

Velopharyngeal Insufficiency: Subjective and Objective Assessments

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Objective: To determine 1) the role, function of structures, and patterns of Velopharyngeal [VP] gap formation among people with Cleft lip and palate [CLP], and 2) the inter-reliability between 2 investigators on the objective assessment of VP gap and its respective patterns.

Materials and Methods: This was a descriptive and retrospective study data collection among. Patients with velopharyngeal insufficiency [VPI]'s medical records were retrieved. Outcomes of perceptual assessment and objective evaluation of velopharyngeal movement (using Motus plus 2.0 software) were analyzed. Descriptive analyses, intra-class correlations, and Kappa coefficient were used to characterize the data.

Results: Twenty-three participants between 8 and 47 years of age were enrolled. Oronasal fistula was found 23.81% Coronal patterns of the VP gap and tongue compensation movement were common in patients with VPI. The respective correlation coefficient between the perceptual resonance rating scale and nasalance scores for Thai standard passages My house, Winter, and Laying Hen was 0.37 (95% CI = -0.06, 0.68), 0.26 (95% CI = -0.19, 0.62), and 0.30 (95% CI = -0.14, 0.64). The respective correlation coefficient between the nasalance scores for the Thai standard passages and articulation types was -0.29 (95% CI = -0.63, 0.16), -0.41 (95% CI = -0.71, 0.03), and -0.19 (95% CI = -0.57, 0.25). The correlation coefficient for articulation types and the perceptual resonance rating scale was 0.08 (95% CI = -0.22, 0.58). The intra-class correlation coefficient between the two investigators for VP gap for each sound was 0.78 to 0.82, while the range of kappa coefficient between the two investigators for the VP pattern gap for each sound was 0.16 to 0.30.

Conclusion: Velum had the primary role vis-a-vis VP function while the posterior pharyngeal wall had minimal function in VP gap formation. Coronal patterns of VP gap and tongue compensation movement were common in patients with VPI.

Keyword: VPI, Cleft lip and palate, Objective measurement, Oronasal fistula

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Velopharyngeal function [VPF] is generally formed by the levator veli palatini muscle that is responsible for the elevation and retraction of the velum in order to move it upward and backward and contact the pharyngeal wall⁽¹⁾. Complete velopharyngeal closure is required during production of “oral” phonemes, such as plosives (/p, p^h, t, t^h, k/), fricatives (/s, f/), or all consonants except for the nasal consonants (/m, n, ŋ, _m, _n, _ŋ/).

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Velopharyngeal insufficiency [VPI] refers to failure of movement of the velum and lateral and posterior pharyngeal walls to separate the oral cavity from the nasal cavity during production of oral (non-nasal) sounds because of the inability of the velopharyngeal sphincter to completely close. The primary effects of VPI are nasal air escape and hypernasality. The secondary effects of VPI include speech articulation errors (i.e., distortions, substitutions, and omissions). These effects will reduce the intelligibility of speech⁽²⁾. VPI is common in people with cleft lip and palate [CLP]. The prevalence of VPI/hypernasality is ~43% (95% CI = 36.58 to 49.93)⁽³⁾. Success of VPI correction is primarily assessed by the quality of speech. Speech assessment by a qualified

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speech and language pathologist [SLP]-including video-nasopharyngoscopy [VNP], video-fluoroscopy [VFS], and nasometry-are all used to determine the necessity of performing pharyngoplasty and designation of flap reconstruction. Assessment of VPI should, therefore, be generally considered as comprising three components: oral peripheral examination; perceptual evaluation; and instrumental assessment VNP is a technique for direct velopharyngeal functional examination that allows observation of the velopharyngeal port during speech using an endoscope. Movement of the soft palate, posterior and lateral pharyngeal walls, and the patterns of velopharyngeal closure are observed. It is common that the majority of cleft teams use this procedure for the assessment of VPI; however, the data obtained from VNP depends on the position and angle of the tip of the endoscope and requires subjective interpretation⁽⁴⁾. Although VNP has some limitations with regard to VPI assessment compared to VFS [i.e., abnormalities of the posterior pharyngeal wall, closure level of velopharyngeal valve (VP valve)], it provides excellent clarity and maneuver ability, and allows the operator to see the configuration of adenoid tissue fissures in the adenoid pad that are causing the firmness of closure⁽⁵⁾. In addition, it is a non-radiographic technique and easily performed with less cost than VFS. Conventional VNP is the gold standard for the screening and diagnosis among individuals at risk of VPI⁽⁶⁾.

An international working group established a system for quantifying, recording, and describing movement of the velum, lateral, posterior pharyngeal walls, as well as the relative size, shape, symmetry, and location of the velopharyngeal gaps⁽⁷⁾. Both VPN and VFS should be standardized based on their subjective measurement ratio. The challenge is getting an objective measurement of the VNP. The objective assessment for VPN has been developed and used in speech and ear, nose, throat clinics in 2010 at Srinagarind Hospital, Khon Kaen University, Thailand.

The aims of the current study were to determine: 1) the role of structures and patterns for VP gap formation among people with CLP; and, 2) the inter-reliability of objective assessments of the VP gap and their respective patterns between 2 investigators.

Materials and Methods

Study design

Descriptive study with retrospective data collection.

Participants

Inclusion criteria

People with VPI and referred for VPN registered at the Speech Clinic, Srinagarind Hospital, Faculty of Medicine, Khon Kaen University between February 2011 and June 2012. Reports of perceptual assessment, nasometry, and VPN were also available.

Exclusion criteria

Any person with cleft lip and palate whose report of VPN was not available.

Procedure

We retrieved reports of perceptual speech assessment, VNP, nasometry from the medical records of the Speech Clinic kept at the Department of Medical Record and Statistics.

In the current study, we used the Thai Speech Parameters for Patients with Cleft Palate in a Universal Reporting System, perceptual assessment of resonance disorders or VPI. This is a 5-level scale for evaluating hypernasality with respect to speech outcome among individuals with CLP (-1: hyponasality; 0 = within normal limits; 1 = mild; 2 = moderate; 3 = severe, x = missing data)^(4,8,9). Articulation types were divided into 4 classes: 0= normal; 1= function or phonological; 2 = compensatory; 3 = organic; and 4 = functional or phonological with compensatory articulation disorders. Voice was assessed as 0 = normal, or 1 = abnormal voice; Based on the number of words and a sentence level articulation test, intelligibility was divided into 3 types: 0 = intelligible (understandable $\geq 75\%$); 1 = intelligible if topic known (understandable 50 to 75%); 2 = unintelligible (understandable $< 50\%$).

The other outcomes were patterns of VP closures (i.e., coronal, sagittal, circular, and circular with Passavant's ridge). Consensus was used to resolve disagreements between the two investigators, and assessment by each investigator of (a) the areas of VP gaps (reported in mm²) by prolongation of /a/, /u/, /i/, and /s/, and (b) nasalance scores (reported as a percentage).

Patterns of the respective VP closure (viz., coronal, sagittal, circular, and circular with Passavant's ridge) (Figure 1)⁽⁷⁾ and VP gaps were visualized using fiberoptic nasopharyngoscopy (as well as by standard circle [diameter 0.5 mm]) while patients produced prolonged /a/, /u/, /i/, and /s/. Pictures of the VP gaps were captured between the 2nd and 3rd minute of prolongation, then the areas of VP gaps were measured (in μm) by Motic Images Plus 2.0 Program. Researchers

calculated real areas of VP gaps in each sound by comparison to the standard circle in square measured in millimicrometers (μm)⁽¹⁰⁾.

Statistical analysis

Descriptive analysis was used for data

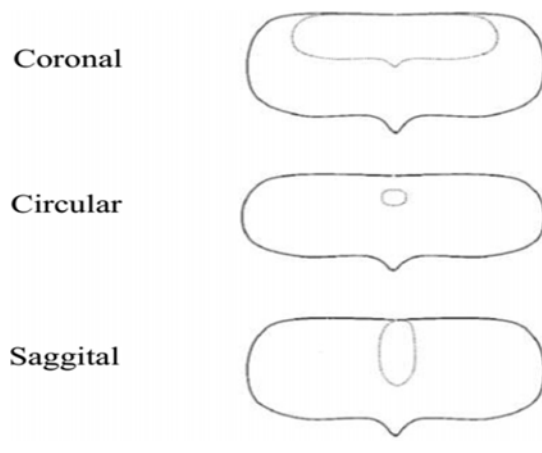


Figure 1. Patterns of the velopharyngeal gap.

Table 1. General characteristics of participants

ID No.	Age (years; months)	Gender	Diagnosis	ONF
1	47	Male	Sub-mucous cleft	1
2	11	Male	Sub-mucous cleft	0
3	13	Female	Bilat. CLP	0
4	24	Male	Bilat. CLP	N/A
5	16	Female	Bilat. CLP	1
6	11	Male	Bilat. CLP	0
7	11	Male	Lt. CLP	0
8	15	Male	Lt. CLP	0
9	13	Male	Rt. CLP	0
10	25	Female	Lt. CLP	0
11	8	Male	Lt. CLP	0
12	13	Female	Cleft palate	0
13	8	Female	Cleft palate	0
14	21	Female	Treacher Collins syndrome	0
15	9	Female	Sub-mucous cleft	0
16	8	Male	Bilat. CLP	1
17	12	Female	Rt. CLP	0
18	9	Male	Bilat. CLP	1
19	16	Female	Cleft palate	N/A
20	13	Female	Mid-facial Hypoplasia	0
21	14	Female	Bilat. CLP	1
22	17	Female	Bilat. CLP	0
23	12	Female	Bilat. CLP	0

ONF = oronasal fistula; N/A = Not available; Bilat. CLP = Bilateral cleft lip and palate; Lt. CLP = Left cleft lip and palate

presentation. Intra-class correlation and Kappa coefficient were used for analyzing the inter-reliability of objective assessments.

This study protocol was reviewed and approved by the Khon Kaen University Ethics Committee in Human Research.

Results

Twenty-three medical records which, reports of perceptual assessment, nasometry, and VPN were available, recruited in this study. The general characteristics of the participants are presented in Table 1. A speech and language pathologist [SLP] and otorhino-laryngologist performed and assessed VP function. Oral examination revealed that the oronasal fistula [ONF] rate was 23.8% (5/21) (95% CI 8.22, 47.17) based on the data available for 21 patients.

Speech abnormalities were assessed based on the Thai Speech Parameters for Patients with Cleft Palate⁽⁹⁾. The percentage of speech outcomes are presented in Table 2.

Participants who had performed a perceptual assessment and demonstrated resonance disorders

Table 2. Percentage of speech abnormalities

Variables	No.	Percentage	95% CI
Resonance			
Mild hypernasality	2	8.70	0, 21.15
Moderate hypernasality	17	73.90	54.50, 93.33
Severe hypernasality	4	17.40	0.63, 34.15
Articulation			
Normal articulation	3	13.00	0, 27.93
Functional/phonological	2	8.70	0, 21.15
Compensatory	4	17.40	0.63, 34.15
Functional and compensatory	14	60.90	39.29, 82.45
Voice			
Normal	19	82.60	61.22, 95.05
Abnormal	4	17.40	4.95, 38.78
Intelligibility			
Intelligible	21	91.30	78.85, 100
Intelligible if topic known	2	8.70	0, 21.15
Unintelligible	0	0	

were referred to an otorhinolaryngologist for an ENT examination and VNP. Data were available for 22 of the 23 patients. Patterns of the VP gap when patients produced prolonged vowels /a/, /u/, /i/, and /s/ were reported by consensus of a SLP and an otorhinolaryngologist. The perceptual assessment and percentage and patterns of velopharyngeal movement were retrieved from the medical records. Some data regarding the amount of tongue compensatory movement were missing. The objective measurement of the reference circle (diameter 0.5 cm) and /a/ are presented in Figure 2 and 3. Information regarding resonance and assessment of VPI are presented in Table 3. The data indicate that the velum had the main role in VP function and the posterior pharyngeal wall had minimal function vis-a-vis VP gap formation. Coronal patterns of the VP gap and tongue compensatory movement were common in patients with VPI.

The respective correlation coefficient for the relationship between the perceptual rating scale of resonance and the nasalance score for the Thai standard passages (My house, Winter, and Laying Hen) was 0.37 (95% CI = -0.06, 0.68), 0.26 (95% CI = -0.19, and 0.30 (95% CI = -0.14, 0.64). The respective correlation coefficient for the relationship between the nasalance scores of the Thai standard passages (My house, Winter, and Laying Hen) and articulation types was -0.29 (95% CI = -0.63, 0.16), -0.41 (95% CI = -0.71, 0.03), and -0.19 (95% CI = -0.57, 0.25). The correlation coefficient for the articulation type and perceptual rating scale of resonance was 0.082 (95% CI = -0.216, 0.576).



Figure 2. Reference circle.

The respective intra-class correlation coefficient with the 95% CI for VNP (based on the Motic Images Plus 2.0 Program) for the VP gap orifice for each vowel for both investigators are presented in Table 4. The respective kappa coefficient and 95% CI for the two investigators for the pattern of the VP gap for each sound are presented in Table 5.

Discussion

Most of the participants in the current study were diagnosed as having bilateral CLP 39.1% (9/23). Approximately a half of the patients (56.52%) were female. ONF as a complication of palatal surgery was

found in 23.80% a prevalence similar to a study done at our centre that revealed a ONF rate of 25%⁽¹¹⁾ and at



Figure 3. Prolongation /a/.

another center where the rate was 23.70%⁽¹²⁾. The occurrence of ONF in published reports varies widely, ranging between 3.40% and 15%^(13,14). The high rate of ONF in the current study might be due to the selection criteria which included only persons with VPI based on a perceptual assessment with need of further investigation. All patients with ONF had bilateral CLP that might require less tissue to repair palatal defects as compared to other types of CLP. Most of the participants had moderate hypernasality (Table 2), so the ONF rate was relatively high. Further study with a larger sample size should be conducted and should take into consideration the classification of CLP.

A large number of patients with VPI had articulatory disorders (87%, Table 2) mostly functional and compensatory types. Most participants were diagnosed with CLP (21 of 23 or 91.3%), so the current study agrees with previous studies which found that patients with CLP had a high risk of difficulties with functional articulation compared to the normal

Table 3. Perceptual assessment and percentage and patterns of velopharyngeal movement

No.	Perceptual assessment	Nasal emission*	Percentage of velopharyngeal movement				Pattern
			Velum	PPW	Left LPW	Right LPW	
1	+2	1	70	0	20	20	Coronal
2	+3	2	80	0	0	0	Sagittal
3	+2	2	50	20	25	25	Coronal
4	+2	1	5	0	5	5	Circular with tongue compensatory 90%
5	+3	0	5	20	10	10	Circular with Passavant's ridge
6	+2	1	10	5	5	5	Coronal
7	+2	1	50	5	5	20	Sagittal with tongue compensatory
8	+2	1	20	20	20	35	Circular
9	+2	1	5	0	10	5	Coronal with tongue compensatory 90%
10	+2	1	50	25	25	50	Coronal
11	+1	1	50	0	0	0	Coronal
12	+3	1	25	25	25	25	Circular and circular with Passavant's ridge
13	+2	1	70	10	25	25	Coronal
14	+3	2	0	0	5	0	Coronal tongue compensatory
15	+2	1	5	5	10	5	Circular
16	+2	2	10	0	10	10	Circular
17	+2	2	20	10	30	30	Circular with tongue compensatory 10%
18	+2	1	60	0	25	20	Coronal
19	+2	2	0	30	20	20	Coronal tongue compensatory
20	+1	2	80	0	0	0	Coronal
21	+2	2	N/A	N/A	N/A	N/A	N/A
22	+2	2	25	0	10	10	Coronal
23	+2	1	0	0	-20	20	Sagittal with tongue compensatory 80%

* 1 = sometimes or not frequent; 2 = frequent or invasive nasal emission; PPW = Posterior pharyngeal wall; Left LPW = Left lateral pharyngeal wall; Right LPW = Right lateral pharyngeal wall

Table 4. Intra-class correlation coefficients between two investigators for the VP gap for each sound

Sounds	VP gap (mm ²)
Rest	0.78, (0.49, 0.91)
/a/	0.87, (0.69, 0.95)
/u/	0.79, (0.51, 0.91)
/i/	0.82, (0.58, 0.93)

Table 5. Kappa coefficients between two investigators for the VP gap pattern for each sound

Sounds	Pattern
Rest	0.30 (0.00 to 0.48)
/a/	0.25 (0.00 to 0.71)
/u/	0.25 (0.00 to 0.61)
/i/	0.16 (0.00 to 0.23)

population^(15,16). For voice abnormality (Table 2), the rate was 17.40%. The prevalence was only 14.30% (3/21), if the determination were based on diagnosis of CLP alone (i.e., excepting No. 14 (hoarseness) and No. 20 (normal voice) who were not diagnosed as CLP) (Table 1), which puts our study in range of previous studies that had a prevalence of 5.50 to 25%^(3,17-20). Even though a previous study showed no relationship between VPI and hoarseness⁽²⁰⁾, the result of this study support that patients with CLP were at risk for dysphonia from compensation of hyperfunctional vocal cord.

Perceptual assessment and percentage and patterns of the velopharyngeal gap (Table 3) showed that the coronal type of VP gap and tongue compensatory movement (40.90% and 31.80%, respectively) were common among persons with VPI. Most of the VP function implicated the velum while the posterior pharyngeal wall had a secondary role for compensatory VP gap formation in persons with VPI. Tongue movement is also a major compensatory mechanism for persons with VPI. This information might be helpful when considering surgical approaches, like VPI reconstruction.

Twenty-two of 23 patients with VPI (95.70%) had nasal emission (Table 3). For people with CLP (with exception No. 14 and 20), majority of them had hypernasality, and the ones who had moderate hypernasality, also had nasal emission. Most of our patients had moderate hypernasality and supports the hypothesis that patients with CLP have nasal emission

caused by a small VPI with resistance from the VP gap while air flows to the nasal cavity^(21,22).

The correlation coefficients between the two investigators for VP orifice in each sound (Table 4) showed that there was good to excellent agreement (78 to 87%) for objective measurement of the VP gap using the Motic Plus 2.0 Program. Agreement of the kappa coefficient, however, was low to fair for the VPI pattern. The current study was the first and primary report of objective VPI assessment; thus, the criteria for measuring VP need corroboration through a larger group of participants and a larger group of investigators.

Conclusion

ONF was a common complication of palatal surgery for persons with CLP (14.3 %). Coronal patterns of the VP gap and tongue compensation movement were also common among persons with CLP. VP function for people with VPI is mainly determined by the velum. The velum had the main role vis-a-vis VP function and as such the posterior pharyngeal wall had minimal function in cases of VP gap formation. The findings of the current research are useful for planning VPI surgical reconstruction. The inter-reliability of objective assessments for patterns of the VP gap needs further study.

What is already known on this topic?

VPI is a common complication in CLP after palatoplasty. Patterns of the VP gap and tongue compensation movement have not been widely studied. To our knowledge, objective assessment of VP function has not been done.

What this study adds?

Coronal patterns of VP gap and tongue compensation movement are common in people with VPI. VP function is primarily determined by the velum. Objective assessment of VP function is possible. These findings are beneficial for planning surgical techniques in VPI reconstruction. Further study should be conducted with a larger number of participants to validate the reliability tests of objective measurement.

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Potential conflicts of interest

The authors no declare conflicts of interest.

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ภาวะความบกพร่องการทำงานของเพดานอ่อนและผนังคอหอย: การประเมินแบบนามธรรมและรูปธรรม

เบญจมาศ พระธานี, สวงศักดิ์ ธนาวิรัตน์นิจ

วัตถุประสงค์: เพื่อศึกษา 1) บทบาท การทำงานของโครงสร้างและรูปแบบการเกิดช่องเพดานอ่อนและผนังคอหอยและ 2) ความเที่ยงของการตรวจแบบรูปธรรมของช่องเพดานอ่อนและผนังคอหอย และรูปแบบระหว่างผู้ประเมิน 2 คน

วัสดุและวิธีการ: การศึกษาเชิงพรรณนาและการเก็บข้อมูลย้อนหลังภายใต้เกณฑ์การคัดเลือกเข้า คือ ผู้ป่วยปากที่มีภาวะความบกพร่องของการทำงานของเพดานอ่อนและผนังคอหอย เวชระเบียนของและรายงานของการตรวจช่องปาก การตรวจด้วยเครื่องวัดสัดส่วนพลังงานของเสียงที่ออกปากและจมูก (nasometry) และการส่องกล้องทางจมุกด้วยวิดีโอถูกสกัดข้อมูล ผลลัพธ์ของการประเมินการฟังเกี่ยวกับความบกพร่องของการทำงานของเพดานอ่อนและผนังคอหอย และการประเมินแบบรูปธรรมของการเคลื่อนไหวของเพดานคานอ่อนและผนังคอหอย (Motic plus 2.0 program) ถูกวิเคราะห์สถิติเชิงพรรณนา การวิเคราะห์ความสัมพันธ์แบบสัมพันธ์สัมพันธ์ (intraclass correlation) and Kappa coefficient ถูกนำมาใช้ในการแสดงผลการศึกษา

ผลการศึกษา: มีผู้เข้าร่วมการศึกษา 23 ราย มีอายุระหว่าง 8 ถึง 47 ปี มีอัตราของรูรั่วระหว่างจมุกและปาก ร้อยละ 23.81 รูปแบบการปิดแบบ Coronal ของเพดานอ่อนและผนังคอหอย และการเคลื่อนไหวแบบซเซของลิ้นเป็นสิ่งที่พบบ่อยในผู้ป่วยที่มีความบกพร่องของการทำงานของเพดานอ่อนและผนังคอหอย ค่าสัมประสิทธิ์สหสัมพันธ์ระหว่างสัมประสิทธิ์ความสัมพันธ์ของการประเมินด้วยการฟังการสั่นพ้องและค่าสัดส่วนพลังงานของเสียงที่ออกทางจมุกและปาก (nasalance scores) ของบทความมาตรฐานภาษาไทยเท่ากับ 0.37 (95% CI = -0.06, 0.68); 0.26 (95% CI = -0.19, 0.62); 0.30 (95% CI = -0.14, 0.64) สำหรับบทความบ้านของฉันทน์, ฤกษ์นาว และโก๋ออกไล่ตามลำดับ ค่าสัมประสิทธิ์สหสัมพันธ์ระหว่างค่าสัดส่วนพลังงานของเสียงที่ออกทางจมุกและปาก (nasalance scores) ของบทความมาตรฐานภาษาไทย และรูปแบบของการแปรเสียงเท่ากับ -0.29 (95% CI = -0.63, 0.16); -0.41 (95% CI = -0.71, 0.03); -0.19 (95% CI = -0.57, 0.25) ค่าสัมประสิทธิ์สหสัมพันธ์ระหว่างรูปแบบของการแปรเสียงและการประเมินด้วยการฟังการสั่นพ้องเท่ากับ 0.08 (95% CI = -0.22, 0.58) สำหรับค่าสัมประสิทธิ์สหสัมพันธ์ (intraclass correlation) ระหว่างผู้วิจัย 2 คนสำหรับ VP gap ในแต่ละเสียงมีค่าระหว่าง 0.78 ถึง 0.82 ในขณะที่ Kappa coefficient ระหว่างผู้วิจัย 2 คน สำหรับ VP gap ในแต่ละเสียงเท่ากับ 0.16 ถึง 0.30

สรุป: เพดานอ่อนเป็นอวัยวะที่ทำหน้าที่หลัก และผนังคอหอยด้านหลังเป็นอวัยวะที่ทำหน้าที่น้อยในการปิดช่องเพดานอ่อนและผนังคอหอย รูปแบบการปิดแบบ Coronal ของเพดานอ่อนและผนังคอหอย และการเคลื่อนไหวแบบซเซของลิ้นเป็นสิ่งที่พบบ่อยในผู้ป่วยที่มีความบกพร่องของการทำงานของเพดานอ่อนและผนังคอหอย
