

**Original article**

**A perceptual evaluation of speech disorders in children with repaired unilateral cleft lip and palate in Hospital Universiti Sains Malaysia**

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**Abstract**

**Aims and objective:** The purpose of this cross sectional study was to determine the types and severity of speech disorders in children with repaired unilateral cleft lip and palate (UCLP) in Hospital Universiti Sains Malaysia (Hospital USM) and describe the inter- and intra-judge reliability of perceptual evaluation of speech disorders using GOS.SP.PASS'98 and five-point rating scale. Four children with repaired UCLP with ages ranging from 8 years old to 12 years old were included in this study. Prior to data collection, participants were contacted through telephone call from the list of patients with UCLP obtained Combined Cleft and Craniofacial Deformity Clinic (Combined clinic) in Hospital USM, Kelantan from year 2013-2015 as well as from clinical records from the database of the Record Unit of Hospital USM from year 2003-2015. Following strict inclusion criteria participant has been selected. After that, history taking was first conducted with the participant's parents, followed by collection of participant's speech sample and finally concluded with oral motor examination. **Results:** Fifty percent of the speech samples obtained from data collection were then duplicated for use in inter-rater and intra-rater reliability investigations. Exact agreement and kappa values were used for reliability measures. Seventy-five percent (3/4) participants exhibited speech disorders and 25% (1/4) participant had no evidence of speech disorder. The type of speech errors that occurred the most in the three participants were glottal articulations (39%), while the least speech errors included lateralizations/lateral articulations (2%), backing to velar (1%), pharyngeal articulations (1%), active nasal fricatives (1%) as well as nasal realizations of fricatives (1%). The severity of speech disorder ranged from some occurrences to occurring always or almost always in the participants. Inter-judge reliability showed poor to slight agreement while intra-judge reliability revealed almost perfect agreement. **Conclusion:** The results obtained from this study were more or less similar to other studies conducted on the speech outcome of individuals with repaired UCLP. It is important that referrals be made to speech-language pathologists (SLPs) to evaluate the individuals' speech outcomes even though surgical intervention has been provided to them to ensure a comprehensive management for the individual.

**Keywords:** Unilateral cleft lip and palate; Speech disorder

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**Introduction**

Communication is made up of several components including hearing, receptive language, expressive language, speech, resonance, voice, and the social use of language most commonly referred to as "pragmatic skills"<sup>1</sup>. Speech is the 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<sup>2</sup>. The four substructures of speech consists of respiration, which is our breathing, phonation when sound is made by the vocal folds, articulation

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which is the production of sounds using the lips, teeth, tongue and jaw movements and resonance or the quality of voice regulated by the integrity and the movement of the soft palate and surrounding structures. Among one of the populations that SLPs work with children with craniofacial anomalies, such as cleft lip and palate (CLP).

CLP is one of the most common congenital craniofacial anomalies that occurs at birth<sup>3, 4</sup>. It affects about 1.5 per 1000 live births worldwide<sup>4</sup>. Children with CLP do not represent a homogenous population, and the signs and symptoms associated with clefting depend on a variety of factors<sup>5,6,7,8</sup>. Common issues that children with a history of cleft palate (CP) can encounter include problems related to feeding, dental and occlusal deviations, hearing and middle ear function, psychosocial development and in speech sound acquisition and language development<sup>9,10</sup>. Besides that, all of the components of communication can be affected by the presence of a cleft, particularly speech<sup>11</sup>. In individuals with CLP, errors in speech production are noticed due to the abnormalities in oronasal structure/function, orofacial structure and growth, learned neuromotor patterns during early infancy, and/or disturbed psychosocial development<sup>11</sup>. Due to this, children with CLP often exhibit 'cleft palate speech' that includes atypical consonant productions, abnormal nasal resonance, abnormal nasal airflow, altered laryngeal voice quality, and nasal or facial grimaces<sup>12</sup>. Since it is common for children born with CP or with CLP to have speech problems at some time in their lives, the aim of this study to determine the types and severity of speech disorders exhibited in children with repaired UCLP in Hospital Universiti Sains Malaysia and to describe the inter-rater and intra-rater reliability of perceptual evaluation of speech disorders using GOS.SP.ASS'98 and five point rating scale of the assessed speech parameters among SLPs.

### **Methodology and Methods**

This study was approved by the Ethical Committee of the Hospital Universiti Sains Malaysia (HUSM) [USM/JEPeM/15100355].

This study was designed including children with repaired UCLP who attended Combined Cleft and Craniofacial Deformity Clinic (Combined Clinic) as patients in Hospital USM, Kelantan from year 2013-2015 and from clinical records from the database of the Record Unit of Hospital USM from year 2003-2015. Only the children who fitted the age range of 8-12 years old were taken into consideration. Patients' parents were then contacted through

telephone to briefly explain about the research and their willingness to participate. Time and date was then set to meet the participants.

The inclusion criterias of our study were -

1. Non-syndromic UCLP patient.
2. Individual aged 8-12 years.
3. Lip surgery and palatoplasty had been performed.
4. Speak in Bahasa Melayu

The exclusion criterias of our study were

1. Have language or developmental problems
2. Have history of hearing problems

The participants were included in this study by convenience sampling, which is a form of nonprobability sampling. All the participants' caregiver from the list were attempted to be contacted. Those who were able to be contacted, fitted the inclusion criteria and were willing to participate in this study, were then recruited as participants for the study. Participants were selected based on caregivers' ease in volunteering and their availability. This ease was mainly due to the fact that the caregivers were provided liberty in determining the time and date to come in for data collection according to their convenience.

Sample size was not calculated for this study as the sample size was taken based on those who were able to participate in the research study. The total number of patients obtained from both sources was 21 patients. Therefore, by only recruiting those who were able to participate in the research study, the sample size consisted of four participants. **Table 1** describe the status of each patient from the two lists.

### **Instruments**

There are some instruments used during the testing which is the perceptual speech assessment form, GOS.SP.ASS' 98. The GOS.SP.ASS' 98 form has a component for perceptual assessment of speech disorders associated with CP and/or VPD under the heading 'Cleft Type Characteristics'. Under Cleft Type Characteristics (CTCs), it can be identified through the transcriptions of the participants' speech samples. The number of the relevant characteristic/s as listed in the form is circled to show its presence in the speech sample. CTCs can be categorized into dentalization, lateralization/lateral articulation, palatalization/palatal, double articulation, backing to velar, backing to uvular, pharyngeal articulation, glottal articulation, active nasal fricatives, weak/nasalized consonants, nasal realizations of fricatives, nasal realizations of plosives, absent pressure consonants and finally gliding of fricatives/affricates. Space is provided in the form of dotted lines adjacent

to the process for the transcription of speech, where specific examples, whole word transcription or atypical consonant cluster production may be entered to provide extra information and provide a clearer view.

Another instrument used is a five-point rating scale with description of the scale values for the assessed speech parameters (Appendix C, page 77) used to determine the severity of the CTCs present. This scale ranges from 0 (no occurrence), 1 (single occurrence), 2 (some occurrence), 3 (frequently occurring) and 5 (occurring always or almost always). The scales are similar to the five degrees used in Clinical Standards Advisory Group study<sup>13</sup> and to the four-point scale used by Van Lierdeetal<sup>14</sup>. These scales are used nationwide at Swedish cleft palate centers<sup>15</sup>.

Besides that, validated speech stimuli in BahasaMelayu (Appendix D, page 78), in which permission was already obtained from its' developer for usage in this study to obtain the speech sample of the participants (Appendix E, page 79) was also used, along with a history taking (Appendix F, page 80) and oral motor examination form (Appendix G, page 81) and audio recorder (SONY Stereo IC Recorder, ICD-UX543F, Appendix H, page 82) to record participant's speech sample.

### **Method**

On the day of data collection, history taking was first conducted to obtain information regarding the antenatal history, birth history, postnatal history, medical history, hearing status, language development as well as involvement of other professionals of the participants. After that participants' speech samples were then collected. Conversational speech was collected through building rapport with the participants. Participants were then required to read a series of speech stimuli in BahasaMelayu and their speech samples were recorded using an audio recorder and used for data analysis for the study. Once the participant's speech sample was obtained, oral motor examination was performed to examine the participant's oral structures and function, so as to identify or rule out the structural or functional factors that relate to a communicative disorder. A group of five listeners participated in this study. Three of the listeners were certified SLPs while the other two were the researchers. The three certified SLPs are with more than five years of experience in the field of CP speech. The researchers were trained through clinical exposure to cleft type speech disorders from January 2016 till April 2016. A perceptual analysis was performed from the

speech sample recordings. This was done by the researcher listening to all of the audio recordings of speech samples and transcribing each of the speech samples accordingly using International Phonetic Alphabet (IPA) and extra IPA (extIPA) for disordered speech (Appendix I, page 83). After transcriptions for each of the speech samples were done (Appendix J, page 84) the researcher listened to the speech samples again and classified the types of CTCs based on the GOS.SP.ASS '98 form and determined overall severity of the speech disorder according to the five-point rating scale for the assessed speech parameters while referring to the transcriptions at the same time for each of the participants' speech samples.

Once perceptual analysis was done, the total number of CTCs that occurred in each of the participants' speech sample was calculated. For each of the participant's speech sample, the total number of CTCs was obtained by calculating the number of times a CTC occurred throughout the speech sample. Percentage of each CTC for each participant was then calculated by taking the number of a particular CTC divided by the total number of CTCs present within the speech sample and multiplied by 100.

Overall number of CTCs from all participants was then calculated by adding all of the CTCs from each participant. Each CTC was added accordingly into its own classification. Total percentage of each CTC was calculated by taking the total number of that particular CTC and divided by the total number of CTCs that occurred throughout all of the speech samples and multiplied by 100.

### **Statistical analysis**

The data was analyzed statistically using IBM Statistical Package for Social Sciences (SPSS) Version 22.0. The analysis of the data regarding the types and severity of speech disorders were interpreted in the form of tables. The intra- and inter-examiner agreements were analyzed with the kappa statistics. According to Landis & Koch<sup>16</sup>, the kappa values of the intra- and inter-examiner agreements were interpreted.

### **Results**

#### **Demographic Data**

Table 2 shows demographic data of each participant in this research study including their age, gender, type of cleft, age of lip repair and palatoplasty, age at which started speech therapy and duration of speech therapy.

#### **Perceptual Evaluation**

Perceptual evaluation was carried out by first

determining the types of CTCs exhibited by the participants. Table 3 shows the CTCs as exhibited by the participants.

Besides determining the different CTCs exhibited by each participant, a rating of the severity of their overall speech disorder was given using the five-point rating scale of the assessed speech parameters. Therefore, by referring to the list of CTCs from GOS.SP.ASS '98 and the five-point rating scale, the different CTCs and overall severity of speech disorders of each participants' are summarized in the Table 4.

#### **Intra- and Inter-examiner agreements**

The exact agreement for all ratings was 100% and the measure of agreement for Kappa for all ratings was 1.00. This Kappa value suggested almost perfect agreement<sup>9</sup>. **Table 5** shows the intra-rater reliability for all speech samples.

Inter-rater reliability was determined between three raters who were SLPs in Hospital USM for 50% of the speech samples from all four participants using Kappa. The Kappa was -0.33 to 0.00 calculated from the exact agreement scores which indicate poor to slight agreement<sup>9</sup>. Table 5 shows the inter-rater reliability between all three raters.

#### **Discussion**

Previous studies revealed speech disorder is one of the common problems associated with CLP patients. The outcome of our study is consistent with most previous studies investigating speech outcomes in children with CLP. According to Normastura et al<sup>2</sup>, there is 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, hence it is one of the unavoidable complication in the cleft child. Hortis-Dzierzbicka et al<sup>17</sup> investigated the speech outcomes of complete UCLP after one-stage lip and palate repair in the first year of life and found articulation development and incidence of compensatory articulations in the sample were mostly satisfactory. In a study by Albustanji et al<sup>18</sup> found that out of 80 participants with CLP, 21 participants had normal articulation and resonance, 59 of participants (74%) showed speech abnormalities. Timmons et al<sup>19</sup> found final speech outcomes were similar for CP and CLP patients, whereby CTCs were noted in 11 (41%) CP and nine (53%) CLP patients.

There could be a number of reasons why speech disorders still occur even after the appropriate surgical management or speech therapy have been conducted with the UCLP children. A possible factor

that may affect development of normal articulation pattern could be attributed to age of the repair or surgery. Previous studies suggest that children who receive early palatal repair demonstrate better overall speech than their peers who receive surgery at a later age<sup>20</sup> and those children who receive late palatal surgery are at greater risk for developing atypical articulation patterns<sup>21-25</sup>.

Clinical outcomes of primary CP repair, such as articulatory deficits are related to several factors, including cleft type, the extent of innate clefting, surgical repair techniques, expertise of the operating surgeon, preoperative orthopedics, and timing of primary palatal repair<sup>26-33</sup>. Thus, a possible explanation for the problem above could have been one of the factors or a combination of factors that resulted in the presence of speech disorders even after appropriate surgical correction has been provided to the participants of this study.

Glottal articulations were discovered to be the most number of CTCs produced by the participants of this study. Authors have hypothesized that glottal and pharyngeal articulations could develop because a child compensates by valving the airstream at a point in his/her vocal tract inferior to the velopharyngeal port so as to normalize pressure<sup>34, 35</sup>. According to D'Antonio & Scherer<sup>1</sup>, the most common and distinctive of the compensatory articulation errors that frequently occur in the speech of individuals with CP is the glottal stop, and these misarticulations are often difficult to eradicate even after therapy, which supports the findings of this study where the most number of CTCs observed was glottal articulations. Compensatory articulation errors such as glottal or pharyngeal articulations occur when a child with CP attempts to compensate for velopharyngeal inadequacy, thus they are learned behaviours that typically do not resolve even after secondary management to repair the CP and when a potentially adequate velopharyngeal mechanism has been achieved<sup>36</sup>. Most of the time speech therapy will be needed to correct these compensatory articulations.

On the other hand, weak consonants, nasal realizations of plosives and nasal realizations of fricatives are passive consequence of velopharyngeal dysfunction, or due to presence of fistulae. These reduces the child's ability in achieving and/or sustaining intraoral pressure<sup>37</sup> causing certain consonants to sound weak or nasalized. However during oral-motor examination of all four participants in this study, no evidence of fistula was found in any of the participants. Therefore,

it could be assumed that these passive CTCs are due to VPD such as velopharyngeal insufficiency (lack of velum tissue) or velopharyngeal incompetence (lack of proper movement of the lateral and posterior pharyngeal walls as well as the velum). Both of these will cause impaired velopharyngeal closure.

In terms of severity of the overall speech disorders, results of this study indicated that three of the participants (75%) who exhibited CTCs had a severity rating ranging from some occurrences to occurring always or almost always. Out of those three participants who exhibited CTCs, two participants showed a severity of some occurrences while one participant had a severity of occurring always or almost always. This result is almost similar to a study by Nyberg et al<sup>38</sup> where in general, most of the investigated children had minor speech problems. Similar speech outcomes were also reported by Hortis-Dzierzbicka et al<sup>17</sup>

For this study, a factor that could be attributed to the differences in severity rating of speech disorder is because of the different duration of speech therapy for each of the participants. Generally, better speech outcome is associated with a more frequent and longer duration of undergoing speech therapy. However, in this study one of the participants who had undergone speech therapy for four years still exhibited occurring always or almost always CTCs while another participant who had been undergoing speech therapy for one month only demonstrated some occurrences of CTCs. However, it should be noted that although there is a difference between the overall duration of undergoing speech therapy and the severity rating of the participant, the frequency in which participants went for speech therapy was not known. Therefore it is possible for a participant who frequently went for speech therapy in a shorter duration to have only some occurrences of CTCs when compared to a participant who did not frequently attend speech therapy in a longer duration to exhibit occurring always or almost always CTCs. Therefore, motivation and commitment from both the parents and children is critical in ensuring successful speech therapy<sup>2</sup>.

In summary, perceptual speech assessment is central to the evaluation of speech outcomes associated with CP and VPD<sup>39</sup>. The primary purpose of the perceptual speech evaluation is to determine the characteristics and cause of the speech problem so that appropriate treatment recommendations can be made. It is critical to make the right diagnosis when there has been a history of CP or if there are characteristics of VPD because the diagnosis will determine whether appropriate treatment includes surgical intervention, speech therapy, or both<sup>40</sup>. Information such as the types and severity of speech disorders gathered through perceptual assessment will be useful in acting as a baseline for SLPs to further manage a CLP case, whether in determining which therapy techniques are suitable to be applied in rehabilitating the specific CTCs as exhibited by the child or in terms of frequency of speech therapy required.

### **Conclusion**

Based on the results, following conclusion can be drawn

1. Seventy five percent (75%) (3/4) participants exhibited speech disorders, while 25% (1/4) participant had no speech disorder
2. In terms of types of speech errors exhibited by children with repaired UCLP the most errors consisted of glottal articulations, while the least were lateralization/lateral articulations, double articulation, backing to velar, pharyngeal articulation, active nasal fricatives and finally nasal realizations of fricatives. This indicated that glottal articulations are the most common CTC in the CP population.
3. Severity of the speech disorders exhibited by children with repaired UCLP, it ranged from some occurrences to occurring always or almost always, which may be due to the different duration and frequency of speech therapy attended by each of the participants.

### **Acknowledgements**

None declared

### **Conflict of interest**

The authors have declared that no COI exists.

**Table 1.** Number of patients from Combined Cleft and Craniofacial Deformity Clinic HUSM (2013-2015) and from Record Unit HUSM (2003-2015) according to status

Status	Number of patients from Combined Cleft and Craniofacial Deformity Clinic HUSM (2013-2015)	Number of patients from Record Unit HUSM (2003-2015)
Voicemail	3	3
Could not be contacted	2	1
Refused to participate	1	2
Could be reached but did not fit the inclusion criteria	2	0
Agreed to participate	3	1
Live in other states	0	3
<b>Total</b>	11	10

**Table 2.** Demographic data of participants

Participant	Age (years)	Gender	Side of UCLP	Age (months) of Lip Repair	Age (months) of Palatoplasty	Age (years) at which started Speech Therapy	Duration of Speech Therapy
1	9	Female	Left	3	7	3	6 years
2	12	Male	Left	3	3	6	4 years
3	9	Female	Right	3	11	4	1 month
4	12	Male	Midline	6	12	5	1 year

**Table 3.** Cleft type characteristics exhibited by participants

Cleft type characteristics (CTCs)	Number of participants
Lateralization/Lateral articulation	1
Double articulation	1
Backing to velar	2
Pharyngeal articulation	1
Glottal articulation	3
Active nasal fricatives	1
Weak/nasalized consonants	3
Nasal realizations of fricatives	1
Nasal realizations of plosives	2
Absent pressure consonants	2
Palatalization/Palatal	0
Backing to uvular	0
Gliding of fricatives/affricates	0

**Table 4.** Cleft type characteristics and overall severity of speech disorder of each participant

Participant	Cleft type characteristics	Overall severity of speech disorder
1	Glottal articulation Weak consonants Absent pressure consonants	Grade 2 (some occurrences)
2	Lateralization/Lateral articulation Double articulation Backing to velar Pharyngeal articulation Glottal articulation Active nasal fricatives Weak/nasalized consonants Nasal realizations of fricatives Nasal realizations of plosives Absent pressure consonant	Grade 4 (Occurring always or almost always)
3	Glottal articulation Weak consonants	Grade 2 (Some occurrences)
4	None	Grade 0 (No occurrence)

**Table 5.** Kappa values of ratings for 4 speech samples

Intra-examiner agreements		
Speech Samples	Exact Agreement (%)	Kappa value
1	100	1.00
2	100	1.00
3	100	1.00
4	100	1.00
Inter-examiner agreements		
SLP	Exact Agreement (%)	Weighted Kappa
1 and 2	50	-0.33
2 and 3	50	0.00
1 and 3	50	0.00

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