cleft care is the provision of centralised and multidisciplinary expert care.<sup>6</sup> Following a national commission to study the outcomes of cleft care in the UK, the Clinical Standards Advisory Group (CSAG) Cleft Lip and Palate Committee reported on overall poor results and fragmented cleft care by low volume operators.<sup>7</sup> Sell *et al.* reported speech outcomes of 238 5-year-olds and 218 12-year-old children with unilateral cleft lip and palate (UCLP) in the UK.<sup>8</sup> The overall findings show poor speech outcomes for both groups despite approximately two-thirds of the sample having undergone speech therapy. 51% of the 5 year-old children had speech intelligibility ratings that would provoke comments, unintelligible to strangers or impossible to understand compared to 19% in the 12-year old group. In addition, 18% of the 5-year-old group and 17% in the 12-year-old group presented with consistent hypernasal speech between mild and severe in degree. The findings provide valuable insights regarding the state of cleft care. Despite the advances in cleft care, there are still a significant number of children with CLP that continue to receive suboptimal benefits from the treatment received.

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In Malaysia, there is no national registry that documents the incidence of CLP. The results of the National Oral Health Surveys (NOHS) indicate approximately 1 in 941 live birth are born with CLP each year.<sup>9</sup> Management typically follows the protocol by the designated hospital of where the child is born. In Malaysia, for decades, surgical (dental, general and/or plastic) management were the main services offered to children with CLP. The role of the speech-language pathologist (SLP) had been minimal if any and only recently available in public hospitals.<sup>10</sup> Cleft teams are available mainly in teaching university hospitals such as Universiti Kebangsaan Malaysia Medical Centre (UKMMC), University Malaya Medical Centre (UMMC) and Hospital Universiti Sains Malaysia (HUSM); and in government hospital, the cleft teams are available at Hospital Kuala Lumpur (HKL), Hospital Sg Buloh and Hospital Angkatan Tentera Tuanku Mizan. The latter two hospitals only started operating in 2015. Although, children with CLP in Malaysia receive surgical care for primary lip and palate closure, little is known about the outcomes and standard practice.

To date, studies reporting speech outcomes in children with CLP in Malaysia is sporadic. Abdullah examined the speech, language and hearing of children and adults with CLP who attended the Speech Therapy Unit at Universiti Kebangsaan Malaysia Medical Centre over a 4-year period.<sup>11</sup> The results showed a high number of patients; 73.7% of the bilateral cleft lip and/or palate (BCLP) patients (n=26) and 74.3% in the unilateral cleft lip and/or palate (UCLP) group (n=48) were hypernasal (mild to severe level). The findings further reported poorer speech outcomes (hypernasality, intelligibility and articulation errors) in the isolated cleft palate group than the UCLP and BCLP groups and called for closer attention to less overt cleft type in rehabilitation. In a separate study, Nurmastura *et al.* reported speech outcomes of children with CLP from the east coast of Malaysia.<sup>12</sup> Ninety eight children from Kelantan between the age of 3 and 12 years old were categorised into cleft (UCLP and BCLP) and non-cleft groups. The mean age was 5.8 years (SD 2.61) for the UCLP and 7.3 years (SD 3.06) for the BCLP respectively. In the study, 61.2% of the 120 children had speech disorders and 75% of children with bilateral cleft lip and palate and 57.7% of children with UCLP were perceived to be hypernasal. No articulation errors were reported for both UCLP and BCLP groups. The authors attributed the astoundingly poor outcomes mainly to the age of surgical repair and the delay in receiving speech therapy and/or shortage of speech-language pathologists but it could possibly be the result of using unstandardised tool, procedures and analysis (i.e. type and experience of rater, rating scale, speech parameters) for measuring the outcomes.

Drawing from the main gaps in the previous study, this study aims to explore the speech and hearing outcomes of children born with CLP living in and around the capital city, Kuala Lumpur. Specifically, the objectives of this study is to profile speech (nasality, nasalance scores, articulation, speech intelligibility) and hearing (hearing threshold) outcomes in children with CLP after primary palatal surgery in children between 5 and 7 years old.

## MATERIALS AND METHODS

An exploratory study was conducted on a sample of children between the age of 5 and 7 years who were registered with the Cleft Lip and Palate Association of Malaysia (CLAPAM) between the year 2004 and 2007. This registry was used to recruit participants because there was no specific registry for reporting CLP in local hospitals and to date, the CLAPAM registry is the single and largest registry locally to capture data on children born with CLP in Malaysia. The participants were recruited through phone contact. While all respondents from the registry were contacted, those who had secondary surgery, syndromic CLP or significant health issues as reported by the parents were excluded. In total there were 251 registered CLAPAM members for the specified age group. Based on the information obtained from the registry, 53% (n=133) were initially excluded because they did not fulfil the inclusion criteria. The remaining 118 members were contacted. There was a very high number of members 66% (n=67) whose contact number was wrong and/or unreachable. Eight members were interested but were not able to attend the speech and hearing appointment given, six declined participation, seven had moved to a different state, three children had passed away while the remaining six were further excluded because they had undergone secondary surgery. The final number of participants who agreed to participate was 21.

Participants were given an appointment for the hearing and speech-language assessment sessions at the Audiology and Speech Sciences Clinic Universiti Kebangsaan Malaysia (UKM) where data were collected. The hearing assessment was carried out first, followed by the speech-language assessment. Each participant attended one speech and hearing assessment session respectively. For the hearing assessment, an audiologist conducted tympanometry and pure tone audiometry testing. The participants then continued with speech-language assessment conducted by a speech-language pathologist. The speech assessment consisted of perceptual assessment of nasality, an articulation assessment, an oral motor examination and nasometry evaluation using the Nasometer II Kay Elemetric 6450 (KayPentax). The data obtained were analysed using the Statistical Package for the Social Sciences (SPSS) 18th edition. Descriptive analysis was used to analyse the results. This research was approved by the Research Ethics Committee of Universiti Kebangsaan Malaysia (UKM 1.5.3.5/244/NN-130-2011) and informed consent was obtained from all participants.

## RESULTS

Speech and hearing results from 21 participants were reported in this study. The mean age of the overall participants was 5:08 years. There were more male (n=15; 71%) participants. Participants were classified into three cleft types; the majority of the participants (n=12; 57%) had unilateral cleft lip and palate (UCLP), five (24%) had cleft palate only (CPO) and four (19%) had bilateral cleft lip and palate (BCLP). All participants in this study were Malays.

Table I: Summary of speech measures in children with cleft lip and palate (n=21)

Speech measures	N (%)
<b>Nasalance score (oral-nasal passage)</b>	
Below cut-off $\leq 30$ (normal limits)	6 (28)
Above cut-off $> 30$	14 (67)
Not tested (did not cooperate)	1 (5)
<b>Nasalance score (oral passage)</b>	
Below cut-off $\leq 22$ (normal limits)	6 (28)
Above cut-off $> 22$	14 (67)
Not tested (did not cooperate)	1 (5)
<b>Perceptual rating of nasality (conversation sample)</b>	
Normal	10 (48)
Mild hypernasality	5 (24)
Moderate hypernasality	4 (19)
Severe hypernasality	2 (9)
<b>Speech intelligibility rating</b>	
Normal	4 (19)
Different from other children but not enough to provoke comment	10 (48)
Different enough to provoke comment but possible to understand most speech	1 (5)
Only just intelligible to strangers	3 (14)
Impossible to understand	3 (14)

The mean age for lip repair was 6 months (age range= 2-18 months) and 24 months for palate repair (age range= 6-30 months). The majority of participants (n=15; 71%) had undergone primary surgery for lip and palate closure at government hospitals while the remaining received surgery at private hospitals. Three (14%) of the participants had a family history of CLP. In terms of education, two (10%) participant were enrolled in special education school while the remaining participants attended normal kindergarten or mainstream school. Seven (33%) participants had never been to an audiologist while five (24%) had never seen a speech-language pathologist. Of the 21 participants, only four was seen by a cleft team. Sixteen (76%) had undergone some speech therapy but only seven (33%) were still receiving services.

The results for speech status are summarised in Table I. Six (29%) participants were judged as "impossible to understand" or "only just intelligible to strangers". Fifteen (71%) had at least one cleft type articulation error in their speech. The most frequent cleft type articulation errors noted among the participants were the posterior oral cleft type characteristics (i.e. backing to velar/uvular) and non-oral cleft type characteristics (i.e. glottal articulation and active nasal fricatives). Five (24%) participants presented with passive cleft type characteristics mainly weak or nasalised consonants, nasal realisation of plosives and/or suspected passive nasal fricatives.

Hearing assessment revealed that only four (19%) had normal hearing bilaterally. Six (29%) had unilateral conductive hearing loss (mild to moderate), seven (33%) had conductive hearing loss in at least one ear (mild to severe) and three (14%) had conductive hearing loss bilaterally (mild to moderate). One participant failed to complete the hearing assessment. Of the 42 ears tested for tympanometry, 17 (40%) was Type B, followed by eight (19%) Type A and C respectively, and five (12%) was Type As. Tympanometry results for four (10%) ears was not obtained.

## DISCUSSION

The findings of this study provided preliminary speech and hearing outcomes and information regarding the treatment received by a small sample of children with CLP in Kuala Lumpur. Overall, children with CLP in this sample had poor speech and hearing outcomes post primary palatal surgery. Only four of the 21 children with CLP had normal speech intelligibility rating and normal hearing bilaterally. Furthermore, in terms of cleft care, four participants were seen by a cleft team while the remaining children received uncoordinated and incomprehensive cleft treatment. Considering the fact that the children in this sample were beginning formal schooling (kindergarten and primary one), the speech and hearing results and cleft care received were far from optimal.

In the present study the mean age for palate repair was 24 months which was much later than the recommended optimal age for primary surgery for lip and palate repair; between the age of six and 18 months,<sup>2,13</sup> while Hardin-Jones and Jones advocated for primary palatal surgery no later than 13 months of age.<sup>14</sup> Compared to the recommend treatment regime, participants in this study were late in receiving primary palatal surgery. In terms of overall CLP management, only four participants were seen by a cleft team while others may have not received comprehensive care. Seven children had never had their hearing tested prior to this study and five children had never had SLP consult before. Essentially, a multidisciplinary approach with expert care beginning from birth is required for successful management of cleft.

Additionally, for those who had been referred for speech and language services; there was a tendency for parents to default such services. Factors to account for this may be the lack of awareness among parents about such services, a possible lack of parental commitment, or the long durations between appointments in public hospitals due to SLP shortages which hamper successful therapy. Thus, in this study, more than half of participants who have had previous speech therapy defaulted and attended less than five sessions and only seven

participants were still being followed up by SLPs for treatment. This further reflects the lack of understanding of the importance of continuous and longitudinal care in CL/P amongst parents and caregivers.

Hearing outcomes in this study were poor with only four or 19% of children had normal hearing bilaterally leaving the remaining 81% of children with some form of hearing problems. Almost 60% had abnormal tympanometry findings suggesting the presence of otitis media. Compared with other similar published studies, the results of the current study had the poorest findings. In Turkey, Tuncbilek *et al.* found that 63% of their participants had normal hearing levels ( $\leq 15$  dB)<sup>15</sup> while in the USA, 75% of children with CL/P had normal hearing levels at both 12-month and 2-year old follow-ups.<sup>16</sup> The high frequency of hearing issues in the present study is of concern. This finding may again point to the late palatal repair received in this group of children coupled with the lack of access and timely audiological management.

With regards to speech intelligibility, findings from the current study were similar to the findings reported by Sell *et al.* where 19% of the participants had normal speech intelligibility.<sup>8</sup> However, the present study had a higher percentage of participants (28%) who were judged to be "impossible to understand" or "only just intelligible to strangers". In the present study, 71% children with CL/P had at least one cleft type articulation error compared to 34% of children with CL/P who had articulation errors in the UK study. The types articulation errors noted in children with CL/P in United Kingdom were dentalisation/ interdentalisation, velar palatalisation and backing to velar. However in the current study, the most frequent cleft type articulation errors noted among the participants were the posterior oral cleft type characteristics (i.e. backing to velar/uvular) and non-oral cleft type characteristics (i.e. glottal articulation and active nasal fricatives). Five participants presented with passive cleft type characteristics mainly weak or nasalised consonants, nasal realisation of plosives and/or suspected passive nasal fricatives. Findings for perceptual evaluation of nasality and nasometry further added to overall poor speech outcomes. In the current study, 43% of participants demonstrated hypernasal speech from mild to severe levels compared to Sell *et al.* where only 18% of the 5-year old children with CL/P had hypernasal speech.<sup>8</sup> Nasometry results further showed that 67% of children with CLP had nasalance scores that were higher than the cut-off scores for the oral passage ( $\leq 20$ ) and oral-nasal passages ( $\leq 30$ ) further confirming the occurrence of hypernasality in speech.<sup>17</sup>

The outcomes reflected in this sample of children with CLP show that the existing management of cleft care is not optimal and together with late palatal surgery may be a factor to explain the poor hearing and speech results. It is evident that a majority of the children in this sample will benefit from receiving timely audiological management, consistent speech therapy as well as be evaluated further for secondary surgery. Left untreated, these children are predisposed to teasing and bullying which leads to poor psychological well-being<sup>18</sup> and difficulty in literacy skills that

is important for academic success.<sup>19</sup> Long term effects of hearing problems in children with CLP include elevated hearing thresholds in high frequencies that may lead to difficulty in communication in noisy environments as well as negatively impact on academic performance.<sup>20</sup>

It is important to point out that the results of this study were confounded by several methodological constraints primarily the sampling method and the small sample size. It was difficult to recruit participants with CLP from local hospitals in the Kuala Lumpur area primarily because there is no comprehensive cleft registry for local hospitals. The number of cases per hospital may be too few for conducting large scale research in CLP or measuring outcomes. Furthermore, coordinated care for CLP patients in this sample is heterogeneous and dependent upon the cleft treatment adopted by the hospital or the primary health care provider. In addition, the poor updates of members' data in the registry as reflected by the high number of wrong and/or unreachable contact details further contributed to the small sample size. This situation not only confines research in the area of CLP but also hinders the process of measuring long term outcomes in cleft care management in Malaysia.

## CONCLUSION

This is the first study that attempted to profile the speech and hearing status of children with CLP following primary palatal surgery in Kuala Lumpur. The finding of this study reflects the shortcomings in the management of CLP in this sample particularly with difficulty to access multidisciplinary expert care. Although the results were confounded by methodological constraints (i.e. sample size) the findings reflect the pressing needs for better cleft care; essentially one that is provided by a multidisciplinary team in a coordinated manner. Further large scale study incorporating all aspects of cleft care is needed to measure treatment outcomes successfully.

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