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A descriptive epidemiology study of cleft lip and palate in Northern Finland

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Abstract

Objective. The aim of this study was to determine the incidence of cleft lip and/or cleft palate in a population uniquely from Northern Finland. ***Materials and methods.*** The records of a total of 214 cleft patients treated between 1998–2011 at the Oulu Cleft Lip and Palate Center at the University of Oulu were assessed on a retrospective basis. Data regarding cleft type, sex and side of cleft was collected and analyzed. Family history of clefting was investigated. ***Results.*** Cleft palate (68.7%) was most frequently found, followed by cleft lip and palate (18.7%) and cleft lip with or without alveolus (12.6%). Cleft palate occurred more frequently in females (63.3%) and cleft lip and palate was more frequently found in males (62.5%). The left side was more frequently affected in both male and female patients. Left-sided clefts were observed in 82% of patients compared to right-sided clefts in 18%. A family history of clefting was detected in 20.1% of patients. ***Conclusion.*** The incidence of clefts in Northern Finland is higher than the corresponding incidence in other European countries. Cleft palate was the most frequent cleft type and it was more frequent in females. In males, cleft lip and palate was more frequent. The left side was more frequently affected in both genders. One fifth of the patients had a family history of clefts.

Key Words: *cleft lip and or palate, incidence, side of cleft, family history of cleft*

Introduction

Cleft lip and/or palate (CL/P) is the most frequently encountered congenital craniofacial anomaly [1]. Generally clefts are divided into two groups, cleft lip with or without cleft palate (CL/P) and isolated cleft palate (CP). Cleft palate appears when the palatal shelves fail to fuse between the 8–12th weeks of embryonic development [2]. Clefting varies in severity, for example only involving the lip, only involving the palate and, in more extreme cases, extensive facial clefts can also occur.

The incidence of clefts varies between countries and racial or ethnic groups [3]. Generally, the worldwide incidence of clefts is estimated to be between 1–2.21 cases per 1000 live births [4]. In Europe, significant differences in incidence have been found within countries and also between registries. The overall incidence of clefts is clearly greater in Finland than in other European countries. The incidence of clefts in Finland is 2.56 cases per 1000 live births and abortions. The corresponding incidence in other European countries is reported as 1.34 cases per live births and abortions [5].

The incidence of CP in Finland is 1.36 cases per 1000 live births, which is also greater than in other European countries where the incidence is 0.53 cases per 1000 births. The incidence of CL/P in Finland is 0.96 and elsewhere in Europe there are 0.81 cases per 1000 live births. The ratio of CP/CL/P is 1.3 in Finland and in addition the distribution of clefts varies regionally. In Northern and Eastern Finland CP incidences are higher, whereas in Southern and Western Finland the CL/P incidence is higher [6]. Previous data from other parts of the world has reported CL/P to have a higher incidence than CP [4], whereas the opposite seems to occur in North Finland where CP is more common than CL/P.

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When observing the side involved by the cleft lip, the left side seems to be more frequently affected than the right side. A study in Kenya [7] found that cleft lip was more frequent in male patients, especially on the left side. A parallel outcome was reported in a Mexican study [8] and a Belgian study [4].

With such a variable incidence throughout the world, the authors aimed to analyze the incidence of lip and palatal clefts in a population of patients uniquely from Northern Finland.

Materials and methods

This retrospective study included 214 cleft patients who were treated between 1998–2011 at the Oulu University Hospital Cleft Lip and Palate Center. The study was approved as a retrospective study by the ethical committee of the Oulu University Hospital and the data were treated according to the principles of the Helsinki Declaration of 1975, as revised in 1983. The patients' charts were accessed through the hospital's medical database. The following retrospective data were collected including: patient's sex, type of cleft, cleft side, presence of syndromes and history of cleft in other members of family, if known. The data were analyzed with the use of IBM SPSS statistics 20.0 software (Chicago, IL) for Windows and differences with $p < 0.05$ being considered as statistically significant.

Results

The study included 214 patients where 25 patients had an associated syndrome with Robin sequence occurring in eight patients. The subjects were grouped as follows: cleft lip with or without alveolus (CL), cleft lip and palate (CLP) and cleft palate (CP). Cleft types were further sub-divided into unilateral (UCLP) and bilateral (BCLP) and according to the side involved by the cleft (left or right). Of the 214 patients in the study, 27 had cleft lip, 40 had cleft lip and palate, 28 had unilateral cleft lip and palate, 12 had bilateral cleft lip and palate and 147 had cleft palate (Table I).

The patient pool consisted of 119 girls and 95 boys. Cleft lip (CL) was mildly more frequent in male patients (16; 59%) than in female patients (11; 41%).

Table I. Cleft distribution according to sex, *n* (%).

	Cleft classification			
Sex	CP	CLP	CL	Total
Female	93 (63.3)	15 (37.5)	11 (40.7)	119 (55.6)
Male	54 (36.7)	25 (62.5)	16 (59.3)	95 (44.4)
Total	147 (68.7)	40 (18.7)	27 (12.6)	214 (100)

CP, cleft palate; CLP, cleft lip and palate; CL, cleft lip.

Table II. Cleft side distribution according to sex, *n* (%).

	Cleft side distribution according to sex, n (%)			
Sex	Left	Right	Bilateral	Total
Female	19 (45.2)	3 (33.3)	4 (26.7)	26 (39.4)
Male	23 (54.8)	6 (66.7)	11 (73.3)	40 (60.6)
Total	42 (19.6)	9 (4.2)	15 (7.0)	66 (100)

Cleft lip and palate (CLP) were more frequent in males (25; 63%) than in females (15; 37%). When the cleft lip and palate data was sub-divided into unilateral clefts (UCLP), the distribution of males was 15 (54%) and females 13 (46%). Bilateral cleft lip and palate (BCLP) was more frequent in boys, being 10 (83%) compared to two (17%) in girls. Cleft palate (CP) was more frequent in females (93; 63%) than in males (54; 37%). The distribution of cleft types by sex was statistically significant when considering CP, CLP and BCLP ($p = 0.006$).

The left side was more frequently affected in both male and female patients compared to the right side in patients with UCLP. The cleft was located on the left side in 42 (82%) patients and on the right side in nine (18%) patients. The distribution between males and females was not statistically significant, males had 23 and females had 19 left-sided clefts. The right side was affected in six males and three females (Table II). The cleft was bilateral in 16 patients.

Regarding a history of clefts in other family members, 20.1% of all patients had at least one relative with a cleft. There was no statistical significance when comparing maternal and paternal hereditary (Table III). CP was the most common cleft type when regarding family-related clefting (76.7%), whereas family history was found in 16.3% of CLP patients and in 7% of CL patients.

Discussion

Many epidemiologic studies from different countries and regions have evaluated the distribution of orofacial

Table III. Family history of cleft.

Cleft family history	Cleft classification			
	CP	CLP	CL	Total
Maternal	14 (6.54)	5 (2.34)	1 (0.47)	20 (9.35)
Paternal	11 (5.14)	2 (0.93)	2 (0.93)	15 (7.00)
Both parents	3 (1.40)	0 (0)	0 (0)	3 (1.40)
Sibling	5 (2.34)	0 (0)	0 (0)	5 (2.34)
No	114 (53.27)	33 (15.42)	24 (11.22)	171 (79.91)
Total	147 (68.69)	40 (18.69)	27 (12.62)	214 (100)

CP, cleft palate; CLP, cleft lip and palate; CL, cleft lip.

clefts. These studies have shown that incidences and distribution in clefting varies substantially between different countries, regions, ethnic groups and races [4,5,9,10]. To our knowledge, there is no previous study involving the population of Northern Finland. In Finland, it is fortunate that there is an already existing thorough and reliable registration system for birth defects. The National Institute for Health and Welfare collects information regarding congenital anomalies from all regions of Finland and its University Hospital districts.

In the current study the frequency distribution for clefts was CP (68.7%), CLP (18.7%) and CL (12.6%) (Table I). These results are in agreement with other studies that involve Scandinavian populations. The overall incidence of cleft in all Scandinavian countries is similar, but there is a difference in the distribution of cleft types. In Finnish populations patients with CP are the largest group, but in other Scandinavian countries CL/P constitute a more common cleft type than CP [11–14]. The incidence of clefts is higher in Finland than other European countries [6]. Studies have also shown that CL/P is more common in Europe than CP [4,15,16]. When comparing Finland to other countries of the world such as Australia [17], the US [18], Brazil [19,20] and Taiwan [21] CL/P incidences are reported to be higher than CP. The groups of CL and CLP are the minority in this study. This makes this sub-set of patients interesting, as other European studies have different distributions in the cleft patients.

The population sample from Northern Finland that was studied showed a greater incidence of CP in females (63.3%) than in males (36.7%). This result is similar to studies in several European countries [4,16,22], Australia [17], Pakistan [23] and Kenya [7]. In the case of CL/P, these were more frequent in males (59.3 and 62.5%) than in females (37.5 and 40.7%). Parallel outcomes have been found in different populations: Sweden [13], Spain [10] Mexico [8], Kenya [7] and Brazil [19].

Numerous studies have shown that CL/P occurs more frequently on the left side [4,7,8,13,14,16]. According to our study, the left side was also more frequently affected (82%) than the right side (18%) (Table II), although there was no statistically significant difference between genders.

The two most common named syndromes or associated malformations in this study were Catch-22 (Velocardiofacial Syndrome) and Pierre Robin Sequence.

In our study, 20.1% of patients had a family history of clefts (Table III). There was no statistical significance regarding paternal or maternal hereditary. The highest incidence in family-related clefting was found in patients with CP (76.7%). A family history of clefts was found in 16.3% of CLP patients and in 7% of CL patients. Distribution of family-related clefts seems to follow the overall distribution of cleft types. Other studies have shown variable proportions of patients with family history of clefts. A study from Spain [10] found that 21.9% of patients had an associated family history of clefts. The same study noted that clefting seemed to occur more often on the father's side of the family than on the mother's. A study from Sweden [13] reported a positive family history in 26.2% of CLP patients. Another study from Kenya [7] found that 3.5% of CLP patients had a positive family history. In a Pakistani study [23], a positive family history was noticed for an orofacial cleft in 17% of cleft patients. These diffrences could be explained partly due to regional and racial variations. Differences could also be explained due to statistical unreliability.

The high percentage on clefts in other family members noted in this study supports the multifactorial and hereditary characteristic of clefts. Many genes and environmental teratogenes have been implicated to play a role in the etiology of clefting [5,24]. It would be worthwhile to integrate genetic research and epidemiologic studies to help explore further the linkage between etiology and incidence of clefting.

Knowing that the most common cleft deformity in Northern Finland is isolated cleft palate has an important effect on resource planning. Cleft lip and palate is associated with a significant burden of care for both the parents of such children and for their communities. The need for speech-related treatment would be higher in cleft palate patients than those patients with isolated cleft lip deformities for example.

Conclusion

We conclude that there are some key differences in this population of patients uniquely from Northern Finland when compared to other reports from elsewhere in the world. In Northern Finland cleft palate is more frequent than cleft lip and palate. Cleft palate occurs more frequently in females than in males. As for cleft lip and palate, males are more frequently affected than females and clefting located on the left side is most common. This cohort of cleft patients was associated with a strong family history of clefting. In the future, genetic analysis could be included with epidemiologic studies.

Declaration of interest: The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

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