Speech, Language, Voice, Resonance and Hearing Disorders in Patients with Cleft Lip and Palate

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Objective: *To estimate the prevalence of speech, language, and hearing disorders in patients with cleft palate with or without cleft lip.*
Material and Method: *All data were retrieved from 384 medical records, transferred to case record forms and analyzed.*
Results: *The oronasal fistula rate was 15.25% (95% confidence interval: CI 11.49-19.02). The overall rates of delayed language development, articulation disorders, resonance disorders, voice disorders, and hearing disorders were 16.33% (95% CI = 12.65-20.69), 88.56% (84.47-92.65), 43.26% (95% CI = 36.58-49.93), 19.13% (95% CI = 14.26-24.82), and 79.49% (95% CI = 74.28-84.70), respectively.*
Conclusion: *For speech and hearing, rates of abnormality were very high compared with those reported in the previous studies. Treatment protocols should receive more attention and intervention.*

Keywords: *Speech disorder, Language disorder, Hearing disorder, Cleft palate, Cleft lip*

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Most children with cleft palate suffer from several speech, language and hearing problems, including speech, language, articulation, and hearing difficulty, dysphonia, and in particular, velo-pharyngeal insufficiency. The incidence of these conditions varies widely and is related to factors such as cleft type and palate treatment, including the timing of cleft repair, age at surgery, surgical procedure, hearing detection and intervention and speech and language intervention.

The majority of children with cleft palate did not exhibit compensatory misarticulation or velo-pharyngeal insufficiency after they had had a two-step closing procedure at 15-34 months of age and speech assessments were made at 2-3 years of age[1]. Children with late cleft palate closure (12-27 months) showed compensatory articulation more frequently than those with early cleft palate closure (5-12 months) (80-90% vs. 5%)[2]. Overall hypernasality and misarticulation have higher rates in children with clefts including the soft palate and late repair (22 months) compared with those who have undergone early repair (13 months)[3]. There are no differences in speech inventory (consonants) except nasal assimilation and backing articulation between children with clefts and non-cleft children. This might suggest that severe phonological problems are associated with late palatal closure, i.e. closure after the onset of meaningful speech[4]. For primary veloplasty at 8 months and closure of hard palate at 8 years, most of the children with clefts demonstrated 1) hoarseness of the voice (43%) due to hyperfunctional compensation, 2) moderate to severe hypernasality (6%), 3) compensatory articulation disorders (23%)[5]. For voice disorders, the rate of hoarseness varies from 0.6-47% in the population of cleft palate children[6-10]. It is higher, 41-50%, in children with clefts and velopharyngeal insufficiency[11,12]. Hearing status is an important variable that influences speech and language problems in children with clefts. The incidence of middle ear pathology may be as high as 97%[13] and Eustachian tube function does not improve significantly after veloplasty until adulthood[13,14]. Fluctuating mild to moderate conductive hearing loss in early childhood can result in impaired speech, language and even cognitive development[15-18].

Velopharyngeal insufficiency affects articulation defects, resonance disorders and dysphonia which results in hypernasality, compensatory articulation disorders, hyperplasia of vocal folds or vocal nodules and hyperemia[19,20] due to abnormal laryngeal valving as a compensatory mechanism for

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velopharyngeal competency[12,21]. The vocal folds are inappropriately adducted in order to provide a constriction inferiorly to the inadequately functioning velopharyngeal closure. Altering velopharyngeal competency often improve resonance, articulation and voice symptoms[22].

In summary, surgical procedures for closure of palate clefts, the age at the time of palatoplasty and treatment of hearing and speech and language abnormalities are important factors that influence the outcome in the speech, language, and hearing domains. Speech defects require early surgery for developing normal articulation, however, maxillofacial surgery would likely be undertaken after maxillofacial the development period finishes and will not disturb or interrupt maxillofacial development. Therefore, compomization of 2 field rationale, labioplasty surgery needs to be done at 3 months and palatoplasty around 1 year. The treatment protocol used by the interdisciplinary team in the Cleft Lip/Palate and Craniofacial Abnormalities Center of Khon Kaen University is concerned with the balance of these outcomes. The scheduling of labioplasty is at 3 months of age and palatoplasty and veloplasty at around 12 months of age, which aims to improve speech, language, and hearing outcomes.

The purpose of this study was to estimate retrospectively the prevalence of speech, language, resonance, voice and hearing disorders in patients with cleft palate with or without lip (CP $\pm$ L).

Material and Method

Study design

This is a descriptive study with retrospective data collection. According to the Helsinki Declaration, Khon Kaen University Ethics Committee for Human Research reviewed and approved (July 21, 2011) the research protocols (HE541129).

Setting

Otorhinolaryngology, Speech and Audiology Clinics, Srinagarind Hospital, Faculty of Medicine, Khon Kaen University, Khon Kaen, Thailand.

Participants

Participants were all patients with CP $\pm$ L who registered for the project "Smart Smile and Good Speech", a celebration of the 50th birthday of Her Royal Princess Sirinthorn, from June 2007-September 2010 at Srinagarind Hospital, Faculty of Medicine, Khon Kaen University, Thailand.

Inclusion criteria

Patients with CP $\pm$ L attending Srinagarind Hospital under the project "Smart Smile and Good Speech" from June 2007-September 2010 and available medical records were included in the present study.

Exclusion criteria

Patients with cleft lip and palate associated with syndromic or any underlying conditions that might affect speech, language, ear, nose, throat, and hearing problems in cleft (e.g., global delay development, facial clefts, velo-cardio facial syndrome, etc.) were excluded.

All data were retrieved from medical records to case record forms and analyzed.

Outcomes

The main outcomes of the present study include the proportions of patients with language delay, articulation disorders, resonance disorders, and hearing disorders. Binary rating was used in which 0 = within normal limit/none (no abnormality) and 1 = present (any abnormality). Data were considered as follows:

Language delay

The child's language skill is scored based on 2 tests:

1) Thai Early Language Mile Stone: TELM[23] assessed language skill for young children aged 0-3 years.

2) Speech and Language Screening[24] assessed language skill for young children aged more than 3 years.

Language skills were scored as 'pass' (0) when the child passed all items of the test and as 'fail' (1) when the child did not pass any item of the test.

Articulation disorders

Articulation was scored as 'pass' or 'fail' (fail = 1, pass = 0) based on the Thai articulation development[25] and articulation screening test which was established by the principle investigator. Articulation was scored as 'pass' (0) for correct production and as 'fail' (fail = 1) for incorrect production or articulation defects (functional or compensatory or both articulation disorders).

Resonance disorders

Resonance assessment was based on the evaluation of a speech sample which was composed of nonsense syllables, serial speech (counting 1-20 and 40-50), 3 simple phrases and sentences with high oral

pressure consonants, as well as 3 nasal sentences. It was evaluated by the principle investigator, who has experience in cleft palate management for more than 20 years, as 'normal' (0) or 'resonance disorder' (1): hypernasality (mild, moderate, and severe) and or hyponasality.

Voice disorders

The rating of voice disorders was based on a whole speech sample that is elicited by perceptual assessment. Six parameters (G: Grade e.g., overall impression, I: Instability e.g., fluctuation of voice, R: Roughness e.g., hoarseness, B: Breathiness e.g., breathy voice, A: Asthenia e.g., a weak voice or speaking with minimal air volume, S: Strain e.g., forced or stressed voice) were used for voice evaluation for counting. They are known as GIRBAS and this is a popular and reliable perceptual scale[26,27]. Each parameter was scored on a scale of 0-3 (0 was considered normal; 1 = slight disturbance; 2 = moderate disturbance; 3 = severe disturbance)[26-28]. Voice was evaluated as normal or no voice disorder (0) and voice disorder when GIRBAS score $\geq$1 (1).

Ear, nose, and throat examinations

The results of ear examinations were retrieved from the information in the medical records. The information included otoscopic findings of the ear drum and the results of hearing tests. Otoscopic findings were classified as 'normal', 'retraction' or 'perforation'.

Hearing disorders

Pure tone audiometry and behavioral audiometry including distraction testing and visual reinforcement audiometry (for young children who could not be assessed by standard audiometry) were exacted from medical records.

Hearing loss was categorized according to standard definitions[29,30]. These were as follows: normal hearing ($\leq$25 dB), mild hearing loss (26-40 dB), moderate hearing loss (41-55 dB), moderately severe hearing loss (56-70 dB), severe hearing loss (71-90 dB) and profound hearing loss (>90 dB). Tympanograms were described as Type A: normal middle ear, Type B tympanogram: flat with no discernable peak, and Type C tympanogram: a peak at negative pressures and occurs in the presence of negative middle ear pressure.

Statistical analysis

Descriptive analyses were used for presentation of the prevalence of language delay development, articulation disorders, resonance disorders, voice disorders and hearing disorders as percentages.

Results

There were 384 patients with CP $\pm$ L who were included in the present study. Thirty children (8.47%) with cleft palate associated with syndromes were excluded, leaving 354 patients with CP $\pm$ L for analysis. Of these patients, the male: female ratio was approximately 1.08: 1, the oronasal fistula rate was 15.25% (95% confidence interval: CI 11.49-19.02). The characteristics of the participants are listed in Table 1.

Every child were evaluated for speech and language disorders, however, the overall rate of speech and language delay was derived from 349 patients because 5 patients' medical records were not available. The prevalence of delayed speech and language development was 16.33% (57 of 348, 95% confidence interval: CI = 12.65-20.69). For speech disorders in patients with CP $\pm$ L, there were available data from

Table 1. Demographic characteristics of patients with cleft palate with or without lip

Characteristics	Number	Percentage
Gender		
Female	170	48.02
Male	184	51.98
Total	354	100.00
Age (years)		
0-2	131	37.01
2-4	103	29.10
4-7	45	12.71
7-15	50	14.12
>15	25	7.06
Total	354	100.00
Cleft type		
Left cleft lip	0	0.00
Right cleft lip	2	0.57
Bilateral cleft lips	1	0.28
Cleft palate	73	20.62
Lt. cleft lip and palate	119	33.62
Rt. cleft lip and palate	57	16.10
Bilateral cleft lip and palate	97	27.40
Submucous deft	5	1.41
Total	354	100.00
Complication or comorbidity		
No	298	84.18
Yes (ONF)	54	15.25
N/A	2	0.57
Total	354	100.00

pre-school and older children's medical records that had potential for assessment, 236 cases for articulation, 215 cases for resonance, 230 for voice, and 227 for intelligibility. Estimations of the prevalence of speech disorders are presented in Table 2.

The available data from ENT examinations are summarized for 218 of 274 ears. Most of them had normal ear canals and ear drums. The results of these assessments of ear canals and ear drums are displayed in Table 3.

Hearing problems were classified according to the type and degree of hearing loss from the available data (234 patients). Of these, 186 patients had hearing loss (356 ears). Tympanometry data from 461 ears were available for analysis. The types and degrees of hearing loss are displayed in Table 4.

Discussion

The overall rate of true oronasal fistula, the most common complication of cleft repair, was 15.25% which is in the range reported in previous studies (12.8-21%)[31,32].

Our findings confirm that patients with cleft palate are at increased risk for communication disorders due to compensatory hyperfunction secondary to VPI[33,34]. The prevalence of delayed speech and language development was 16.33% which is less than that reported in previous studies (13 vs. 92 %)[16,35-38]. A systematic review revealed that the overall prevalence of primary speech and language delay among children aged 2-6 years old was 6.0% (1.4-19%)[39]. The prevalence reported by speech clinics across the country in Thailand for speech and language delay ranges from 23.4-46.8%[40-43]. The rate of speech and language delay in the present study was in the range of the general population's prevalence; however, it was quite low compared to the prevalence in the cleft palate populations in previous studies. This suggests that early diagnosis and early intervention programs in our protocol might be effective for prevention of delayed language development for patients with CP $\pm$ L. Our data supported the view that most children with CP $\pm$ L have the potential to develop normal language if they receive early intervention, which agrees with previous

Table 2. Overall prevalence of speech and language disorders

Disorders	Number	Percentage	95% confident interval
Articulation			
Normal	27	11.44	7.35-15.53
Articulation disorders	209	88.56	84.47-92.65
Functional articulation disorders	55	23.31	17.87-28.74
Compensatory articulation disorders	26	11.02	6.99-15.04
Functional and compensatory articulation disorders	126	53.39	46.98-59.80
Organic articulation disorders	2	0.85	-0.33-2.03
Total	236	100.00	
Resonance			
Normal	122	56.74	50.07-63.42
Resonance disorders	93	43.26	36.58-49.93
Hyponasality	6	2.79	0.57-5.01
Hypernasality			
Mild	42	19.53	14.19-24.88
Moderate	40	18.60	13.36-23.85
Severe	5	2.33	0.29-4.36
Total	215	100.00	
Voice			
Normal	186	80.87	75.18-85.74
Abnormal	44	19.13	14.26-24.82
Total	230	100.00	
Intelligibility			
Intelligibility	226	99.56	97.57-99.99
Unintelligibility	1	0.44	0.01-2.43
Total	227	100.00	

Table 3. Ear examination

Ear examination	Number	Percentage	95% confident interval
Ear canal			
Left ear			
Normal	239	87.55	83.03-91.22
Abnormal	34	12.45	8.78-16.97
Impact cerumen	29	10.62	6.94-14.30
Stenosis	4	1.46	0.03-2.90
Atresia	1	0.37	-0.35-1.09
Total	273	100.00	
Right ear			
Normal	244	89.05	84.74-92.49
Abnormal	30	10.95	7.51-15.26
Impact cerumen	24	8.77	5.39-12.13
Stenosis	5	1.82	0.23-3.42
Atresia	1	0.36	-0.35-1.08
Total	274	100.00	
Ear drum			
Left ear			
Normal	181	83.03	77.37-87.76
Abnormal	37	16.97	12.24-22.63
Retraction	33	15.14	10.34-19.93
Central perforation	4	1.83	0.04-3.63
Total	218	100.00	
Right ear			
Normal	185	83.72	78.17-88.32
Abnormal	36	16.28	11.68-21.83
Retraction	34	15.38	10.59-20.18
Central perforation	2	0.90	-0.35-2.16
Total	221	100.00	

findings[35].

For speech disorders that effect intelligibility, the previously reported prevalence is 44-63%[36,44,45]. However, the prevalence of articulation disorders in this study was higher at 88.56% (Table 2). The rate of hypernasality (43.30%) (Table 2), indicating velopharyngeal insufficiency (VPI), was slightly higher than in previous review articles (range 20-40%)[32,46-51]. This might be due to the lack of an early speech correction programs in our center; the majority of children (62.99%, Table 1) were enrolled for their 1st visit for speech and language treatment at age ≥2 years and 33.89% of them registered for their 1st assessment at age ≥4 years. This might be too late for early prevention of compensatory articulation disorders and speech disorders because the standard management of articulation disorders in cleft palate should start during the prelingual stage[52]. Our treatment protocols, including surgery and early speech and language intervention program, require more attention.

The rate of voice abnormality was 18.59% which is similar to that reported in previous studies that found a prevalence of 5.5-20.8%[7,9,53]. This study's rate was approximately the same as that reported in a recent study that found a hoarseness rate was of 18.4-20.8%[7], but lower than that found in another recent study[54]. These results support the theory that patients with cleft palate are at increased risk for voice disturbances due to laryngeal hyperfunction as a compensatory articulation mechanism for VPI[2,12,21,22]. Dysphonia is usually caused by the strain from the explosive effort required when trying to build up the pressure necessary for speech[33]. It is an obligatory speech disorder that requires the physical management of VPI.

Middle ear disease is common in children with cleft palate and, unlike the case of children without clefts, and has a prolonged recovery period and a

Table 4. Types and degrees of hearing loss

Hearing	Number	Percentage	95% confidence interval
Type of hearing (patients)			
Normal	48	20.51	15.30-25.72
Hearing loss	186	79.49	74.28-84.70
Conductive hearing loss both ears	165	70.51	64.63-76..40
Unilateral conductive hearing loss	16	6.84	3.58-10.10
Mixed hearing loss	4	1.71	0.04-3.38
Sensorineural hearing loss	1	1.27	-0.41-1.27
Total	234	100.00	
Hearing loss level (ears)			
Mild	130	36.12	31.49-41.54
Moderate	181	50.84	45.62-56.06
Moderately severe	39	10.96	7.69-14.22
Severe	4	1.12	0.02-2.22
Profound	2	0.56	-0.22-1.34
Total	356	100.00	
Tympanogram (ears)			
Type A	98	21.26	17.50-25.01
Type B	335	72.67	68.58-76.75
Type C	28	6.07	3.89-8.26
Total	461	100.00	

substantial incidence of late sequelae. Otitis media with effusion has been demonstrated to be almost universally present in infants with cleft palate[13,55]. Otitis media with effusion and hearing loss are also common findings among older children with cleft palate[56]. The present study revealed that the prevalence of hearing abnormalities was 79.49%. Conductive hearing loss was the most common type in the present study (77.35%) (Table 4), in line with the results of previous studies[37,57,58], but it was very high when compared to one recent study (13.6%)[59]. The rate of mixed hearing loss from this study is similar to the rate reported in a previous study[59] (1.71% vs. 2.2%). With regard to the degree of hearing loss, the data revealed that patients with CP $\pm$ L had mild to moderate losses (86.96%: 26-55 dB) (Table 4). Of these, 36.12% had a mild hearing loss that might improve with age[60] and 63.48% had a moderate level (41^{+} dB) that might not significantly improve with age[60], potentially interfering with language and speech development, as their hearing levels are beyond those required to understand normal conversation (40-60 dB). This will affect children's language development if it persists long enough.

Based on the current theory, the reason for having secretory otitis media in children with CP $\pm$ L is that these abnormalities are associated with chronic Eustachian tube dysfunction because of a failure of the tube opening mechanism. The Eustachian tube does not open during swallowing because of the abnormality of the soft palate or stiffening from its repair and does not function normally. Inadequate Eustachian tube function is associated with the presence of fluid in the middle ear. Sometimes, the tube can open and cause the aspiration of nasopharyngeal secretions, thus maintaining the secretory otitis media in the middle ear. If there is fluid in the middle ear, perforation of the tympanic membrane, scarring of the tympanic membrane, or erosion of the ossicular chain may occur. In these circumstances the tympanogram, which is a way of assessing the function of the middle ear will be type B or C in the presence of middle ear effusion or will have a high peak if there is ossicular discontinuity. Although many of the ear drums of children in this study were assessed as normal by the examining doctors, the most common type of tympanogram in this study was type B (72.67%, Table 4) in line with the results of a recent study that found tympanogram type B has the highest prevalence in cleft palate patients with middle ear effusion in both study groups (single and repeated ventilating tube insertions)[61]. This result supports the findings for hearing type and hearing loss levels. Regarding the prevalence of hearing loss type

and the degree of hearing loss and tympanogram types, the rates in the present study were very high. The very high rates of hearing problems may be due to the retrospective nature of the present study and our results were interpreted with available data. A further prospective study should be done and early interventional programs for patients with CP ± L should be critically revised.

Conclusion

The prevalence of speech and language delay was a low overall rate. For speech and hearing disorders they were very high compared to previous studies.

Therefore, our treatment protocols, including physical treatment, early speech therapy program, and hearing conservation should be reviewed and improved.

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Potential conflicts of interest

None.

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ปัญหาภาษาการพูดการได้ยินในผู้ป่วยปากแหว่งเพดานโหว่

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วัตถุประสงค์: เพื่อศึกษาความชุกของความผิดปกติทางการพูด ภาษา การสั่นพ้องของเสียง เสียง และการได้ยินในผู้ป่วยเพดานโหว่ที่มีหรือไม่มีปากแหว่งร่วมด้วย

วัสดุและวิธีการ: ข้อมูลทั้งหมดถูกค้นจากประวัติการรักษาของผู้ป่วยจำนวน 384 ราย ลงในแบบบันทึกรายบุคคลแล้วทำการวิเคราะห์ผล

ผลการศึกษา: ความชุกของรูรั่วระหว่างช่องปากและเพดานพบร้อยละ 15.25 (95% ของช่วงเชื่อมั่น = 11.49-19.02) ส่วน ความชุกของการพัฒนา ภาษาล่าช้า การพูดไม่ชัด การสั่นพ้องผิดปกติ เสียงผิดปกติ และการได้ยินผิดปกติพบได้ร้อยละ 16.33 (95% ของช่วงเชื่อมั่น = 12.65-20.69), ร้อยละ 88.56 (95% ของช่วงเชื่อมั่น = 84.47-92.65), ร้อยละ 43.26 (95% ของช่วงเชื่อมั่น = 36.58-49.93), ร้อยละ 19.13 (95% ของช่วงเชื่อมั่น = 14.26-24.82), และร้อยละ 79.49 (95% ของช่วงเชื่อมั่น = 74.28-84.70) ตามลำดับ

สรุป: พบความผิดปกติของการพูดและการได้ยินข้างสูง แผนการรักษาควรได้รับความสนใจและปรับขบวนการรักษาให้ดีขึ้น