# Speech Outcome Analysis after Primary Cleft Palate Repair: Interim Siriraj Hospital Audit

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## ABSTRACT

**Objective:** To evaluate the speech outcomes after primary cleft palate repair in a single tertiary medical institution of Thailand.

**Materials and Methods:** A prospective cohort study was performed. Patients who had cleft palate with/without cleft lip and underwent primary cleft palate repair were included. Speech assessment was performed using the Pittsburgh weighted speech score (PWSS) by a speech-language pathologist.

**Results:** Forty patients (21 males and 19 females) who underwent primary cleft palate repair at Siriraj Hospital were included. The median age at the time of speech evaluation was 7 years. The median age at primary cleft palate surgery was 12 months. The predominant cleft palate type was Veau 3 (47.5%). Oronasal fistula occurred 40%. Two-flap palatoplasty and intravelar veloplasty were the most common procedures. Median PWSS was 7, in which the competence velopharyngeal mechanism was found 5%, borderline competence 10%, borderline incompetence 32.5%, and incompetence velopharyngeal mechanism 52.5%. Among the velopharyngeal incompetence group, articulation disorder was the most common disorder with median score of 3. Besides, the median scores for nasality, nasal emission, phonation, and facial grimace disorder were 1, 2, 0 and 0, respectively. There was no statistically significant association between velopharyngeal incompetence and cleft types, age at primary surgery, type of operation, the width of cleft palate and prevalence of postoperative oronasal fistula or otitis media effusion.

**Conclusion:** Velopharyngeal incompetence has been commonly identified after cleft palate repair in our institute. The articulation disorder is the most common characteristic.

**Keywords:** Cleft palate; speech outcome; velopharyngeal insufficiency; craniofacial abnormalities (Siriraj Med J 2021; 73: 744-751)

## INTRODUCTION

Cleft lip and/or cleft palate (CLP) are common craniofacial anomalies in Thailand. Chuangsuwanich et al., in 1998, reported that the incidence of patients with CLP was around 1 in 600 per live births at Siriraj Hospital.[1] Recently, Chowchuen et al. also reported a prevalence of CLP of 1.93 per 1000 live births in the northeast region of Thailand.[2] Patients with CLP have cosmetic and functional concerns, which typically require comprehensive multidisciplinary team (MDT) care and long-term follow-up, starting with cleft lip repair at around 3-6 months and cleft palate repair between 6-18 months of age.

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Cleft palate (CP) repair aims to create an anatomically intact and functional palate to improve feeding, achieve normal speech, and minimize maxillary growth restriction and middle ear infection.[3] In order to produce normal speech, a child must have velopharyngeal competence, defined as the ability to completely close the velopharyngeal sphincter, which separates the oro- and nasopharynx. The absence of this ability is termed velopharyngeal insufficiency.[4] The primary effects of velopharyngeal insufficiency are nasal air escape and hypernasality. A wide range of postoperative velopharyngeal insufficiency cases requiring secondary operation have been reported (15%–50%).[4-5]

The phonemic system in the Thai language is different from English. A Thai syllable consists of an initial, a vocalic nucleus, a final, and a tone.[6] The Thai language is a tonal language with sentences typically comprising a subject, verb, and object in order. The subject is usually not obviously stated but contextually assumed, similar to the case with the object. The verb has no declensions, tense, or conjugations.[7] According to this, if cleft palate patients have a decreased intelligibility of speech expression or are especially prone to articulation errors, it may disturb their communication skills, resulting in difficult social circumstances and communications.

Perceptual evaluation of speech by an experienced speech-language pathologist remains a widely accepted standard tool.[8-9] The Pittsburgh weighted speech score (PWSS), originally described by McWilliams and Phillips[10], is one of the key standardized methods used for a perceptual speech assessment. This tool rates the severity of velopharyngeal incompetency (VPI) by evaluating five speech components: nasality/resonance, nasal air emission, facial grimace, phonation/voice, and articulation.

Due to the lack of postoperative speech outcomes after CP repair in our centre, we conducted this study using PWSS to evaluate the postoperative speech outcomes after primary palatal repair.

## MATERIALS AND METHODS

### Patient enrollment

This research involved a prospective cohort study. Ethical approval was granted by the Institutional Review Board committee, Faculty of Medicine Siriraj Hospital, Mahidol University (Si 382/2018(EC2)). Informed consent was obtained. Patients who were diagnosed with CP with/without CL and underwent primary CP repair at Siriraj Hospital were enrolled. Exclusion criteria were syndromic patients with associated anomalies and patients who had follow-up time less than 3 years (co-operable assessment issue). After reviewing medical records, the eligible patients were contacted via telephone and invited for study participation.

### Speech outcome assessment

The speech evaluation was conducted between October 2019 and 2020. Perceptual speech outcome assessments using PWSS were conducted face-to-face by a qualified speech-language pathologist (K.L.) (Fig 1). Five components were investigated: nasal air emission, facial grimace, nasality/resonance, phonation/voice, and articulation. The sum of scores equal to 0 indicated velopharyngeal competency, 1-2 indicated borderline velopharyngeal competency, 3-6 indicated borderline VPI, and ≥7 indicated VPI.

### Data collection

The patients' medical records were reviewed to collect the following data: age and weight at primary palatoplasty, gender, cleft palate type based on Veau classification[11], cleft gap, techniques used in hard/soft palate procedures, the use of Vomerine flap, operation time, estimated blood loss, hospital length of stay, and the incidence of postoperative oronasal fistula (ONF) and otitis media effusion (OME). The Veau classification[11] categorizes cleft palate into four groups: Veau 1 (defects involving the soft palate only), Veau 2 (defects involving the hard and soft palate), Veau 3 (defects involving the soft palate to the alveolus, usually with lip involvement), and Veau 4 (complete bilateral cleft palate). Intraoral examination and photography were performed to identify postoperative ONF, defined as an abnormal connection or hole between the oral and nasal cavities, while intentionally unrepaired anterior hard palate and lingual–alveolar or labial–alveolar fistulas were not defined as this particular condition.

### Statistical analysis

As appropriate, continuous variables were summarized using Mean ± standard deviation (SD) or Median (range). Categorical variables were summarized using counts with the percentage. The clinical and peri-operative characteristics between patients with and without VPI were compared using the Student's t-test or Mann-Whitney U test for continuous variables, while Pearson chi-square, Yates' continuity correction or Fisher's exact test was used for categorical variables, as appropriate. P-value of ≤0.05 was considered statistically significant. Statistical analyses were performed using PASW Statistics for Windows, Version 18.0. Chicago: SPSS Inc.

**Nasal Emission:**

Not present _____0

Inconsistent, Visible _____1

Consistent, Visible _____2

Nasal escape on nasals appropriate _____0

Reduced _____0

Absent _____0

Audible _____3

Nasal Turbulence _____3

******Summary: Nasal emission scores: _____***

**Phonation:**

Normal ____0

Hoareseness or Breathiness

Mild ____1

Moderate ____2

Severe ____3

Reduced Loudness ____2

Tension in system ____3

Other_____

******Summary: Phonation scores:** ____*

**Facial Grimace**: ____2

**Nasality:**

Normal ____0

Mild Hypernasality ____1

Moderate Hypernasality ____2-3

Severe Hypernasality ____4

Hypo-Hypernasality ____2

Cul de sac resonance ____2

Hyponasality ____ 0

******Summary: Facial grimace and nasality scores:** ____*

**Articulation:**

Normal ____0

Development errors ____0

Errors from other causes not related to VPI ____0

Errors related to anterior dentition ____0

Reduced intraoral pressure for sibila nts ____1

Reduced intraoral pressure for other fricatives ____2

Reduced intraoral pressure for plosives ____3

Omission of fricatives or plosives ____2

Omission of fricatives or plosives plus hard glottal attacks for vowels ____3

Lingual-palatal sibilants ____2

Pharyngeal fricatives, snorts,inhalation or exhalation substitutions ____3

Glottal stops ____3

Nasal substitutions for pressure sounds ____4

******Summary: Articulation scores:** ____*

******TOTAL SCORE (from all sections above): ____

**Speech Indicates:**

____0 Competent Velopharyngeal Mechanism

____1-2 Competent to Borderline Competent

____3-6 Borderline to Boderline Incompetent

____7 Incompetent Velopharyngeal Mechanism

**Fig 1.** Perceptual speech evaluation form adapted from the Weighted Values for Speech Symptoms Associated with Velopharyngeal Incompetence of the Cleft Palate Center of the University of Pittsburgh[10]

## RESULTS

Forty patients (21 males and 19 females) with CP between 3-15 years of age who underwent primary cleft palate repair in Siriraj Hospital were included in the study. The median age at primary palatoplasty was 12 months old (range 8-40). Veau type 3 was found in 47.5% of patients, while Veau type 4, type 2, and type 1 were found in 30%, 15%, and 7.5%, respectively. The mean cleft gap width was 12.3 mm. The Bardach two-flap palatoplasty technique was performed in 94.6% of patients, while 5.4% underwent Veau-Wardill-Kilner palatoplasty for hard palate surgery. Intravelar veloplasty was performed in 82.5% of patients, while 2.5% underwent radical intravelar veloplasty, and 12.5% underwent Furlow z-plasty for soft palate surgery. One patient (2.5%) has unknown detail of the soft palate procedure. The vomerine flap was used in 42.5% of patients. The mean operation time was 127 min. Table 1 summarizes the patients' demographic data.

At the time of speech evaluation, the median age was 7 years (range 3-15 years). Among 40 patients, 21 (52.5%) had VPI, while 13 (32.5%) had borderline VPI, and 4 (10%) had borderline velopharyngeal competency, and 2 (5%) had velopharyngeal competency. The median total PWSS score was 7.0 (0-18). Median score for nasal emission was 2 (0-3), phonation was 0 (0-5), facial grimace was 0 (0-2), nasality was 1 (0-4), and articulation was 3 (0-10) (Table 2). There was no significant difference in age at primary palatoplasty, or for the type and width of cleft palate, type of surgery for hard and soft palate, or incidence of postoperative ONF and OME between patients with or without VPI (data shown in Table 3).

Overall, 16 patients (40%) had postoperative ONF. All of those underwent fistula closure operations. However, 8 patients had recurrence fistula at the time of speech evaluation. The median interval between primary CP surgery and ONF occurrence was 22 months (range 3-40 months). OME was found in 30 patients (75%).

## DISCUSSION

We present interim data on the perceptual speech outcomes after primary cleft palate repair from a tertiary referral centre in Thailand. We found over half of our patients with CP had postoperative VPI after primary CP repair at around 7 years of age. Moreover, making articulation errors was the most common characteristic affecting achieving a higher PWSS.

Normal speech achievement is one of the most important goals of CP surgery. Perceptual evaluation (i.e., listening) is the "gold standard" clinical assessment method for speech and voice disorders in the cleft palate population, and has been used in several previous studies.[8,12-16] In this study, VPI was identified by perceptual speech evaluation using PWSS in 53% of patients. This rate is higher than those reported in prior studies, ranging from 15% to 45%.[4-5] Articulation errors were found to be the most common speech distortions in our series. Pratanee et al., in 2016, found that articulation errors were the most common speech and language defects in Thai cleft palate patients.[17] Oopanasak et al. organized a case-control study of Thai children aged 6-13 years old and found that patients with CLP had significantly higher articulation defects than normal children, with velar and trill errors the most common articulation patterns.[18] Another study in Saudi Arabic-speaking children aged between 6-15 years old by Albustanji et al. found speech abnormalities, including articulation, hypernasality, and resonance, in 74% of patients after CP repair.[19] A study from Korea reported that 20% of patients had postoperative VPI and 50% demonstrated articulation deficits.[20] Recently, a study of Arabic-speaking Egyptian children between 3-9 years old demonstrated that articulation disorders, especially substitution, were the most common errors in CP patients with VPI.[21]

There are many factors involved in articulation. Every element of the speech apparatus, including the lips, teeth, palate, tongue, velum, and larynx, are engaged in producing intelligible sounds.[22] Patients in our study were evaluated at 7 years old, which is in the mixed dentition phase. Abnormal dental alignment (e.g., severe crowding), including transverse maxillary collapse during this time, maybe a causative factor in articulatory disorders in cleft palate patients. Another factor, including the prevalence of remaining alveolar cleft during this particular phase, may interfere with the incidence of articulation errors. Significantly, most children with CP in our study did not receive any regular long-term speech therapy after surgery. Although speech therapy in Thailand is offered to cleft palate patients free of charge due to our universal health care program, regular long-term speech therapy can be burdensome. Ideally, patients are required to attend 30-minute speech therapy sessions at least every 2-3 months for several years. Further, the speech therapy service is only available in a restricted number of tertiary referral hospitals. In addition, as the patients are children, their parents need to accompany them to the hospital for the service. This would cost them transportation expenses and a need to miss work, resulting in reduced income. Therefore, access to and take-up speech therapy in our patients is limited, especially when considering the long distance to the service and socioeconomic status of many of our patients' families.[23-24] Unsurprisingly, many

**TABLE 1.** Demographic data of 40 patients with cleft palate who underwent primary cleft palate repair.

| Characteristics | (Total=40) |
|---|---|
| **Genders, number (%)** | |
| Male | 21 (52.5%) |
| Female | 19 (47.5%) |
| **Surgeons** | |
| Attending staff | 33 (82.5%) |
| Trainees | 7 (17.5%) |
| **Age at primary palatoplasty (months), Median (range)** | 12 (8-40) |
| **Age at speech assessment (years), Median (range)** | 7 (3-15) |
| **Cleft types, number (%)** | |
| Veau type I | 3 (7.5%) |
| Veau type II | 6 (15%) |
| Veau type III | 19 (47.5%) |
| Veau type IV | 12 (30%) |
| **Cleft gap width (mm.), Mean ± SD** | 12.3 ± 4.1 |
| **Type of hard palate procedure, number (%)** | Total 37 |
| Two-flap palatoplasty | 35 (94.6%) |
| Veau–Wardill–Kilner palatoplasty | 2 (5.4%) |
| **Type of soft palate procedure, number (%)** | Total 40 |
| Intravelar veloplasty | 33 (82.5%) |
| Radical intravelar veloplasty | 1 (2.5%) |
| Furlow Z-plasty | 5 (12.5%) |
| Unknown detail of the procedure | 1 (2.5%) |
| **Vomerine flap use, number (%)** | 17 (42.5%) |
| **Blood loss (ml.), Median (range)** | 20 (1-150) |
| **Surgery duration (minute), Mean ± SD** | 127 ± 49 |
| **Hospital stay (days), Mean ± SD** | 4 ± 1 |
| **Presence of oronasal fistula, number (%)** | 16 (40%) |
| **Presence of otitis media effusion, number (%)** | 30 (75%) |
| **Speech indicates, number (%)** | |
| Velopharyngeal competency | 2 (5%) |
| Borderline velopharyngeal competency | 4 (10%) |
| Borderline VPI | 13 (32.5%) |
| VPI | 21 (52.5%) |

**TABLE 2.** Modified weighted values for speech symptoms associated with velopharyngeal incompetence of the Cleft Palate Center of the University of Pittsburgh by perceptual evaluation.

| Modality | Score |
|---|---|
| Total : Median (range) | 7 (0-18) |
| Articulation : Median (range) | 3 (0-10) |
| Nasality : Median (range) | 1 (0-4) |
| Nasal emission : Median (range) | 2 (0-3) |
| Phonation : Median (range) | 0 (0-5) |
| Facial grimace : Median (range) | 0 (0-2) |

**TABLE 3.** Various parameters and the association with VPI.

| Parameter | PWSS < 7 (n= 19) | PWSS ≥ 7 (n= 21) | P-value |
|---|---|---|---|
| **Age at primary palatoplasty, Median (range) (months)** | 12 (8-40) | 12 (9-39) | 0.539 |
| **Cleft types, number (%)** | | | |
| Veau type I | 3 (15.8) | 0 (0) | 0.238 |
| Veau type II | 3 (15.8) | 3 (14.3) | |
| Veau type III | 7 (36.8) | 12 (57.1) | |
| Veau type IV | 6 (31.6) | 6 (28.6) | |
| **Cleft gap width, Mean ± SD (mm.)** | 12.2 ± 3.4 | 12.3 ± 4.7 | 0.909 |
| **Type of hard palate procedure, number (%)** | | | |
| Two-flap palatoplasty | 16 (84.2) | 19 (90.5) | 0.495 |
| Veau–Wardill–Kilner palatoplasty | 0 (0) | 2 (9.5) | |
| **Type of soft palate procedure, number (%)** | | | |
| Intravelar veloplasty | 13 (68.4) | 20 (95.2) | 0.130 |
| Radical intravelar veloplasty | 1 (5.3) | 0 (0) | |
| Furlow Z-plasty | 4 (21.1) | 1 (4.8) | |
| **Vomerine flap use, number (%)** | 7 (36.8) | 10 (47.6) | 0.713 |
| **Presence of oronasal fistula, number (%)** | 6 (31.6) | 10 (47.6) | 0.477 |
| **Presence of otitis media effusion, number (%)** | 15 (78.9) | 15 (71.4) | 1.000 |

factors, not only velopharyngeal anatomical deficiencies, are associated with VPI in our patients in this centre.

Our study revealed no significant association between age at primary surgery, types and width of cleft palate, type of CP surgery, or incidence of postoperative ONF, OME, and severity of VPI. These data are consistent with a previous study from a tertiary university hospital in Northeast Thailand.[25]

Postoperative ONFs in cleft palate have been reported in 9%-50% of cases.[26-27] The ONF rate in the present study was 40%, which is consistent with previous literature.[28-31] A large case series from Khon Kaen University, Thailand, revealed that ONF formation was significantly associated with higher types of Veau classification, syndromic cleft patients, and a cleft width more than 11.5 mm.[32] In our series, we found that the majority of patients had Veau type 3 or 4 (77%), and the mean cleft gap was 12.3 mm. From our study, 48% of patients with VPI had postoperative ONF. Although an ONF closure procedure was performed in all these patients, 50% still had residual fistula at the time of the study. In the borderline VPI group, 38% of patients had ONF. Also, 60% of those still had residual ONF at the time of the study despite a fistula closure procedure having been performed. There were no ONF cases in the velopharyngeal competency and borderline velopharyngeal competency group. ONF might be the cause for this, producing a higher hypernasality score in the PWSS.

OME is common in CLP, often resulting in hearing loss, speech delay, and learning disabilities. If untreated, chronic otitis media or cholesteatoma may occur, leading to permanent middle ear damage. In our hospital, the prevalence of OME was reported by Ungkanont et al. to be around 50%–80%.[33] They also found a significant improvement in the audiograms after palatoplasty. In this series, we found 75% of patients had OME. This condition might be another confounding factor that affected the poor speech outcomes in this study, despite the statistical analysis suggesting no significant association between OME and speech outcomes in this study. Further subgroup analysis should be performed to confirm this outcome.

We realize that one of the significant limitations of our study was the small population sample. We had previously planned to enrol a study population of 200 by calculating the predicted sample size using nQuery Advisor (San Diego, CA) and by assuming the rate of velopharyngeal incompetency as 37.5% with confidence interval of 95%. However, due to the COVID-19 pandemic in Thailand during 2020, we, unfortunately, could only enrol 40 patients to take part in face-to-face speech evaluation. Ongoing speech evaluations are continuing for a complete long-term speech outcome assessment.

This interim study should, however, be of benefit for our hospital and can reinforce the need for MDT care, including cleft surgeon, speech pathologist, ear nose throat surgeon, and dental team, which is mandatory for all patients with CLP. Objective investigations, such as nasendoscopy and/or video-fluoroscopy, should be further used in patients who have VPI from initial perceptual speech evaluation as the diagnostic tools and preoperative planning before any subsequent correction procedure.

## CONCLUSION

Postoperative VPI is common after cleft palate repair in our hospital. The articulation disorder is the most common characteristic affecting speech outcomes. We encourage establishing a cleft and craniofacial centre to deliver MDT care in our hospital to achieve the best benefits in CLP care.

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