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Original article

Articulation disorders and patterns in children with a cleft

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Background: Compensatory articulation disorders (CAD) are the most common speech defects in patients with a cleft. Early prevention programs are needed to avoid CAD.
Objectives: To examine articulation disorders, patterns, and related speech outcomes in children with a cleft palate with or without lip defects.
Methods: Articulation test record forms and clinical records of 42 children were accessed retrospectively to provide the data of speech outcomes related to cleft palate. Double data entries and incorrect completion type errors were corrected.
Results: Prevalence of articulatory defects was 88% (functional articulation disorders, 12%; compensatory articulation disorders, 10%; functional articulation disorders and CAD, 67%), resonance disorder was 50%, and voice abnormalities was 19%. Abnormal backing of oral consonants, particularly glottal substitution was the most common pattern of CAD (40%), follow by velar substitution (36%), and nasal consonant for oral pressure consonant (21%). There was high incidence of functional articulation disorder in patients with a cleft (76%). Younger children (≤7 years old) had more articulation defects than older children (>7 years old) (mean difference = 3.308, $P = 0.002$, 95% confident interval 1.683–6.971). Levene's test for equal variance found that resonance disorder seems unaffected by the number of articulation errors (mean difference = 0.253, $P = 0.897$, 95% confident interval –3.736–4.241).
Conclusion: CAD, particularly abnormal backing of oral consonants and hypernasality were the most common speech defects in children with cleft. Refinement and revision of timing for referring for early speech intervention should be reconsidered.

Keywords: Cleft palate, compensatory articulation disorder resonance disorder, speech defect, speech outcome

There are two viewpoints for timing of primary surgical closure of the cleft palate. The first viewpoints advocates for late palatal closure because early surgical closure of cleft palates negatively affects maxillary development and had an adverse effect on the aesthetic appearance of the face and occlusion. It also inhibits maxillofacial growth development [1]. Late palatoplasty is considered to produce a better result for speech outcomes [2-4] and less maxillary growth restriction [5-7]. The incidence of compensatory articulation disorders (CAD), hypernasality (moderate to severe), and voice disorders (because of hyperfunctional compensation) were 23%, 6%, and 43%, respectively in children with a cleft palate who received late palatoplasty (veloplasty at 8 months and closure of hard palate at 8 years) [8]. Children with delayed closed clefts had significantly more nasal escape and a higher prevalence of compensatory retracted articulation than children with functionally closed clefts [9].

A second viewpoints supports the early surgery of palatal closure, which is associated with better speech outcomes [9-11]. From this viewpoint, surgery should be performed before the onset of the first word to provide the primary goal of palatoplasty: an intact hard and soft palate to produce a normally functioning velopharyngeal mechanism and speech outcomes [10-12].

In consideration of speech and language development, the first year of life is of major importance for acquiring speech, normal children at 6 months of age begin producing the first frontal consonant; however, those with clefts have a structural defect of the palate that makes this

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production difficult or impossible, and usually compensate by producing glottal or pharyngeal articulations that are typical patterns of speech associated with a cleft. Particularly in relation to speech, signs of velopharyngeal insufficiency (VPI), which is the inability of the soft palate to make contact with the upper throat wall to close off the nose during speech. Speech defects from VPI include hypernasality, nasal air escape, weak intraoral air pressure during oral sound production, and use of atypical places of production known as CAD or cleft characteristic articulation disorders [11, 13]. CAD are generally developed from inadequate oral pressure for oral consonants caused by velopharyngeal insufficiency. Production of sounds further back in the mouth (or throat) are substituted to compensate for the inadequate oral pressure therapy, such as glottal stop for oral plosive, pharyngeal or nasal fricative for oral fricative, and, use of mid dorsum palatal place of production [14]. CAD severely affects intelligibility and usually requires a prolonged period of speech therapy. Thus, normal anatomical conditions in clefts should be reconstructed in infancy to allow for correct kinesthetic patterns before CAD becomes misarticulation in the speech system. Speech and language pathologists have generally recommended that the primary surgery of palatoplasty should be performed at the age of 1 year [15-17].

There have been comparisons between the two viewpoints and they found no differences of speech, or mandibular or maxillary growth between one stage (early primary palatoplasty) and two stages (delayed hard palate closure) of palatoplasty [18, 19]. A current review also revealed that there is no evidence to support which timing of primary palatoplasty gave better maxillary, mandibular growth and speech outcomes. However, well-designed controlled studies and long-term studies are needed [20].

For balance of maxillofacial growth and the development of speech and language abilities, the scheduling for labioplasty is approximately 3 months and palatoplasty and veloplasty is around 12 months. This objective of treatment for children with clefts aims to have the clefts closed soon as reasonable to minimize faulty habits of speech articulation and intraoral, oropharyngeal, laryngeal compensatory movements for improvement the speech outcomes. This protocol showed CAD including lateralization 4%, palatalization 19%, velar 11%, uvular 4%, pharyngeal 23%, glottal 7.4%, active fricative 4%, nasal consonant 7% in 27 children with isolated cleft palate and that lateralization 24%, palatalization 24%, velar 18%, glottal 6%, nasal consonant 24% in 17 children cleft lip and palate, respectively [21]. Recent study revealed overall rates of delayed language development, articulation disorders, resonance disorders, voice disorders, and hearing disorders were 16.33%, 88.56%, 43.26% 19.13%, and 79.49%, respectively [22]. Velar production was the most common pattern, followed by glottal and pharyngeal productions at word level (43.75%) and at sentence level test (37.5% and 18.75%), respectively [23].

The purpose of this study was to examine articulation disorders, patterns and related speech outcomes in children with cleft palate with or without lip involvement (CP ± L) in The Center of Cleft Lip Cleft Palate and Craniofacial Deformities, Khon Kaen University, in association with the Tawanchai Project.

Materials and methods

Study design

A descriptive study with retrospective data collection was conducted. According to the Helsinki Declaration (HE541129), the Ethics Committee of Khon Kaen University reviewed and approved (July 21, 2011) the research protocols.

Participants

Forty-two children with cleft lip and/or palate who registered in the Project "Smart Smile and Good Speech", in honor of the 50 year celebration of Her Royal Highness Princess Sirinthorn and had completed formal articulation test records.

Chart records of formal articulation tests and clinical history were reviewed, data accessed and filled into case record forms. Double data entry errors and incorrect completion type errors of data were corrected.

Outcomes

The main outcomes of this study were number of articulation errors and patterns, children's characteristics and related data including resonance disorders, voice abnormalities, and intelligibility from articulation tests and clinical records. Binary rating was used in which 0 = within normal limit/none (no abnormality) and 1 = present (any abnormality). Data were considered as follows:

Articulation disorders: articulation screening test (composed of 4 sentences of all of the Thai initial and

final consonants), which was performed on the first visit and related speech outcomes including screening of resonance, voice and intelligibility were retrieved from articulation test and medical records. The formal articulation test [24] was performed on the second or later visit. Articulatory production was scored as pass (0) for correct production and as fail (1) for incorrect production or articulation defect based on the norm Thai articulation development [24].

Resonance disorders: resonance was screened based on the perception of speech samples, which were composed of nonsense syllables of high oral pressure sounds, serial speech (counting 1–20 and 40–50), 3 simple phrases and sentences with high oral pressure consonants, as well as 3 nasal sentences. The resonance test was perceptually as normal (0) or resonance disorders; –1, hyponasality; and +1, mild; +2, moderate; and +3, severe hypernasality.

Voice disorders

Voice disorders: voice was rated based on a whole speech sample that was elicited during the first screening assessment. The voice was evaluated as normal or no voice disorder (0) and voice disorder (1).

Intelligibility: rating of intelligibility was scored base on a whole speech sample during the first screening assessment based on the whole speech sample as 0 = intelligibility, 1 = unintelligibility.

Statistical analysis

Descriptive analysis was used for presentation of prevalence of articulation disorders, resonance disorders, and voice disorders and expressed as percentages. Levene's test for equal variance was used to determine differences of articulation between age groups and resonance on articulation types.

Results

The characteristics of children with CP ± L are presented in **Table 1**. The proportion of girls to boys was approximately equal. Most of children were diagnosed with bilateral cleft lip and palate.

For articulation screening in the first visit, 5 of 42 (12%) children had normal articulation development (**Table 2**). When articulation patterns were classified, children with clefts had the highest incidence of both functional articulation disorders and CAD.

Related speech disorders that included resonance, voice disorders, and intelligibility were presented as **Table 3**. Moderate hypernasality was the most common in children with clefts.

Table1. Demographic characteristics of the children

Characteristic	Number	Percentage
Sex		
Female	22	52
Male	20	48
Total	42	100
Age (years)		
<4	4	10
4–7	22	52
7–15	10	24
16+	6	14
Total	42	100
Diagnosis		
1. Cleft palate	5	12
2. Left cleft lip and palate	12	29
3. Right cleft lip and palate	3	7
4. Bilateral cleft lip and palate	19	45
5. Bilateral complete cleft lip and palate	3	7
Total	42	100

Table 2. Articulation patterns

Articulation patterns	Number	Percentage
Normal	5	12
Functional articulation disorders	5	12
CAD	4	9
Functional articulation disorders and CAD	28	67
Total	42	100

CAD: compensatory articulation disorders

Table 3. Related speech outcomes

Speech outcomes	Number	Percentage
Resonance		
Normal	21	50
Hyponasality	0	0
Hypernasality	21	50
Mild	7	17
Moderate	13	31
Severe	1	2
Voice		
Normal	34	81
Voice abnormality	8	19
Intelligibility		
Intelligibility	42	100
Unintelligibility	0	0

Formal articulation tests that were performed during the second or later visits: 3 of 24 (7%) children with CP ± L had normal articulation development. The articulation pattern presented by most of CAD children was the abnormal backing of oral consonants (**Table 4**).

Table 4. Type of cleft characteristic articulation disorders

Cleft characteristic articulation disorders	Number	Percentage
Within normal limit/None	3	7
Abnormal backing of oral targets to post-uvular place		
- Pharyngeal	9	21
- Glottal	17	40
Abnormal backing of oral targets, but place remains oral		
- Mid-dorsal palatal	4	10
- Velar/uvular	15	36
Nasal consonant for oral pressure consonant	9	21
Nasalized voiced pressure consonants	2	5
Weak oral pressures	3	7
Developmental articulation errors	20	48
Phonological errors	6	14
Functional/other oral misarticulation	32	76
Coarticulation	2	5
Dental lisping	1	2

According to age groups (≤7 and >7 years), numbers of articulation errors were classified and Levene's test for equality found variances were unequal. An independent *t* test was used for analysis of mean difference, and results revealed that there were statistically significant differences between age groups when $P < 0.05$ was considered significant (**Table 5**).

For classification of articulation error types by resonance (with or without hypernasality), numbers of articulations were explored and Levene's test for equality found variances were equal. The independent *t* test was used for analysis of mean differences and results showed no statistically significant differences between resonance groups (**Table 6**).

Discussion

Twenty-six of total 42 children (52%) had the first visit for speech assessments when ≤7 years old, the appropriate period for speech therapy with easy correction because it is the stage of articulation development. The rest (38%) had the first visit or were referred for speech and language evaluation at >7 years old, which is the period for complete Thai articulation development [24]. This might be more difficult and take a longer time for speech correction because age and type of cleft are statistically significant factors in the development of normal articulation skills [25]. We recommend the protocol for early intervention and publicity to inform both health care providers and parents to realize the magnitude of articulation disorders and to therefore refer children with cleft to speech and language pathologists earlier.

For articulation screening, only 5 of the 42 (12%) children with CP ± L presented normal articulation (**Table 2**). Most of them (76%) had CAD (10% had functional articulation disorders and 67% had functional articulation and CAD). This confirmed that children with CP ± L had typical characteristics of cleft speech types and supports previous studies [26, 27]. When data from the first assessment (**Table 2**) were compared with the formal articulation test in the second or later visits (**Table 3**), the results showed the number of children with CP ± L who had normal articulation decreased from 5 (12%) to 3 children (7%). This indicated that the screening articulation test had false negatives in 2 of 5 children with CP ± L. This false negative might be difficult to detect because each child had only a single error; one child had only [b]/[w] in context of the vowel [u:a]: [bu:a]/[wu:a] and another had only [n]/[ɳ] in context of vowel [a]: [na:]/[ɳa]. They might be the screening test had short or fewer speech samples. Therefore, a formal articulation test is necessary for all children with CP ± L in a well-organized therapy plan.

Consistent with previous findings [27-30], most of children with CAD had the typical type of articulation in clefts in both the screening evaluations and formal articulation tests (**Tables 2 and 3**). **Table 4** displays that 39 (93%) of the children with CP ± L had articulation disorders. Most of the CAD is abnormal backing of oral consonants. Twenty-six or

Table 5. Independent *t* tests between age groups

Group	N	Mean of articulation number	Standard deviation	Mean difference	*P*	95 % Confidence interval
Children CP ± L ≤7 years	26	26	8.58	3.308	0.002	1.683–6.971
Children CP ± L >7 years	16	16	4.25			

CP ± L: Cleft palate with or without lip involvement

Table 6. Independent *t* test between articulation types by resonance

Group	N	Mean of articulation number	Standard deviation	Mean difference	*P*	95 % Confidence interval
Functional and compensatory with hypernasality	14	8.71	5.21	0.253	0.897	–3.736–4.241
Functional and compensatory without hypernasality	13	8.46	4.82			

61% of the children with CP ± L showed retracted oral articulation including that occurring in the posterior and beyond the uvular place: pharyngeal or glottal area and 19 children with CP ± L (46%) showed abnormal backing of oral targets, but oral: mid-dorsal palatal, velar, or uvular articulation. These results confirmed the previous article that the abnormal articulatory type in clefts was palatalized articulation that showed less of a tendency to correct itself spontaneously than did glottal stops [28]. These productions are the individual's response to inadequate intraoral air pressure for normal articulation or velopharyngeal or a palatal opening and considered as active speech characteristics [31] or compensatory errors [32]. While the compensatory productions are used, the manner of production is usually maintained. However, the places of articulation are altered and moved posteriorly to the pharynx or larynx. These allow the individual to take advantage of the air pressure that is available in the pharynx before it is reduced because of the velopharyngeal opening. Both articulation patterns, that were posterior and beyond the uvular place: pharyngeal or glottal and abnormal backing of oral targets, but oral: mid-dorsal palatal, velar or uvular were the most frequent substitution for oral sounds since occurrence of CAD requires higher levels of oral air pressure, therefore, abnormal backing is the common substitution for oral sounds. These CAD abnormalities are under patients' control and can be corrected with speech therapy [14, 33].

Thirty-two children (76%) with CP ± L presented with functional articulation disorders and 6 children (14%) with CP ± L had phonological errors. This prevalence was higher than articulation disorders in typically developing peers that found a prevalence of only 4%–31% [34, 35]. The most common functional articulation defect was [l]/[r]. This finding was consistent with that of the study by Van Lierde et al. [36]. It could be implied that children with clefts had a trend to miss-learn in acquisition of the general articulation development more than children without clefts because of an incomplete sensory and motor structure of the speech organ and abnormality of structures. Whenever there are structural anomalies, speech can be affected by obligatory distortions or compensatory errors. Obligatory distortions, including hypernasality because of VPI, are caused by abnormal structure and not by abnormal function. Therefore, surgery or other forms of physical management are needed for correction. By contrast, speech therapy is indicated for CAD where articulation placement is changed in response to the abnormal structure. Speech therapy is much more effective if it is performed after normalization of the structure. When speech therapy is appropriate, the techniques involve methods to change articulation placement using standard articulation therapy principles. These require performing the therapy and subsequent use during early preschool to prevent later difficulty with literacy.

Other speech outcomes such as resonance disorders were found in 50% of patients (**Table 3**). This prevalence was higher than those found in previous studies [8, 27, 37, 38], where the prevalence of resonance disorders or VPI ranged from 20% to 30%. Voice disorders were found in 8 of 42 children (19%) and this was in range of prevalence of 0 to 47% found previously [4, 39, 40]. This is usually because of increased respiratory and muscular effort, and hyper-adduction of vocal folds while attempting to close the velopharyngeal valve [33].

According to the numbers of articulation defects between age groups, younger children had more errors than older children. It was possible that the younger ones had not started or delayed speech therapy or some older ones might have received previous speech correction or have corrected by self-improvement when they passed the age of articulation development. For articulation types by resonance, there were no significant differences between groups with and without resonance abnormalities. It can be implied that articulation defects among children with clefts still persisted even though reconstruction of VPI was accomplished and that confirms previous studies [25, 28]. These compensatory habits should be corrected by speech therapy.

In summary, the prevalence of speech disorders in CP ± L in this study was higher than previous studies. It is possible that because data in this study were collected only from available retrospective records it might constitute a selective bias. Nevertheless, our findings suggest that continued refinement and revision of timing for referring to speech assessment and therapy should be considered and related factors should also be focused on improving the treatment goals and outcomes. Further prospective research is needed to confirm this.

Conclusion

CAD, abnormal backing of oral consonants, and resonance disorders were common speech defects

prevalence in children with CP ± L in our center. We recommend refinement and revision of timing for referring of CP ± L patients to speech assessment and early speech intervention should be considered, and related speech outcomes should also be brought into focus for improvement of the treatment goals and outcomes. Further prospective research with standard assessment is also required to confirm protocols and to provide evidence for improvement of treatment goals and outcomes.

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