Original Article

Prevalence of Speech and Language Delay in Children with Cleft Lip/Palate

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Background: Speech and language disorders—particularly delayed speech and language development [DSL]—are not uncommon for children with cleft lip and cleft palate [CLP]. If DSL is not treated in a timely and appropriate manner, it might affect the child's communication, literacy, and education. The magnitude of DSL in children with CLP has not been formally studied in Thailand.

Objectives: To investigate the prevalence of DSL in children with CLP.

Materials and Methods: 397 children with CLP registered at the Srinagarind Hospital Speech Clinic between 2009 and 2014 were included in the retrospective study. The speech and language screenings were scored (pass = 0 and fail = 1) based on the normal level with Adapted Early Language Mile Stone for Thai Children; Thai speech and language test for children between 1 and 2 years of age; Thai speech and language norms for children 2 1/2 to 4 years of age; and Utah Test of Language Development. Speech and language errors were listed as a guideline for speech therapy planning.

Results: Prevalence of DSL in children with CLP ranged between 4.3 and 44.40% (average 18.70%; 95% confidence interval = 14.90, 22.90). The common item of speech and language errors was drawing or painting (100%), copying common words (75%), telling days of the week (50%), telling full names and surnames (28.57%). This information is useful for speech therapy planning and further speech and language promotion.

Conclusion: Prevalence of DSL was a common defect in children with CLP. Early diagnosis and early intervention are needed to facilitate good quality of development, learning, and life.

Keywords: Prevalence, Speech and language delay, Cleft lip, Cleft palate, Delay speech and language development

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Cleft lip and cleft palate [CLP] are facial and oral malformations that occur in the 1st trimester of pregnancy. A cleft results when there is not enough tissue in the mouth or lip area, and the tissue that is available does not join together properly. A cleft lip is a physical split or separation of the two sides of the upper lip while a cleft palate is a split or opening in the roof of the mouth. A cleft palate can involve the soft palate (the soft back portion of the roof of the mouth) and/or the hard palate (the bony front portion of the roof of the mouth). Overall worldwide prevalence of CLP ranges between 0.11 and 1.00 per 1,000 live births[1-6] and between 0.58 to 2.49 per 1,000 live births in Thailand[7-10]. The prevalence of CLP in northeastern Thailand is the highest (2.4/1,000 live births) in Thailand[9]. Clefts require several surgical procedures and multidisciplinary treatments and care; the estimated conservative lifetime medical cost for each child with an orofacial cleft is $100,000, amounting to $750 million for all children with orofacial cleft born each year in the United States[5] or between 214,789 and 298,345 baht/child with CLP in Thailand[11]. According to the statistics for patients with CLP at Srinagarind Hospital, Khon Kaen, between 1984 and 2007, the majority of children with CLP were in the 4 provinces in the Nation Health Security System Area 7[12]. The rate of children with CLP registered to get speech services at Srinagarind Hospital increases annually.

One of the most common burdens regarding

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health care after cleft repair for the correction of the configuration and social stigma was speech and language defects, particularly delayed speech and language development [DSL]. The prevalence of DSL in children with CLP ranges between 20 and 92%[13-15]; ~50% of whom need speech and language therapy. Delayed 1st meaningful word, limitation of vocabulary, mean length of utterance, grammatical complexity and psycholinguistic are the most common defects[16], however, DSL was not directly related to hearing loss[17].

Due to the high prevalence of DSL, children with CLP should be assessed early for speech and language skills and get early intervention before or around their 1st birthday (9 to 12 months) and followed-up until 36 months[18,19]. The traditional articulation approaches and linguistic stimulation techniques-such as phonetic and phonological approaches as well as focused stimulation or whole-language approaches-should be evaluated early and the patient provided speech and language therapy in preschool. Other specific intervention models that require further testing include multisensory input modeling[20]. Language development in children with CLP should be complete within 5 to 6 years of age: a period of neurological development critical for brain development[21,22]. If patients are not assessed early and early interventions implemented, they will be at high risk for DSL. DSL will result in other development delays, including cognitive impairment, psychological problems, reading, and writing difficulties. Early speech and language assessment as well as early stimulation are necessary for children with CLP.

In Thailand, there are limited speech services due to a relatively small number of speech pathology professionals[23]. Most children with CLP cannot access speech services within the guideline period. The magnitude of DSL in CLP has not been formally studied in Thailand. In previous studies, the rates of DSL mostly derived from the limited number of participants or special characteristics[13,24,25]. Reports on the factors related to DSL in children with CLP are also not available.

The objectives of this study were to investigate the factors related to speech and language errors of DSL in children with CLP, and their prevalence. This information will be useful for developing further guidelines and management for speech and language stimulation as well as parental consultation.

Materials and Methods

This descriptive study with retrospective data collection was conducted by including children with CLP registered at the Speech Clinic at Srinagarind Hospital between 2009 and 2014. Children with CLP and other abnormalities (i.e., global delayed development, mental retardation, Down's syndrome, brain damage), which might have a negative affect on language skills were excluded. All data were retrieved from medical records including demographic data.

Speech and language tests used for speech and language evaluation on the 1st visit included (a) the Adapted Early Language Mile Stone for Thai Children (Adapted ELM) for children between 0 and 48 months[26], (b) the Thai speech and language test for children between 1 and 2 years (TSLT1-2 years)[27], (c) the Thai speech and language norms for children between 2 1/2 and 4 years (TSLT2-4 years)[28], and (d) the Utah Test of Language Development [UTAH] for children between 2 and 9 years[29]. The language test was scored as 0 = pass (or normal) and 1 = not pass (or fail) based on the criteria for normative speech and language development of each test[26-29]. The main outcome was a language score (pass or fail). Speech and language scores at the 1st assessment were analyzed as a percentage or rate of delayed DSL. For the 2nd through the 7th speech and language assessment, children with CLP were reassessed or followed-up in the same group after they received therapy or were older.

Descriptive statistics was used for data presentation.

Results

There were 765 children with clefts registered at the Speech Clinic at Srinagarind Hospital between 2009 and 2014. Three hundred and sixty-eight children were excluded because they (a) had abnormal conditions affecting speech and language development and/or (b) record forms of speech and language tests were not completed, so 395 children with CLP were enrolled. The baseline characteristics of children with CLP are presented in Table 1. Each child performed language tests 1 to 7 times depending on the age level of follow-up. Data analyses were based on language scores (pass or fail) at the first visit for nine age levels.

All children with clefts under 1 year of age were assessed for speech and language development using the Adapted ELM. The majority of children over 1 year of age were assessed using the UTAH, albeit the report for one child (age 4 years) was not available. The prevalence of DSL in children with clefts was analyzed based on the 1st language assessment of the

Table 1. Baseline characteristics of children with clefts at the first speech and language assessment

Age (year)	Number	Gender		Cleft type				
		Girl n (%)	Boy n (%)	CP n (%)	Lt. CLP n (%)	Rt. CLP n (%)	Bilat. CLP n (%)	Submucous cleft n (%)
<1	6	4 (66.67)	2 (33.33)	2 (33.33)	1 (16.67)	1 (16.67)	2 (33.33)	-
1	81	36 (44.44)	45 (55.56)	25 (30.86)	22 (27.16)	15 (18.52)	18 (22.22)	6 (1.23)
2	135	62 (45.93)	73 (54.07)	34 (25.19)	51 (37.78)	18 (13.33)	32 (23.70)	-
3	42	20 (47.62)	22 (52.38)	8 (19.05)	14 (33.33)	9 (21.43)	11 (26.19)	-
4	30	9 (30)	21 (70)	6 (20)	13 (43.33)	4 (13.33)	7 (23.33)	-
5	23	14 (60.87)	9 (39.13)	3 (13.04)	9 (39.13)	3 (13.04)	8 (34.78)	-
6	28	14 (50)	14 (50)	7 (25)	14 (50)	2 (7.14)	4 (14.29)	1 (3.57)
7	18	6 (33.33)	12 (66.67)	6 (33.33)	4 (22.22)	3 (16.67)	5 (27.78)	-
8	16	11 (68.75)	5 (31.25)	6 (37.5)	2 (12.50)	1 (6.25)	6 (37.50)	1 (6.25)
9	18	6 (33.33)	12 (66.67)	2 (11.11)	6 (33.33)	3 (16.67)	7 (38.89)	-
Total	397	182 (45.84)	215 (54.16)	99 (24.94)	136 (34.26)	59 (14.86)	100 (25.00)	3 (0.76)

N = Number; Lt. CLP = Left cleft lip and palate; Rt. CLP = Right cleft lip and palate; Bilat. CLP = Bilateral cleft lip and palate

Table 2. Prevalence of DSL in children with clefts

Age (year)	1st time			
	N	Non pass		95% CI
		n	%	
<1	6	0	0.00	0.00, 45.90*
1	81	6	7.40	2.80, 15.40
2	135	26	19.30	13.00, 26.90
3	42	15	35.70	21.60, 52.00
4	29	7	24.10	10.30, 43.50
5	23	1	4.30	0.10, 21.90
6	28	3	10.70	2.30, 28.20
7	18	3	16.70	3.60, 41.40
8	16	5	31.30	11.00, 58.70
9	18	8	44.40	21.50, 69.20
Total	396	74	18.70	14.90, 22.90

* one-sided, 97.50% confidence interval.
95% CI = 95% confidence interval

396 participants. The percentage of DSL and its 95% confident interval was calculated and presented in Table 2. The prevalence of DSL in all of the children under 1 year of age passed the normal scale indicating that none had DSL (prevalence = 0), so a one-sided, 97.50% confidence interval was used. Most of the children (69.77%) were assessed for speech and language assessment 1 to 2 times within 6 years (between 2009 and 2014), while the remainder (30.20%) were assessed more than 2 times within 6 years (average 3.57 times or once in 2 years). Regarding the 2nd to 7th language assessments, the data for prevalence of DSL are presented in Table 3. The percentage of children that passed or failed the various speech and language tests and the types of errors made are presented in Table 4.

Discussion

Most of the subjects with CLP were boys (54.20%) and the most common type was right CLP (34.30%). Submucous cleft palate was the least prevalence (3.00%). A speech and language assessment on the first visit was performed within 2 years (55.90%), while the remainder (44.10%) were evaluated after 2 years (Table 1); these tests indicated that most of the children did not have access to timely speech and language services, so treatment was delayed, especially among children with cleft who had delayed speech and language skills.

The 1st assessment showed that DSL ranged between 4.30 and 44.40% (Table 2) or averaged 18.70% (95% CI = 14.90, 22.90). Most of the children with CLP had appointments for speech therapy and re-evaluation; however, most could not come because (a) the family had a low economic status, (b) caregivers were grandparents and could not come, and/or (c) there was no time for bringing the children early and regularly. In addition, speech services in the northeastern region of Thailand are limited.

Table 3. Prevalence of DSL in the 2nd to 7th visits

2nd time				3rd time				4th time			
N	Non pass		95% CI	N	Non pass		95% CI	N	Non pass		95% CI
	n	%			n	%			n	%	
220	61	27.70	21.90, 34.10	120	16	13.30	7.80, 20.70	72	8	11.10	4.90, 20.70
5nd time				6rd time				7th time			
N	Non pass		95% CI	N	Non pass		95% CI	N	Non pass		95 % CI
	n	%			n	%			n	%	
37	3	8.10	1.70, 21.90	12	0	0.00	0.00, 26.50*	2	0	0.00	0.00, 84.20*

The prevalence of DSL was high, especially among children 3 years old, suggesting children with CLP might have DSL during infancy. This result is supported by a previous study that found these children produced less babbling noises[30], persistent vocalization, and vocabulary deficits well beyond palate closure[31]. By contrast, a previous study found there was no significant difference between typical infants and children with cleft[32]. Further research is needed to explain delays among prelinguistic children with clefts.

Early diagnosis and early intervention or prelinguistic programs should be provided for language promotion and prevention of delays for children with CLP[33-40]. For older children (8 to 9 years), the prevalence of DSL was highest (i.e., 31.30% and 44.40%, respectively). This might be because Thai children were not concerned about the items represented on the speech and language screening test (i.e., telling days the day of the week (50%) or imitating sentences longer than 10 words (30%). The trend in DSL (Table 3) seemed to decline among as children grew older as interventions were implemented. The number of participants undergoing a 2nd to 7th visit, particularly the 6th to 7th visit, was small, so the rate of DSL should be interpreted with care.

When data were grouped, the prevalence of DSL for children with CLP, under 2 years, was 14.40% (32/222), and the prevalence of DSL for children with CLP over 2 years was 24.10% (42/174). These results suggest that the prevalence of DSL among older children with CLP was significantly higher than for younger children with CLP; however, this study was retrospective research so the data on older children were less available from the medical records-(a) not available (b) the patients did not come for follow-up or (c) the patients did not undergo language testing. The results of the present study should, thus, be interpreted with care based on the limitation of available data.

The most common deficits (Table 4) included telling full name and surnames (28.60%) for children 4 years old, drawing or painting (100%) for children 5 years old, copying 2 to 3 common words (75%) for children 6 years old, and telling the days of the week (50%) for children 7 years old. As most other children pass all these items, these represent key indicators that if failed early intervention is advised for children with CLP.

In summary, speech and language early in life are integral to other development. Early diagnosis and early intervention are needed to facilitate good quality of development, learning, and life.

What is already known on this topic?

DSL is one of the common deficits among children with CLP. There are very few studies related to this topic in Thailand. A prior study provided the overall prevalence via speech and language screening tests, but that did not include items of speech and language errors that might be important guides for planning interventions.

What this study adds?

The prevalence of DSL at each age level was presented. The results indicate high rates of DSL among

Table 4. Speech and language errors

Age (years)	Test	Items of speech and language errors	%
<1	ELM (100%)	-	
1	ELM (100%)	- Mono babbling	5.60
		- Poly Syllabic babbling	5.60
		- Mama/Papa: Any	16.70
		- Single word imitation	16.70
		- Mama/Papa: Correct	11.10
		- First word	11.10
		- Production 4 to 6 single words	11.10
		- Tell want	5.60
		- Point to body parts	11.10
	TSLT (1%)	-	
2	ELM (100%)	- 2 step command	2.00
	TSLT (16.30%)	- Identify common objects	12.90
		- 2-step command	9.30
		- 3-step command	11.10
		- Identify dress	4.00
		- Understand common verbs	11.10
		- Naming common actions	7.40
		- Imitation sounds in environment	7.40
		- Tell want	4.00
	UTAH (80.70%)	- Identify common object pictures	5.60
		- Identify body 3 to 5 parts	4.00
		- Imitation 3 to 5 words	2.00
3	ELM (2.38%)	- Using preposition	5.30
		- Carry conversation	5.30
	UTAH (97.60%)	- Naming 1 color	26.30
		- Understand common opposite words	21.10
4	ELM (3.30%)	-	
	UTAH (96.70%)	- Imitation 3 number	14.30
		- Telling full name and surname	28.60
		- Copy a cross	14.30
5	UTAH (100%)	- Drawing or paint	100
6	UTAH (100%)	- Copy square	25.00
		- Copy common 2 to 3 words	75.00
7	UTAH (100%)	- Write number 1 to 30	25.00
		- Telling story	25.00
		- Reading easy word	25.00
8	UTAH (100%)	- Telling days in a week	50.00
9	UTAH (100%)	- Imitation complicated sentence longer than 10 words	30.00
		- Telling current date and time	10.00
		- Telling direction	10.00

Adapted ELM = Adapted Early Language Mile Stone; TSLT = Thai speech and language test; UTAH = Utah Test of Language Development

children with CLP. The study provides examples of speech and language errors useful for planning early intervention.

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Potential conflicts of interest

The authors declare no conflicts of interest.

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