

A Preliminary Study on Clinical Profiles of Non-Syndromic Cleft Lip with or without Cleft Palate During Community Services in Indonesia 2020-2022

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Abstract

This study aimed to report the clinical profile of individuals with non-syndromic cleft lip with or without palate (NSCL/P) during community services in Indonesia in the period 2020-2022.

We conducted a retrospective study of NSCL/P cases treated during community services in four provinces in Indonesia (West Java, Riau, South Sulawesi, and West Nusa Tenggara), for the period from 2020 to 2022. The medical records of the subjects found to be incomplete were excluded from the study. The chi-square tests were performed to calculate the relationship between gender, the type and the site affected of orofacial cleft.

Total of 4565 individuals with NSCL/P registered during community services in the study period. The majority of the subjects were males (62.3%), in the age group of 3-6 months (24,18%). The most common cleft type was cleft lip (55.4%), and unilateral was the most prevalent site affected (80,5%). There was no significant relationship between the type of cleft and gender ($p>0.05$), but we found a significant relationship between cleft type and age ($p<0.001$) and the site affected ($p<0.001$). The most performed treatment was labiaplasty (56.1%), followed by palatoplasty (25.5%).

The NSCL/P in this study were more common in males. The most frequent cleft type and treatment were unilateral cleft lip and labiaplasty. Cleft type was significantly associated with age and the site affected, but not with gender. Future research should consider investigating the risk factors of NSCLP that may explain the high prevalence of orofacial clefts in Indonesia.

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Introduction

Cleft lip and/or palate (NCL/P) is the most frequent of birth defects or congenital abnormalities that occurs during early embryological development due to failure of the facial processes to grow or fuse properly.¹ It is believed that the causes of NSCL/P are complex and multifactorial, including both genetic and environmental factors.²⁻⁴ The incidence rate of NSCL/P varies among different parts of the world, with approximately 1 in 500-2500 live births.

NSCL/P more frequently occurs among Asian and American Indian descent (1 per 500-1000 live births), while the prevalence rate at about 1 per 1000 live births among European descent, and the lowest prevalence rate has been reported among African American population (1 per 2500 live births).^{2,5} Differences in the incidence of NSCL/P worldwide may be due to various factors, including genetics, ethnicity, geographic origin, socioeconomic status, environmental factors such as vitamin and folic acid deficiency, and maternal behaviours during pregnancy (medication use, alcohol consumption, smoking, maternal obesity).⁵⁻⁷

The type of NSCL/P is divided into cleft lip (CL), cleft palate (CP), and cleft lip and palate (CLP), unilateral or bilateral, most commonly the palate or upper lip, or both.⁸⁻¹² The affected individuals face feeding difficulties early in life and usually require several corrective surgeries,

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dental procedures, and speech therapy during childhood.^{13,14} In addition, individuals born with an orofacial cleft have an increased incidence of mental health problems and overall mortality at all stages of life, even in developed countries with good medical care.^{15,16} When access to medical care is very limited, infants with CL/P generally have a high mortality rate due to breastfeeding difficulties, while untreated cases of CL/P may face social discrimination throughout their lives. As such, these birth defects have been subjected to selective pressure for most of human history.

The aim of this study was to investigate the prevalence and clinical profile of CL/P during community service activities in four region in Indonesia, including West Java, Riau, South Sulawesi, and West Nusa Tenggara, from 2020 to 2022. This study might provide information in monitoring of the occurrence of CL/P and can be used to prevent the diseases in Indonesia.

Materials and methods

This research was a retrospective cross-sectional study. The data were collected from medical records of patients with NSCL/P during community services in four provinces in Indonesia (West Java, Riau, South Sulawesi, and West Nusa Tenggara), for the period from 1 January 2020 until 31 December 2022. The data collected includes demographic information, diagnosis, and treatment. The medical records of the subjects found to be incomplete were excluded from the study. The chi-square test was used to calculate the significant association between gender, the type, and the site affected of orofacial cleft.

This study was approved by the Research Ethics Committee of the University of Padjadjaran (No. 649/UN6.KEP/EC/2021) and was conducted according to the guidelines of the Declaration of Helsinki.

Results

A total of 4565 study subjects with NSCL/P were included in this study. Table 1 showed that the majority of subjects with NSCL/P (24.2%) were in the age range of 3-6 months, followed by the group aged of 2-5 years (21.8%). The least number of patients with NSCL/P were found in the age group of 6-12 months (8.9%).

This study also demonstrated males have a greater proportion (62.3%) than females (37.7%).

Characteristics	Total N = 4565	Percentage (%)
Age		
- 3-6 months	1104	24.2%
- 6 months - 1 years	405	8.9%
- 1 - 2 years	780	17.1%
- 2-5 years	995	21.8%
- 5-10 years	485	10.6%
- > 10 years	816	17.9%
Gender		
- Male	2843	62.3%
- Female	1722	37.7%

Table 1. Characteristics of research subjects.

Type	Male n=2843 (%)	Female n=1722 (%)	Total N = 4565 (%)	P- value*	CI 95% OR (Lower-Upper)
Cleft lip	1554 (61.5%)	973 (38.5%)	2527 (55.4%)	0.130	0.92 (0.83 - 1.01)
Cleft palate	1103 (62.6%)	660 (37.4%)	1763 (38.6%)		
Cleft lip & palate	186 (67.6%)	89 (32.4%)	275 (6.0%)		

Table 2. Association between the type of non-syndromic cleft lip and/or palate and gender.

Type	Location		Total	P- value*	CI 95% OR (Lower - Upper)
	Unilateral (n=3481)	Bilateral (n=1084)			
Cleft lip	2035 (80.5%)	492 (19.5%)	2527 (100.0%)	<0.001	1.55 (1.39 - 1.72)
Cleft palate	1267 (71.9%)	496 (28.1%)	1763 (100.0%)		
Cleft lip & palate	179 (65.1%)	96 (34.9%)	275 (100.0%)		

Table 3. The type of non-syndromic cleft lip and/or palate according to the site affected, one site (unilateral) or both sites (bilateral).

Based on the type of cleft, we found that cleft lip (CL) (55.4%) is more frequent than cleft palate (38.6%) and both cleft lip-palate (6.0%) as shown in Table 2. The chi-square (χ^2) test showed that there was no significant relationship ($p > 0.05$) between the type of cleft and gender. Meanwhile, based on the site affected, the most prevalent of all cleft types were unilateral (Table 3). The chi-square (χ^2) test revealed that there was a significant relationship ($p < 0.001$) between the type of cleft and its site affected (adjusted odds ratio: 1.55; 95% confidence interval: 1.39 - 1.72).

The relationship between the types of cleft and the patient's age was presented in **Table 4**. The chi-square test (χ^2) showed that there was a significant relationship ($p < 0.001$) between the type of cleft and age (adjusted odds ratio: 0.525; 95% confidence interval: 0.388 - 0.712). Table 5 showed the treatment given to the study subjects. The most performed treatment was labiaplasty (56.1%), followed by palatoplasty (25.5%).

Age	Type Cleft Lip (n=2527)	Cleft Palate (n=1763)	Cleft Lip & Palate (n=275)	P-value*	CI95% OR (Lower - Upper)
3-6 months	691 (62.6%)	247 (22.4%)	166 (15.0%)	<0.001	0.525 (0.388 - 0.712)
6 months-1 years	361 (89.1%)	44 (10.9%)	0 (0.0%)		
1 - 2 years	789 (100.0%)	0 (0.0%)	0 (0.0%)		
2-5 years	0 (0.0%)	995 (100.0%)	0 (0.0%)		
5-10 years	156 (33.5%)	268 (57.6%)	41 (8.8%)		
> 10 years	539 (66.1%)	209 (35.6%)	68 (8.3%)		

Table 4. Association between the type of non-syndromic cleft lip and/or palate and age.

No.	Surgical Type	Total N = 4565	Percentage (%)
1.	Labiaplasty	2562	56.1%
2.	Primary Lip Nose Bilateral	53	1.2%
3.	Primary Lip Nose Bilateral + Lip Nose Revision	2	0.0%
4.	Primary Lip Nose Unilateral	248	5.4%
5.	Primary Lip Nose Unilateral + Lip Nose Revision	3	0.1%
6.	Primary Lip Nose Unilateral + Other	1	0.0%
7.	Labio-palatoplasty	72	1.6%
8.	Fistula repair	144	3.2%
9.	Fistula repair +Lip nose revision	3	0.1%
10.	Fistula Repair + Secondary Cleft Palate	1	0.0%
11.	Lip nose revision	97	2.1%
12.	Palatoplasty	1166	25.5%
13.	Reconstructive Labiaplasty	128	2.8%
14.	Gnatoplasty	55	1.2%
15.	Veloplasty	11	0.2%
16.	Bone graft	19	0.4%

Table 5. The treatment intervention for patients with non-syndromic cleft lip with or without cleft palate.

Discussion

The present study revealed that the most common individual with NSCL/P was found at the age of 3-6 months. The patients came to the community services sought the treatment of orofacial cleft. It has been known that cleft lip cases are recommended to be repaired at 3 months of age or in accordance with the rule of 10s that the patient should be at least 10 weeks old, weight 10 pounds, and have a hemoglobin

level over 10 g/dL.^{17,18} This is in line with our finding regarding the type of cleft that the most prevalent cleft type found in the present study was cleft lip. Data from this study was also similar to the previous study showing that orofacial clefts have been reported to be more frequent in males compared to females. The findings are similar to previous studies conducted in South America, South Africa, and Iran that NSCL/P were more frequent in males.^{19,20,21} The predominance of males in the prevalence of clefts has also been reported by studies conducted in Iran and Korea.²²

According to the type of cleft, cleft lip (CL) cases were ranked first, followed by cleft palate, and cleft lip and palate. The results were different from the previous studies that showed cleft palate (CP) had the highest frequency of all diagnoses, followed by CLP, and CL.^{19,23} Meanwhile, Study other studies in Damascus and Turkey showed that the most common type of cleft was CLP, followed by CP, and CL.^{24,25} In North-Eastern Iran, CLP was the most frequent type of cleft found, followed by CL, and CP.²² Our study revealed that there was no significantly relationship between the type of cleft and gender. This finding also similar with the prior study in Iran and Saudi Arabia.^{21,26} The ratio of males to females has been reported including 2:1 ratio of CL, 1:2 ratio of CP, and 2:1 ratio of unilateral cleft lip cases.²⁷

With regards to the affected side, our study showed that the unilateral cleft was more frequent than bilateral. This is in line with previous studies that unilateral clefts are more common than bilateral clefts with a ratio of 4:1.¹⁶ The statistically analysis revealed that there was a significant relationship between the type of cleft and its site affected, similar to a study in Turkey.²⁵ The number and various different types of orofacial/ NSCL/P cleft worldwide may due to geographic origins, race or ethnicity, socioeconomic status, and maternal exposure to environmental factors such as health/disease status, malnutrition, also lifestyle such as tobacco, alcohol, and medication/drugs use.²⁸ The previous studies have reported the genetic variation that may related to the risk of CL/P among Indonesian population.²⁹⁻³²

Various treatments can be performed for orofacial cleft, depend on the age and need of the patients, the severity of the cleft, the presence of associated syndromes, and other

birth defects. This present study indicated that labiaplasty treatment has the highest number of treatments compared to other treatments. Labiaplasty was recommended to be carried out at the age of three to four months, followed by treatment of the palate at the age of six to nine months.³³ But then, palatoplasty is often neglected in developing countries after labiaplasty procedures. The secondary intervention mostly performed were fistula repair and cheiloplasty. Individuals with orofacial cleft will need additional treatments as they get older.³⁴

The limitation of this study is that the secondary data were obtained without conducting direct interviews. There are some data in medical records that are not completely documented.

Conclusions

This study revealed that NSCLP in Indonesia was more common in males than females. Cleft lip was the most prevalent cleft type, and the majority of subject had unilateral cleft. There was no significant relationship between the type of cleft and gender, but cleft type was found to be significantly associated with age. Labiaplasty was the most frequent treatment among the patients in this study. Future research should consider investigating the risk factors of NSCLP that may explain the high prevalence of orofacial clefts in Indonesia.

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Declaration of Interest

The author reports no conflicts of interest in this work.

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