Clinical Outcomes of Primary Palatoplasty in Preschool-Aged Cleft Palate Children in Srinagarind Hospital and Comparison with Other Standard Cleft Centers

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Background: *The purpose of this study was to evaluate the clinical outcomes regarding the rate of hypernasality and oronasal fistula formation in patients with cleft palate who underwent primary palatoplasty at our center and under our management protocol.*

Material and Medthod: *A Cross-sectional study of 40 consecutive non-syndromic cleft palates with/or without cleft lips, born between February 2007 and December 2008, who underwent primary palatoplasty at Srinagarind Hospital, Thailand. Demographic data that were recorded includes: patients with cleft types, age at palatoplasty, operating surgeons and surgical techniques.*

Results: *40 consecutive patients. There were 23 boys and 17 girls. Three patients had associated disease; one patient had amniotic band syndrome and clubfeet, two patients had G-6-PD deficiency. Mean age at time of evaluation was 5.7 years (5.0-6.9 years). Mean age at palatoplasty was 14.1 months (9-64 months). There were three plastic surgeons and plastic surgery residents. The predominant cleft lip type was Veau 3 (52.5%) followed by Veau 4 (27.5%) and Veau 1 (20%). Two-flap palatoplasty was used in all patients. The rate of hypernasality was 37.5% (15 out of 40 patients). Mild hypernasality was 25% and moderate hypernasality was 12.5%. Oronasal fistula occurred in 10 patients, fistula rate was 25%. Oronasal fistula closure was performed on nine patients (90%).Two patients (5%) had residual oronasal fistula at the time of the study. There were no statistically significant differences in the cleft types, age at palatoplasty and operating surgeons in hypernality rates and oronasal fistula formation.*

Conclusion: *The rate of hypernasality and oronasal fistula formation was comparable to results from other standard cleft centers in cleft palate patients who underwent primary palatoplasty during previous rounds of our management protocol.*

Keywords: *Cleft palate, Palatoplasty, Oronasal fistula, Hypernasality*

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Cleft lip with or without cleft palate was the most common congenital anomaly. The prevalence was 1 per 500 to 1 per 700 live births in Europe[1], 7.75 per 10,000 live births in the United States[2], 1.62 per 1,000 live births in Thailand[3] and 2.49 per 1,000 live births in North of Thailand[4]. The cleft palate management requires the long-term care of the multidisciplinary cleft team consisting of plastic surgeons, otolaryngologists, speech therapists, pediatricians, orthodontists and social workers.

The goal of palatoplasty was to achieve complete and intact closure of the palate resulting in avoidance of palatal fistula, restoration of the velopharyngeal sphincter for the development of normal speech, optimization of maxillary growth and also minimizing hearing loss and middle ear complications[5,6].

The 5-6 year-old cleft palate children about to enter school face a potentially traumatic period. The excessive hypernasality was probably the signature characteristic of persons with cleft palate[7]. The fact that half of these children had speech that was different enough to provoke comment, gives cause for concern[8]. The occurrence of oronasal fistula in the palate along the site of original closure represents a failure of surgical repair, may affect speech, socialization, and nutrition, and remain a challenging problem[9].

A number of audits of cleft palate management

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have been published by various centers worldwide. The outcomes of primary cleft palate repair were never properly studied in our center.

The main objective was to determine the rate of hypernasality and oronasal fistula formation. The cleft types, age at palatoplasty and operating surgeons influence on occurrence of hypernasality and oronasal fistula formation was determined.

Material and Method

Patients

A Cross-sectional study of 40 consecutive non-syndromic cleft palate with/or without cleft lip patients between December 2012 to February 2013, 5-6 year-old born between February 2007 and December 2008, who received primary palatoplasty by our cleft palate team at Srinagarind Hospital. The patients who experienced loss of medical records, did not respond to telephone or letter invitations, refused to enroll in study or could not perform speech assessment and palatal examination were excluded. The institutional review board gave approval for the present study.

Treatment protocol

Surgical management occurred at the following time intervals: cheiloplasty was performed at the age of 3-4 months in the case of cleft lip and cleft palate. Primary palatatoplasty was performed at the age of 9-12 months according to Two-flap palatoplasty in most patients. The surgical team consisted of three plastic surgeons and plastic surgery residents.

Orthodontic treatment was performed pre-surgically and subsequently depending upon the individual patient by orthodontists. Otorhinolaryngological examinations were performed by otorhinolaryngologists. In case of middle ear effusion, which required surgical treatment, myringotomy was accomplished simultaneously with primary palatoplasty. Our speech and language therapy program was performed at 1-6 years. Speech outcome was assessed at 4-6 years. Between 6 and 9 years, speech therapy was continued. Velopharyngeal insufficiency, which required secondary corrective surgery, was performed at approximately 6 years (4-9 years). Oronasal fistula closure was performed at any time of detection. Orthodontic management was performed at 6-9 years. Alveolar bone grafting was performed at 9-11 years and orthognathic surgery was performed at 18 years.

Demographic data

Demographic data including gender, co-morbidity, age at palatoplasty, age at time of evaluation, cleft type, type of surgical technique, operating surgeons was obtained from medical records. Patients who were determined to be syndromic, when diagnosed by a pediatrician who specialized in genetics, were excluded.

The Veau classification system was used to describe cleft type including Veau 1 (soft cleft palate), Veau 2 (hard and soft cleft palate), Veau 3 (soft and hard palates and unilateral cleft of the primary palate), and Veau 4 (soft and hard palates and bilateral clefts of the primary palate)[5]. Four operating surgeons were considered and treated as categorical data. Surgeons 1 through 3 were plastic surgeons, and surgeon 4 represented plastic surgery residents.

Speech assessment

Speech evaluation was determined by one qualified speech and language pathologist based in the Srinagarind Hospital with 20 years experience in managing patients with cleft lip and cleft palate. A perceptual speech assessment was estimated based on "Thai speech parameters for patients with cleft in a universal reporting system"[10], the perception of speech samples comprising nonsense syllables, Thai serial speech with high oral pressure consonants (counting from 1-20 and 40-50) with 4 simple sentences loaded with all of the consonants and 3 nasal sentences[11]. The aspects of speech outcome were assessed including resonance.

Each parameter of resonance was rated on a five-point scale[12]: -1 = Hyponasality ; 0 = normal; +1= mild hypernasality; +2 = moderate hypernasality and +3 = severe hypernasality. The hypernasality was defined as +1, +2 or +3.

Oronasal fistula

Oronasal fistula was defined as a patency between the oral and nasal cavities caused by failure of healing or a breakdown in the primary surgical repair of the palate; intentionally unrepaired lingual-alveolar and labial-alveolar fistulas are not included in the condition[13,14].

Any documentation of oronasal fistula from medical records was included in the group of patients with oronasal fistulas. Each patient had examinations by members of cleft palate team for intraoral examination, the original line of the cleft was visually inspected for oronasal fistula and intraoral photography while the evaluation was performed was used to identify residual fistula. Information about the location, size, symptoms,

operation for oronasal fistula closure, age at oronasal fistula closure and recurrence were reviewed.

Data analysis and statistics

The rate of hypernasality and oronasal fistula was reported to be in the 95% confidence interval (CI). The Chi-square and Fisher exact test was used to assess whether the development of hypernasality and oronasal fistula were influenced by cleft type, range of age at palatoplasty and operating surgeons. Comparisons of means were performed using t-test. The level of statistical significance was 0.05.

Results

There were 72 cleft palate patients with/or without cleft lip born between February 2007 and December 2008 who underwent primary palatoplasty at Srinagarind hospital. Two patients who had craniofacial syndrome consisting of facial cleft and Treacher collins syndrome were excluded. Forty consecutive patients were enrolled through patient birth date. There was a gap in the data of 30 patients made up of 20 patients, who had not responded to telephone and letter invitations along with the remaining 10 patients who refused to enroll in the study.

Of the 40 patients, 23 were male (57.50%) and 17 female (42.50%). Three patients had associated diseases consisted of two patients who had G-6-PD deficiency and one patient had amniotic band constriction with clubfeet. Mean age at the time of evaluation was 5.7 years (range, 5.0-6.9 years). The mean age at the time of primary palatoplasty was 14 months (range, 9 months to 64 months). Twenty-nine (72.5%) patients underwent palatoplasty at/or before 12 months of age, 7 (17.5%) patients had operation between 12-18 months and 4 (10%) patients had operations later than 18 months of age. The most prominent cleft type was Veau 3 (52.5%) followed by Veau 4 (27.5%) and Veau 1 (20%). All surgical techniques used for cleft palate repair was Two-flap palatoplasty. The palatoplasty was performed by four surgeons; surgeon 1 did 17 operations (42.5%), surgeon 2 did 4 operations (10%), surgeon 3 did 2 operation (5%) and surgeon 4 did 17 operations (42.5%). Nine of the patients (90%) received secondary palatal surgery for oronasal fistula closure. Two patients (5%) had residual oronasal fistula at time of evaluation. The demographic data was showed in Table 1.

Table 1. Demographic and clinical characteristics of patients with cleft palate with/without cleft lip who underwent primary palatoplasty

Characteristics	Number (%)
Number of patients	40
Male	23 (57.5%)
Female	17 (42.5%)
Comorbidity	2; G-6-PD deficiency 1; Amniotic band constriction, club feet
Mean age at time of evaluation	5.84 (±0.4) years
Mean age at palatoplasty	14.1 (±10.56) months
Age range for palatoplasty	
≤12 months	29 (72.5%)
12-18 months	7 (17.5%)
>18 months	4 (10%)
Cleft types	
Veau 1	8 (20%)
Veau 2	0
Veau 3	21 (52.5%)
Veau 4	11 (27.5%)
Operating surgeon	
Surgeon 1	17 (42.5%)
Surgeon 2	4 (10%)
Surgeon 3	2 (5%)
Surgeon 4	17 (42.5%)
Type of palatoplasty	
Two-flap palatoplasty	40 (100%)
Secondary palatal repair for fistula closure	9 (22.5%)
Residual fistula at time of evaluation	2 (5%)

Speech outcome

Thirteen patients had hypernasality speech. The rate of hypernasality was 37.5% (95% CI = 26.35-55.4). There were mild hypernasality in 10 patients (25%) and moderate hypernasality in 5 patients (12.5%). None of the patients had hyponasality or severe hypernasality (Fig. 1). Mean age at time of speech assessment in the patients with hypernasality was 5.63 years earlier than the patients who had normal resonance which were evaluated at 5.96 years (p = 0.017). Gender and co-morbidity were no difference among these patients. Two patients had residual oronasal fistula at the time of speech evaluation, both patients exhibited mild hypernasality. Hypernasality occurred in 4 out of 9 patients who had previous oronsal fistula closure, three patients had it to a mild degree and one patient had a moderate degree. The comparison data between patients with or without hypernasality are demonstrated in Table 2.

Speech outcome according to cleft types,

hypernasality occurred in 60% (9/15) of patients with Veau 3, 33.3% (5/15) in Veau 4 and 6.7% (1/15) in Veau 1. Patient with Veau 4 and Veau 3 had a higher overall rate of hypernasality than Veau 1. This difference was not statistically significant (p = 0.219) (Fig. 2) (Table 3). The majority of hypernasality occurred in patients who underwent operations by surgeon 1 (8 out of 15, 53%) followed by surgeon 4 (3 out of 15, 20%), surgeon 2 (2 out of 15, 13%) and surgeon 3 (2 out of 15, 13%). There were no statistically significant differences among surgeons in hypernasality rate (p = 0.068) (Fig. 3). The majority (86.6%) of patients who had hypernasality received palatoplasty before 12 months of age. The difference was not statistically significant in hypernasality among the patients who underwent palatoplasty before 12 months, between 12 and 18 months and older than 18 months (p = 0.444) (Fig. 4).

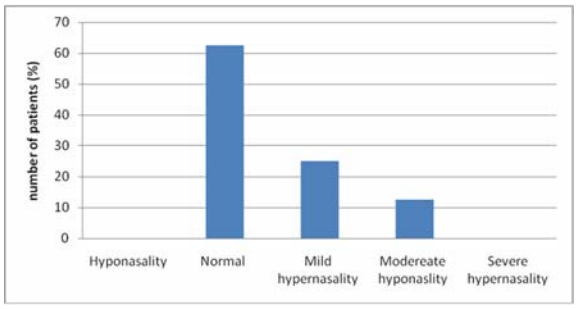

Fig. 1 Speech outcome in 40 preschool-age cleft palate patients who underwent primary palatoplasty.

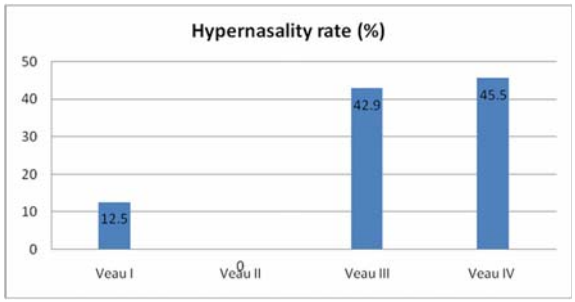

Fig. 2 Hypernasality rate according to cleft types.

Table 2. Characteristics grouped by occurrence of hypernasality

Characteristics	Normal resonance (n = 25)	Hypernasality (n = 15)	p-value
Male: female	14 (56%):11 (44%)	9 (60%): 6 (40%)	0.804
Comorbidity	1	2	0.544
Mean age at speech evaluation (year)	5.96 (±0.4)	5.63 (±0.42)	0.017
Mean age at palatoplasty (month)	14.0 (±8.38)	14.27 (±13.8)	0.938
Residual ONF at time of speech assessment	0	2	0.134
Previous fistula closure	5	4	0.705

Table 3. Data analysis grouped by occurrence of hypernasality

Characteristics	Normal resonance (n = 25)	Hypernasality (n = 15)	p-value
Age at palatoplasty			0.444
≤12 months	16 (55.2%)	13 (44.8%)	
12-18 months	6 (85.7%)	1 (14.3%)	
>18 months	3 (75%)	1 (25%)	
Cleft types			0.261
Veau 1	7 (87.5%)	1 (12.5%)	
Veau 2	0	0	
Veau 3	12 (57.1%)	9 (42.9%)	
Veau 4	6 (54.5%)	5 (45.5%)	
Operation surgeon			0.068
Surgeon 1	9 (52.9%)	8 (47.1%)	
Surgeon 2	2 (50%)	2 (50%)	
Surgeon 3	0 (%)	2 (100%)	
Surgeon 4	14 (82.4%)	3 (17.6%)	

Oronasal fistula

The number of patients who developed oronasal fistula was 10 out of 40. The oronasal fistula rate was 25% (95% CI = 11.99-45.98). Oronasal fistula formation according to cleft types, age at palatoplasty and operating surgeons were presented in Table 4. Veau 3 was the major cleft type found in 7 (77.8%) patients and Veau 4 found in 2 (22.2%) patients (Fig. 5). The oronasal fistula occurred in 38% (8/21) of the patients with Veau 3 and in 18% (2/11) of the patients with Veau 4 (p<0.088) (Table 5).

The authors found that all patients with oronasal fistula underwent primary palatoplasty before 18 months of age (Fig. 6). There was no statistical difference among age of palatolplasty when cut-point was 12 months (p = 0.259). Four patients (40%) with oronasal fistula received palatoplasty by surgeon 1, 3 patient (30%) by surgeon 4, 2 patients (20%) by surgeon 2 and one patient (10%) by surgeon 3. There was no statistically significant difference with the operating surgeon on fistula rate (p = 0.473) (Fig. 7). One patient

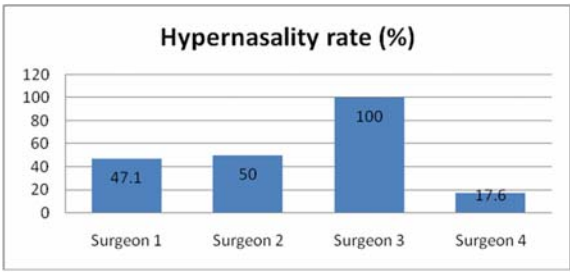

Fig. 3 Hypernasality rate according to operating surgeon.

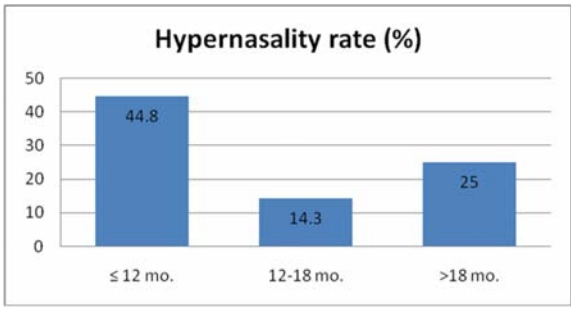

Fig. 4 Hypernasality rate according to age at palatoplasty.

Table 4. Characteristics between patients with and without fistula

Characteristics	Patients without oronasal fistula (n = 30)	Patients with oronasal fistula (n = 10)	p-value
Male: female	18 (60%):12 (40%)	5 (50%):5 (50%)	0.716
Comorbidity	2	1	0.518
Mean age at palatoplasty (month)	15.20 (±12.02)	10.80 (±1.32)	0.259

Table 5. Data analysis grouped by occurrence of oronasal fistula

Characteristics	No oronasal fistula (n = 30)	Oronasal fistula (n = 10)	p-value
Age at palatoplasty			0.259
≤12 months	20 (68.9%)	9 (31.1%)	
12-18 months	6 (85.7%)	1 (14.3%)	
>18 months	4 (100%)	0	
Cleft types			0.088
Veau 1	8 (100%)	0	
Veau 2	0	0	
Veau 3	13 (61.9%)	8 (38.1%)	
Veau 4	9 (81.8%)	2 (18.2%)	
Operation surgeon			0.473
Surgeon 1	13 (76.5%)	4 (23.5%)	
Surgeon 2	2 (50%)	2 (50%)	
Surgeon 3	1 (50%)	1 (50%)	
Surgeon 4	14 (82.4%)	3 (17.6%)	

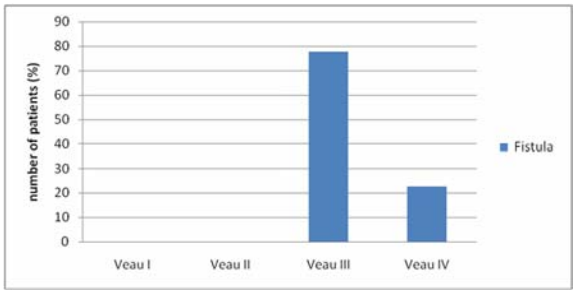

Fig. 5 Occurrence of fistula according to cleft types.

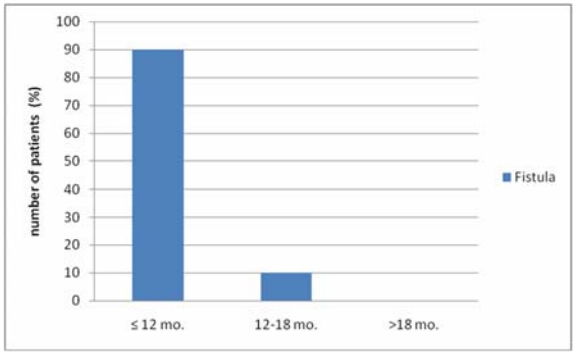

Fig. 6 Occurrence of fistula according to age at palatoplasty.

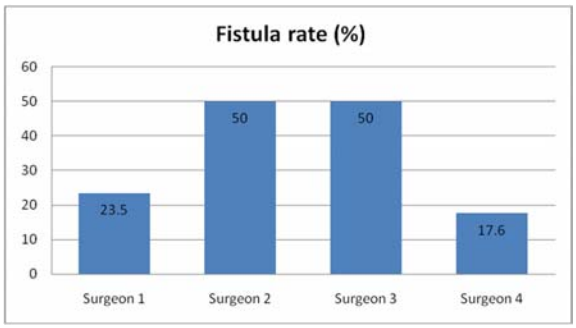

Fig. 7 Occurrence of fistula according to operating surgeon.

immediately had postoperative bleeding and hematoma. The presenting symptoms were regurgitation (88.9%), hypernasality (11.1%) and asymptomatic (11.1%). The fistulas were mostly located in hard palates (88.9%), followed by the junction of hard and soft palate (11.1%). The size of fistula was recorded in nine patients with a variable size between 2 mm and 10 mm. Nine of these 10 patients (90%) required a second operation to achieve fistula closure. Irrespective of the oronasal fistula closure technique, two-flap palatoplasty was the most chosen technique, being used in five patients (62.5%). Two were closed with local flaps (25%) and one with a facial artery musculomucosal (FAMM) flap (12.5%). Clinical characteristic of ten patients who developed oronasal fistula is shown in Table 6.

Recurrence of fistulas occurred in two patients (22.2%). One patient (No. 3) required a second surgery due to symptoms of regurgitation. Two-flap palatoplasty was the chosen technique to close the fistula, and the recurrence fistula was not found at the time of the 20-month follow-up. Another recurrent fistula patient (No. 9) had a fistula in the hard palate; he had symptoms of regurgitation and mild hypernasality and was scheduled for a third surgery after scar tissue maturity. Two patients (case 7, 9) had residual oronasal at the time at study (Fig. 8).

Discussion

The goals of palatoplasty are to achieve complete and intact closure of the palate, avoiding palatal fistula, restoration of the velopharyngeal sphincter for the development of normal speech, optimization of maxillary growth and minimizing hearing loss and middle ear complications[5,6]. Hypernasality was generally considered to be the primary feature associated with velopharyngeal insufficiency[15]. The reliable testing for velopharyngeal insufficiency can be performed somewhere between 3 and 5 years of age[16]. The speech outcomes of the present study will be based on the perceptual analyses of hypernasality. It was feasible to compare our results with other studies due to rates of velopharyngeal insufficiency variances in the reported literature which depended significantly on an exact definition.

The published literature with a detailed evaluation of speech and age at speech evaluation revealed a 29-37% hypernasality rate.

Sell (1999): Great Ormond Street Hospital for Children, London reported speech outcomes in cleft palate children throughout Clinical Standards Advisory Group's investigation of cleft care in the United Kingdom with cross-sectional analysis of 238 5-year-olds unilateral cleft lip and palate patients. The type of palatoplasty was not reported. Cleft Audit Protocol for Speech on a five-point scale was utilized and the hypernasality rate was 29%. Twenty-seven patients previously had secondary velopharyngeal surgery and normal resonance was obtained at the time of data collection. They found that speech outcome was underreported[8]. Mary (2005); University of Wyoming, USA, retrospectively studied 212 preschool patients in all cleft types, but the type of palatoplasty was not

Table 6. Characteristic of 10 oronasal fistula patients

Patient No.	Gender	Cleft type (Veau)	Age at palatoplasty (months)	Type of palatoplasty	Surgeon	Location	Size (mm)	Symptom	ONF closure techinique	Age at ONF closure	Recurrence ONF
1	M	3	10	Two-flap	4	Junction of soft and hard palate	8	R	Two-flap palatoplasty	1 year 9 month	No
2	F	3	9	Two-flap	2	Hard palate	5	R	Local palatal flap	2 year 1 month	No
3	F	4	11	Two-flap	3	Hard palate	10	R	Local palatal flap	2 year 2 month	Yes^{++}
4	F	4	11	Two-flap	4	Hard palate	7	R, H	Two-flap palatoplasty	4 year 10 month	No
5	M	3	11	Two-flap	4	Hard palate	3	R	Two-flap palatoplasty	2 year 7 month	No
6	M	3	12	Two-flap	2	Hard palate	5	R	Two-flap palatoplasty	2 year 6 month	No
7	M	3	9	Two-flap	1	Hard palate	2	Asymptomatic	No*		
8	F	3	10	Two-flap	1	Hard palate	10	R	FAMM	2 year 10 month	No
9	M	3	12	Two-flap	1	Hard palate	10	R	Two-flap palatoplasty	4 year 6 month	Yes*
10	F	3	13	Two-flap	4	Hard palate	N/A	R, H	Two-flap palatoplasty	4 year 2 month	No

M = male; F = female; R = regurgitation; H = hypernasality. * present of residual ONF at time of study; $^{++}$ no residual ONF due to 3rd palatal repair

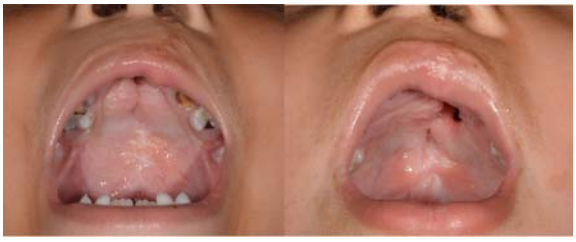

Fig. 8 Two patients who had residual oronasal fistula at time of evaluation. (left) patient No. 7 (right) patient No. 9.

reported. Mean age at evaluation was 3 years 6 months. Mean age of palatoplasty was 16 months. Twenty-five percent of patients received secondary velopharyngeal insufficiency surgery. Judgments of hypernasality were made using a seven-point rating scale. The hypernasality rate was 37%. This study focused on the speech problems of the young preschool child, so a high hypernasality rate was present[17]. Yun Shan Phua (2008); Middlemore Hospital, New Zealand, Cleft Audit Protocol for Speech reported hypernasality in 211 patients of all cleft types including syndromic cleft. Mean age at evaluation was 4 years 10 months. Types of palatoplasty were Veau, von Langenbeck and Furlow Z-plasty. Mean age of palatoplasty was 1 year 1 month. Rate of hypernaslity was 31.8%. Secondary surgery for velopharyngeal insufficiency was required in 13.3%[18].

Regarding surgical technique, there are a few reports of speech outcome in Two-flap palatoplasty. Bardach (1995); University of Iowa who first described this technique reported 75-80% of patients had normal speech production. Oronasal fistulas were found in 5.2% of patients with all clefts types[19]. Sullivan (2006); Children's Hospital and Harvard Medical School, study of 449 patients with Two-flap palatoplasty. Mean age at palate repair was 11.6 months. Velopharyngeal sufficiency was found in 85.1%. Fistula rate was 2.9%[20]. Salyer (2006): Singapore General Hospital, was a study of 150 non-syndromic patients. Palatoplasty was performed before 12 months. 91.14% of patients had normal to mildly impaired resonance. Velopharyngeal insufficiency was 8.92%[21]. Intania (2012); Cipto Mangunkusumo Hospital, Indonesia, a retrospective study of 22 non-syndromic children with complete unilateral cleft palate who performed two-flap palatoplasty. Age at palatoplasty was around 2 years old and utilized the Murthy rating criteria for speech parameters. Overall hypernasalty rate was 36.5%[22].

Our 37.5% hypernasality rate was comparable to the results of other available research reports. However, it was not possible to be statistically compared because of research variables, design, measurement of outcome and protocol practices.

David (2000) mentioned that skill and experience of the surgeon could affect surgical outcomes and the timing of primary palatoplasy may be an important variable. The theory that the palate should be repaired before 2 years of age with an 18-month ceiling has gained wide acceptance[7]. In the present study, the operating surgeons and age at palatoplasty did not affect the development of hypernasality, but it should be noted that most of the patients underwent palate repair before 18 months of age. The extent of the cleft defect is an important determinant of outcome. Other authors have suggested that Veau type 1 clefts are more likely to result in a favorable functional outcome after repair[23,24]. In the present study, hypernasality was higher in patients with a Veau 3 and 4 cleft.

The fistula rate variance depended on the definition of reported fistula. Intentionally unrepaired nasoalveolar and/or anterior hard palate fistulas are not included in the condition[13]. In the present study, lingual-alveolar and labial-alveolar fistulas were excluded.

Yun Shan Phua summarizes the reported fistula rates from studies over the last 30 years (1979 to 2006). The fistula rate was 3-45%[18]. The authors also reviewed the report of fistula rates after 2006 and adapted from article by Yun Shan Phua in Table 7.

The overall fistula rate was 25% in the present study. It will be difficult to compare the results because of variables in surgical techniques and treatment protocols. The authors found four pieces of literature which reported the fistula rate in Two-flap palatoplasty. Bradon (2001); University of Texas Medical Branch and Southern Illinois University, reported a 3.4% fistula rate in 119 consecutive patients who underwent Two-flap palatoplasty by a single surgeon. All cleft types were included. Age at palatoplasty was about 9 months[25]. Murthy (2009): George Washington University Medical Center, reported 2.4% fistula rate in 332 consecutive non-syndromic cleft patients who underwent 2-flap palatoplasty at a mean age 10.8 months[26]. Salyer (2006) reported 10% fistula rate in 382 non-syndromic patients who underwent two-flap palatoplasties before 12 months[21].

The recently published literature by Michael from the University of Pittsburgh shows a meta-analysis rate of oronasal fistula formation following primary cleft

Table 7. Fistula rates reported in previous studies

Literatures	Cleft types included	Fistula rate (%)	Study size
Abyholm et al (1979)	All cleft types	18	1,108
Bardach et al (1984a)	All cleft types	17	45
Bardach et al (1984b)	All cleft types	14	43
Schultz (1986)	All cleft types	22	267
Amaratunga (1988)	All cleft types	21	236
Coghlan et al (1989)	All cleft types	15	20
Rohrich and Byrd (1990)	All cleft types	45	44
Cohen et al (1991)	All cleft types	23	120
Morris et al (1993)	All cleft types	18	40
Canady et al (1994)	All cleft types	14	329
Lehman (1995)	All cleft types	12	34
CSAG-5-y cohort (1998)	Unilateral cleft lip/palate	38	239
CSAG-12-y cohort (1998)	Unilateral cleft lip/palate	10	218
Onizuka et al (1996)	All cleft types	14	28
Emory et al (1997)	All cleft types	11.5	113
Muzaffar et al (2001)	All cleft types	8.7	93
Wilhelmi et al (2001)	All cleft types	3.4	119
Chait et al (2002)	Unilateral cleft lip/palate	3	35
Bekerecioglu et al (2005)	All cleft types	5	39
Inman et al (2005)	All cleft types	4.7	148
Helling et al (2006)	All cleft types	3	31
Middlemore data (2006)	All cleft types	13	211
Agrawal and Panda (2006)	All cleft types	2.95	678
Salyer et al (2006)	All cleft types	10	382

palate repair, and associated risk factors. The papers published between 2000 and 2012 clearly described oronasal fistula and did not include lingual-alveolar and labial-alveolar fistula. The fistula rate was 4.9; data were collected from 11 studies, comprising 2505 children[27].

Multiple factors influenced fistula rates, including severity and cleft size, type of cleft, palate repair technique, timing of repair and the experience level of the operating surgeon[5,13]. The recent study found a significant relationship between Veau classification and the occurrence of fistula, mostly in Veau 4 cleft[27].

The present study reported a 25% fistula rate. The fistula rate was comparable with results from other studies. The authors found all fistulas occur in Veau 3 and Veau 4 patient and were not found in Veau 1. According to cleft type, many of our patients (80%) had a severe cleft extension (Veau 3, 4), which may have led to a higher fistula rate. Although cleft widths were not documented in the present study, the severity of cleft type represents a significant factor, as a greater width of cleft has shown a statistically significant increase in fistula rate[28]. The authors felt the patients in our area had a wide cleft which made it difficult to achieve closure that was tension-free, although grotto-flap technique has been demonstrated to be a safe and reliable procedure[19].

According to the timing of surgery and surgeon experience, the younger or more inexperienced surgeons tend to have higher fistula rates, and more occasional cleft surgeons will have a higher incidence of fistulas and the strongest predictor of the occurrence of a cleft palate fistula, was the surgeon performing the procedure[5,7,29]. In our series, there was no significant difference in fistula rates among surgeons. The timing of the repair appears to lead to mixed results[5]. All of our patients who had fistula underwent primary palatoplasty before 12 months. It should be noted that the most of our patient were operated on before 18 months and all patients who were operated on older than 18 months had Veau 1 cleft, in which fistulas did not occur in this cleft type in our study. Most cleft practitioners would argue in favor of earlier repair of the palate for better speech outcomes[6,30].

Management of fistulas, especially oronasal

fistulas, is important to undertake as soon as possible if it has been determined that they have detrimental effects on speech[31]. There were nine patients who underwent fistula closure; procedures were mostly performed before 3 years of age. The recurrence rate was 22%. The data on speech outcome before fistula closure were not available but the authors found the patients who had previous fistula closure with no residual fistula exhibited normal resonance 62.5% (5 out of 8 patients). In two patients who had residual oronasal fistula at the time of speech evaluation, we found all of them had hypernasal speech. It is recognized that oronasal fistula also contributed to hypernasality and these patients required a more detailed investigation[15].

The data obtained in the present study reflects the outcomes associated with our surgical protocol in the period of 2007-2008. There is no doubt that improving surgical technique has and will lead to better patient outcomes. The variability of factors directly and indirectly affecting speech outcomes in cleft palate patients, surgical parameters, physiology or environment and speech parameters such as the history and amount of speech therapeutic intervention, hearing status, family and quality of service should be mentioned[31]. The high rate of children with hypernasality is a major problem during preschool age. The therapeutic intervention should be continued in these patients. A further study of possible factors that influence the speech outcome was suggested, in order to provide better management protocols.

Conclusion

The rate of hypernasality and oronasal fistula formation was comparable with results from other standard cleft centers in cleft palate patients who underwent primary palatoplasty during our previous management protocol. The data should be further analyzed for possible affecting factors to provide better outcomes in the future.

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Potential conflicts of interest

None.

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ผลลัพธ์ทางคลินิกของการผ่าตัดเสริมสร้างเพดานโหว่แบบปฐมภูมิในเด็กเพดานโหว่ช่วงก่อนวัยเรียนในโรงพยาบาลศรีนครินทร์เมื่อเปรียบเทียบกับศูนย์การดูแลผู้ป่วยปากแหว่งเพดานโหว่อื่นที่เป็นมาตรฐาน

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วัตถุประสงค์: ศึกษาผลลัพธ์ทางคลินิกในด้านอัตราการเกิดภาวะเสียงขึ้นจมูกมากเกินไป (hypernasality) และอัตราการเกิดรูรั่วเพดานปาก (oronasal fistula) ในผู้ป่วยเพดานโหว่ที่ได้รับการผ่าตัดเสริมสร้างเพดานโหว่แบบปฐมภูมิที่ศูนย์การดูแลผู้ป่วยปากแหว่งเพดานโหว่ โรงพยาบาลศรีนครินทร์

วัสดุและวิธีการ: เป็นการศึกษาแบบตัดขวางในผู้ป่วยเพดานโหว่ที่มีหรือไม่มีปากแหว่งร่วมด้วยชนิดไม่มีกลุ่มโรคร่วมด้วย ที่เกิดระหว่างเดือนกุมภาพันธ์ พ.ศ. 2550 ถึง เดือนธันวาคม พ.ศ. 2551 ที่ได้รับการผ่าตัดเสริมสร้างเพดานโหว่แบบปฐมภูมิที่โรงพยาบาลศรีนครินทร์จำนวน 40 ราย โดยมีการศึกษาทั้งด้านข้อมูลทั่วไปรวมทั้งชนิดของเพดานโหว่ อายุเมื่อผ่าตัด แพทย์ผู้ผ่าตัด และเทคนิคการผ่าตัด

ผลการศึกษา: ผู้ป่วยจำนวน 40 ราย ประกอบด้วยเพศชาย 23 ราย เพศหญิง 17 ราย ผู้ป่วยจำนวน 3 รายมีโรคประจำตัวร่วมด้วยได้แก่ amniotic band syndrome และ club feet จำนวน 1 รายและภาวะ G-6-PD deficiency จำนวน 2 ราย อายุเฉลี่ยขณะทำการศึกษาเท่ากับ 5.7 ปี (5-6.9 ปี) อายุเฉลี่ยขณะได้รับการผ่าตัดเสริมสร้างเพดานโหว่เท่ากับ 14.1 เดือน (9-64 เดือน) มีศัลยแพทย์จำนวน 4 ราย ได้แก่ศัลยแพทย์ตกแต่งจำนวน 3 รายและแพทย์ประจำบ้านภายใต้การดูแลของศัลยแพทย์ตกแต่ง ชนิดของเพดานโหว่ประกอบด้วยเพดานโหว่ชนิด Veau 3 คิดเป็นร้อยละ 52.5 ชนิด Veau 4 คิดเป็นร้อยละ 27.5 และชนิด Veau 1 คิดเป็นร้อยละ 20 โดยทุกรายใช้เทคนิคการผ่าตัดเสริมสร้างเพดานโหว่แบบการย้ายแผ่นเนื้อชนิด Two-flap (Two-flap palatoplasty) ผู้ป่วยจำนวน 15 รายมีภาวะเสียงขึ้นจมูกมากเกินไป คิดเป็นอัตราการเกิดภาวะเสียงขึ้นจมูกมากเกินไปร้อยละ 37.5 โดยมีอัตราเสียงขึ้นจมูกมากเกินไประดับเล็กน้อย (mild hypernasality) เท่ากับร้อยละ 25 อัตราเสียงขึ้นจมูกมากเกินไประดับปานกลาง (moderate hypernasality) เท่ากับร้อยละ 12.5 โดยไม่มีรายใดมีภาวะเสียงจมูกมากเกินไประดับรุนแรงผู้ป่วยจำนวน 10 ราย เกิดรูรั่วเพดานปากหลังจากผ่าตัดเสริมสร้างเพดานโหว่คิดเป็นร้อยละ 25 ผู้ป่วยจำนวน 9 รายได้รับการผ่าตัดปิดรูรั่วเพดานปาก (oronasal fistula closure) คิดเป็นร้อยละ 90 มีผู้ป่วยจำนวน 2 ราย คิดเป็นร้อยละ 5 ที่มีรูรั่วเพดานปากหลงเหลืออยู่ขณะที่ทำการศึกษาไม่มีความแตกต่างกันอย่างมีนัยสำคัญทางสถิติระหว่างชนิดเพดานโหว่ อายุเมื่อผ่าตัดและศัลยแพทย์ผู้ผ่าตัดต่อการเกิดภาวะเสียงขึ้นจมูกมากเกินไปและการเกิดรูรั่วเพดานปาก

สรุป: อัตราการเกิดภาวะเสียงขึ้นจมูกมากเกินไปและการเกิดรูรั่วเพดานปากในเด็กเพดานโหว่ที่ได้รับการผ่าตัดเสริมสร้างเพดานโหว่แบบปฐมภูมิในแนวปฏิบัติที่ผ่านมาในโรงพยาบาลศรีนครินทร์ใกล้เคียงกับสถาบันการดูแลผู้ป่วยปากแหว่งเพดานโหว่ที่เป็นมาตรฐานอื่น ควรที่จะมีการศึกษาถึงปัจจัยที่อาจส่งผลต่อผลลัพธ์ด้านการพูดและการเกิดรูรั่วเพดานปากในอนาคตเพื่อเป็นการพัฒนาคุณภาพของการดูแลผู้ป่วยต่อไป