

Original article:

A perceptual evaluation of resonance disorders in children with repaired unilateral cleft lip and palate in Hospital UniversitiSains Malaysia

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Abstract

Background: In most literature regarding speech outcomes in children with repaired cleft lip and palate (CLP), exhibited resonance disorders despite having surgical repair. However, the types and severity of the resonance disorders vary from one individual to another. Thus, perceptual evaluation is important to determine the speech outcomes in individuals with repaired cleft lip and palate. **Aim:** The aim of this cross sectional study was to determine the types and severity of resonance disorders of children with repaired unilateral cleft lip and palate (UCLP) and describe the inter- and intra-rater reliability of perceptual evaluation of resonance disorders using GOS. SP.PASS'98. Four children with repaired UCLP in Hospital USM with ages ranging from 8 years old to 12 years old was included in this study. During data collection, history taking was first conducted, then participants' speech sample was collected and lastly oral motor examination was conducted. **Results:** 50% of the speech sample was then duplicated for inter- and intra-examiner reliability investigations. Exact agreement and kappa values were used for reliability measures. Seventy five percent (75%) (3/4) participants exhibited hypernasality as the type of resonance disorder and 25% (1/4) participant had no evidence of hypernasality or any other types of resonance disorder. No other types of resonance disorders such as hyponasality, mixed resonance or cul-de-saq was noted in the participants. The severity of hypernasality ranged from mild to severe in the participants. Inter-rater reliability showed fair to almost perfect agreement and intra-rater reliability revealed almost perfect agreement. **Conclusion:** The results obtained from this study was more or less similar to our other studies conducted on the speech outcome of individuals with repaired UCLP. Although surgical intervention has been provided to these individuals, it is important that referrals be made to SLPs to evaluate their speech outcomes. This is to determine a proper management for the individual.

Keywords: Unilateral cleft lip and palate, resonance disorder.

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Introduction

The term "cleft" means a split or a divide. Children can be born with a variety of cleft types and with variable severity. Cleft lip and palate (CLP) are considered one of the most common birth defects that possess significant medical, psychological, social, and financial implications on the affected

individuals and families. CLP represent a major public health problem due to the possible associated life-long morbidity, complex etiology, and the extensive multidisciplinary commitment required for intervention. It affects about 1.5 per 1000 live births (250,000 new cases per year) worldwide, with tremendous variations across geographic areas and

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ethnic groups¹). The etiology of CLP has been thought to be multifactorial with genetic and environmental factors^{2,3}). Multifarious functional problems like feeding, speech, hearing, dental functioning and also psychological dilemma can happen to the patient⁴). CLP pt requires coordinated care from multiple specialties to optimize treatment outcome. It's a complex lengthy treatment plan and several surgeries are essential for the good treatment outcome^{5, 6, 7}). Therefore, treatment outcome of these surgeries like cheiloplasty and palatoplasty can be evaluated by several methods⁸⁻¹⁶).

Children with CLP frequently demonstrate speech and resonance disorders following primary surgical repair of the palatal cleft¹⁷). They are at risk for resonance disorders, articulation and also expressive language skills that can affect their communication skills. Despite undergoing palatoplasty surgery, 20% to 30% of repaired cleft palate will demonstrate some degree of velopharyngeal dysfunction, resulting in abnormal speech¹⁷).

When there is an abnormal balance of sound energy in the cavities of the vocal tract (pharyngeal cavity, oral cavity and nasal cavity), this causes resonance disorder. In the speech assessment, the examiner must first determine the type of resonance (normal, hypernasal, hyponasal, cul-de-sac, or mixed) by listening to spontaneous speech¹⁸). Resonance distortion is, for the most part, the direct effect of coupling of the nasal space with the oral-pharyngeal space during vowel and vocalic productions¹⁹).

The strong relationship between palatal clefting and the presence of hypernasality is so well recognized that in the early years of the field, the presence of hypernasality in a speaker was commonly referred to as "cleft palate speech"²⁰). Resonance disorders is known to affect an individual's communication skills which is important for an individual to fulfill their daily and social needs.

However, it is known that many individuals with repaired CLP still exhibit cleft type speech including articulation disorders, resonance disorders as well as nasal emission. However, the severity and types of resonance disorders exhibited by individuals with repaired CLP is still not known especially in Kelantan. So far only one study has been found where the prevalence of speech disorders among operated CLP children in Northeast Malaysia was investigated but the severity of the disorder was not conducted. Other than that, many studies have been conducted to investigate the speech outcomes in CLP patients but less studies focused only on resonance

disorders. The aim of this study to determine the types and severity of resonance disorders exhibited in children with repaired unilateral cleft lip and palate (UCLP) in Hospital Universiti Sains Malaysia and to describe the inter-rater and intra-rater reliability of perceptual evaluation of resonance disorders using GOS.SP.ASS'98.

Materials and Method

This study was approved by the Ethical Committee of the Hospital Universiti Sains Malaysia (HUSM) [USM/JEPeM/15100354].

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 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

Among the instruments used during this research study was the perceptual speech assessment form:

Great Ormond Street Speech Assessment (GOS.SP.ASS' 98). This perceptual speech assessment form was used to determine the types and severity of resonance disorders from participants' speech sample. The section used for administration was on resonance which consisted of 4 types which were hypernasal, hyponasal, mixed resonance and cul-de-saq. Grading system was only present for hypernasal resonance and hyponasal resonance according to the assessment form. **Table 2** shows the grading system (severity rating) of Hypernasal resonance and hyponasal resonance.

A validated speech stimuli in the Malay language was also used. Hasherahet al²¹⁾ conducted a study to establish normative nasalance scores for a set of newly developed stimuli in Malay. Hence, this new standardized stimuli in the Malay language was used to obtain participants' speech sample. Permission was obtained from its developer for usage in this study.

Another instrument used was a case history and oral motor examination form from Klinik Pertuturan Bahasa (KPB), PPSK. The form was used to conduct history taking to obtain more information about the participants and also to perform oral motor examination to check on the structures and functions of participants' speech mechanism. An audio recorder of the model SONY Stereo IC Recorder, ICD-UX543F was also used 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 collected. Conversational speech was first collected through building rapport with the participants. Participants were then required to read a series of speech stimuli in Bahasa Melayu 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. Specifically, participants' range of motion and strength of the tongue, lips and jaw was examined. Any presence of fistula or bifid uvula was also examined.

A group of four listeners participated in this research study. Three of the listeners were certified SLPs and another one was the researcher herself. The

three certified SLPs are with more than five years of experience in the field of cleft palate speech. The researcher was also trained during attachments in the Combined Cleft and Craniofacial Deformity Clinic in Hospital USM from January 2016 till April 2016.

Data analysis

A transcription for each of the participant's speech samples was done first by listening to the audio recordings. The transcription was done by referring to the International Phonetic Alphabets (IPA) and extIPA for disordered speech. After transcribing, a perceptual evaluation was performed from the audio recordings of the speech samples together with the transcriptions. The participants' speech samples were classified into types of resonance disorders and given a rating of severity for their resonance disorders using GOS.SP.ASS' 98 form.

Reliability Investigations

The inter-examiner reliability was done by three certified speech-language pathologists (SLPs). 50% of the audio recordings of the speech samples were emailed to the respective SLPs and GOS.SP.ASS'98 forms were given to each SLP. The SLPs were required to listen to the audio recordings, determine the types of resonance disorders and give ratings on the severity of resonance disorders using the GOS.SP.ASS'98 form for each of the participant. The SLPs were also informed that their ratings will be used as comparison to determine the inter-rater reliability. The SLPs were given one week to complete their ratings.

The intra-examiner reliability was also done with one week duration in between the initial rating and second rating. The researcher listened to all of the audio recordings of the speech samples and together with the transcriptions, determined the types of resonance disorders and gave ratings on the severity of resonance disorders using GOS.SP.ASS'98 form.

Statistical analysis

The data was analysed 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 & Koch²²⁾, the kappa values of the intra- and inter-examiner agreements were interpreted. Kappa is a reliability measure that compares obtained agreements with expected agreements by chance. A kappa value of 0 indicates no agreement and a value of 1 indicates perfect agreement.

Results

Demographic Data

Table 3 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 on Types of Resonance Disorders

Perceptual evaluation was carried out by first determining the types of resonance disorders exhibited by the participants based on GOS.SP.ASS' 98 by listening to the audio recordings of the speech samples. **Table 4** shows the results of descriptive analysis of types of resonance disorders exhibited by the participants where a total of 3 participants exhibited hypernasality. None of the participants exhibited hyponasality, mixed resonance or cul-de-saq. One of the participant had no resonance disorder despite having repaired UCLP.

Figure 1 shows the presence of hypernasality among the participants where 75% (3/4) participants had resonance disorders of hypernasality and 25% (1/4) participant had no hypernasality.

Perceptual Evaluation on Severity of Resonance Disorders

A rating of the severity of the participants' resonance disorders were given using GOS.SP.ASS' 98 by listening to the audio recordings of the speech samples. Transcriptions of the speech samples of the participants were also done to help give ratings of severity of the participants' resonance disorders. Since 3 participants exhibited hypernasality as the type of resonance disorder and one had no resonance disorder, the results of the severity of hypernasality of the 3 participants after performing descriptive analysis has been summarized in **table 5**.

Intra- and Inter-examiner reliability

Table 6 shows the results of Intra- and Inter-examiner reliability.

Discussion

Historically, speech characteristics have been an integral part of any description of sequelae of cleft palate¹⁹. In this study, 75% of the UCLP participants presented with hypernasality and 25% of them had no hypernasality. This results were comparable with findings from a study conducted by Normasturaet al¹⁷ where hypernasality occurred in 75% of BCLP children and 57.7% of UCLP children. Kuehn and Moller¹⁹ reported that excessive nasality or hypernasality is probably the signature characteristics of persons with cleft palate. As seen in this study, most of the

participants exhibited hypernasality as the resonance disorder. None of them had hyponasality, mixed resonance or cul-de-saq. This is supported by a study done by Normasturaet al¹⁷ where the only speech abnormality in BCLP and UCLP children noted was hypernasality. Brunnegard & Lohmander²³ also stated that the prevalence of hypernasality in the UCLP group of their study was higher than in other comparable published materials which was evident in the current investigation.

There are many factors that can lead to the presence of hypernasality. Hypernasality is most often caused by either having a fistula or impaired velopharyngeal closure. As there was no evidence of fistula in the participants of this study through oral motor examination, another possible cause to hypernasality in the participants could have been impaired velopharyngeal closure. This term encompasses two main causes of the impairment of velopharyngeal closure²⁴. The first is velopharyngeal insufficiency which indicates structural abnormalities and the second is velopharyngeal incompetency which indicates functional abnormalities. Either one of them could have caused hypernasality, as the impairment in velopharyngeal closure causes sounds that should be resonated in the oral cavity to resonate in the nasal cavity especially during production of vowels and voiced consonants. However, the cause of impaired velopharyngeal closure is best determined through instrumental assessments such as nasopharyngoscopy for direct imaging which would confirm the cause of hypernasality. Although all participants of this study had already undergone lip surgery and palatoplasty, this is to decide if the particular individual should proceed for further surgical intervention if the cause is of velopharyngeal insufficiency or proceed with speech therapy if the cause is of velopharyngeal incompetency.

Another factor that can lead to hypernasality is hearing problems. According to Sharma and Nanda²⁵, hearing loss is a well-known complication in cleft individuals. Hearing loss in turn will then lead to a distorted speech sound production because the way speech sounds are perceived will be affected. Intact hearing is a crucial aspect in speech sounds development and also in resonance. Thus, it was made sure that the participants involved in the research study were those who had no developmental hearing problems. This exclusion criterion was to eliminate the possibility of including participants who could have resonance disorders due to hearing difficulties. Thus, it is suffice to say that the hypernasality

exhibited by the participants in this study was not caused by any hearing problems.

Results of this study also indicated that 75% of the participants who exhibited hypernasality had a severity rating ranging from mild to severe. A study by Abdullah as cited by Normastura et al., (2008) on Malaysian CLP patients in Hospital Universiti Kebangsaan Malaysia (HUKM) also found that 73.7% of BCLP patients demonstrated mild to severe hypernasality. All of the participants in this study had attended speech therapy for a certain period but have been defaulted later on due to unknown reasons. Only one participant who attended speech therapy for 1 year showed no evidence of resonance disorder. This finding was almost similar to outcomes from another study conducted by Hardin Jones and Jones²⁶⁾ where 37% of the children demonstrated moderate to severe hypernasality despite having undergone speech therapy.

One factor that normally contributes to the difference in severity of resonance disorder is because of the duration of undergoing speech therapy. The longer the duration of attending speech therapy, the better the speech outcome will be. However, in this study one of the participant that has been undergoing speech therapy for 4 years still exhibited severe hypernasality while another participant who had been undergoing speech therapy for 1 month demonstrated mild hypernasality. Although there is a difference between the duration of undergoing speech therapy and the severity rating of the participant, the frequency in which participants went for speech therapy was not known.

In consequence, perceptual speech assessment is central to the evaluation of speech outcomes associated with cleft palate and velopharyngeal dysfunction²⁷⁾. Information such as the types and severity of resonance disorders gathered through perceptual assessment can act as a baseline for SLPs to further manage a CLP case. From the perceptual assessment, the SLP would be able to choose the

correct technique to be used for therapy to tackle the specific type of resonance disorder. In addition, the data serves as a basis for decision making for secondary surgery if needed by the particular individual and also determine the frequency of speech therapy needed.

Conclusion

The research data revealed that 75% (3/4) participants exhibited hypernasality as the type of resonance disorder and 25% (1/4) participant had no evidence of hypernasality or any other types of resonance disorder. Likewise, no other types of resonance disorders such as hyponasality, mixed resonance or cul-de-saq was noted in the participants of this research study. This indicated that hypernasality is one of the most common resonance disorder that is being exhibited.

Correspondingly, the severity of the participants' hypernasality ranged from mild to severe. This could be influenced by the frequency of speech therapy attended by the participants and the age at which they started speech therapy.

Other than that, this study revealed inter-rater reliability of fair to almost perfect agreement with kappa values and an exact agreement ranging from 50% to 100%. On the other hand, intra-rater reliability showed almost perfect agreement with kappa values and an exact agreement of 100%. Similarities in perceptual ratings of resonance disorders by the SLPs and the researcher herself indicates a high reliability in the assessment

Abbreviations

CLP- cleft lip and palate
 UCLP- unilateral cleft lip and palate
 SLPs- speech-language pathologists
 IPA- International Phonetic Alphabets

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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 |
| Total | 11 | 10 |

Table 2. The grading system (severity rating) of Hyper-nasal resonance and hypo-nasal resonance.

| Hyper-nasal resonance | |
|-----------------------|-------------------------------------------------------------------------------------------------------------------------------|
| Grade 0 | Normal tone |
| Grade 1 | Hyper-nasal resonance perceived on vowels and approximants |
| Grade 2 | Hyper-nasal resonance perceived on vowels and approximants, have weakened consonants with nasalization of voiced consonants |
| Grade 3 | Includes all of the above characteristics of hyper-nasality and the replacement of /b d g/ by their nasal equivalents /m n N/ |
| Hypo nasal resonance | |
| Grade 0 | Normal tone |
| Grade 1 | Moderate hypo-nasality where nasal consonants are slightly denasal |
| Grade 2 | Nasal consonants are perceived as plosives |

Table 3. 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 4. Types of resonance disorders exhibited by participants

| Type of Resonance Disorders | No. of Participants |
|-----------------------------|---------------------|
| Hypernasal | 3 |
| Hyponasal | 0 |
| Mixed Resonance | 0 |
| Cul-de-saq | 0 |
| None | 1 |

Table 5. Severity and consistency of hypernasal resonance of the 3 participants

| Participant | Severity of Hypernasality | Consistency of Hypernasality |
|-------------|---------------------------|------------------------------|
| 1 | Grade 2 | Inconsistent |
| 2 | Grade 3 | Consistent |
| 3 | Grade 1 | Inconsistent |

* Grade 0-None, Grade 1-Mild, Grade 2-Moderate, Grade 3-Severe

Table 6. Intra- and Inter-examiner reliability

| 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% | 0.33 |
| 1 and 3 | 100% | 1.00 |

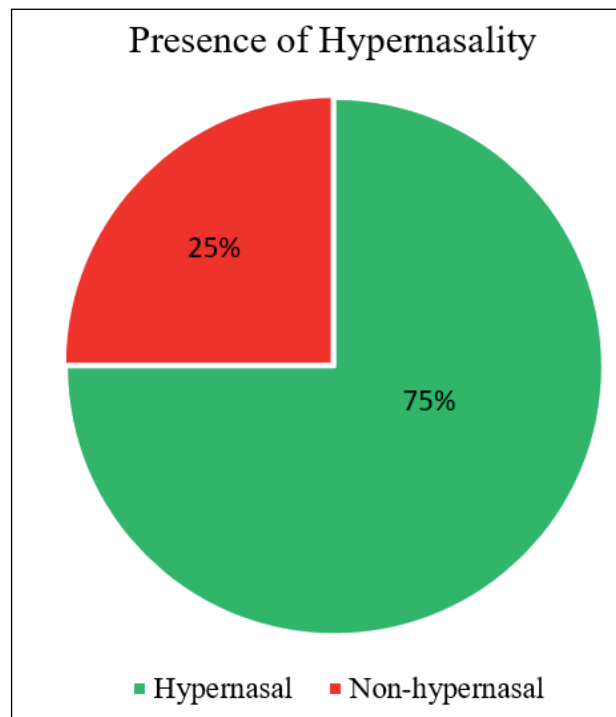


Figure 1: Presence of hypernasality among the participants

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