

Epidemiology, Clinical Aspects and Management of Cleft Lip and/or Palate in Burkina Faso: A Humanitarian Pediatric Surgery-Based Study

Kisito Nagalo^{1,2*}, Isso Ouédraogo^{2,3}, Jean-Martin Laberge^{4,5}, Louise Caouette-Laberge^{4,6}, Jean Turgeon^{4,7}

¹Service of Pediatrics, Clinique El Fateh-Suka, Ouagadougou, Burkina Faso

²Training and Research Unit of Health Sciences, University of Ouagadougou, Ouagadougou, Burkina Faso

³Department of Pediatric Surgery, Charles De Gaulle Pediatric University Teaching Hospital, Ouagadougou, Burkina Faso

⁴Mission Sourires d’Afrique, Montreal, Canada

⁵Department of Pediatric Surgery, McGill University, Montreal, Canada

⁶Department of Surgery, University of Montreal, Montreal, Canada

⁷Department of Pediatrics, University of Montreal, Montreal, Canada

Email: kiki_nagalo@yahoo.fr

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Abstract

Background: Cleft lip and/or palate are the most common orofacial malformations. Many studies, especially in developed countries have been conducted on this malformation, but in Burkina Faso, data are scarce and they are not specific to children. The aim of this study was to report the epidemiological, clinical and therapeutic aspects of cleft lip and/or palate in children in a low-income country. **Materials and Method:** The authors conducted a retrospective descriptive study based on data of three humanitarian missions of pediatric reconstructive facial surgery which took place in 2007, 2010 and 2014 at Clinique El Fateh-Suka in Ouagadougou, Burkina Faso. All children of 0 - 14 years of age, presenting with cleft lip and/or palate, were included in the study. **Results:** A total of 185 cases of cleft lip and/or palate were seen during these three humanitarian surgery missions. There were 100 boys and 85 girls. The average age of the children was 2.4 ± 3.2 years [0 - 12 years]; there were 8.7% newborns. The commonest type of cleft was cleft lip and palate (49.7%) followed by isolated cleft lip (48.7%) and isolated cleft palate (1.6%). The left side was the most affected (49.2%). In 21.1% of cases, clefts were associated with other congenital malformations. In total, 150 of 185 (81.1%) children underwent surgery and there were no postoperative complications reported. **Conclusions:** Epidemiological and clinical characteristics of cleft lip and/or palate

*Corresponding author.

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observed in this study are not very different from those described elsewhere in Africa. However, in our conditions, there are circumstances and structural factors which hinder the diagnosis and constitute challenges that must be addressed for adequate management of this congenital, highly disfiguring malformation.

Keywords

Orofacial Clefts, Cleft Lip and/or Palate, Congenital Malformations, Humanitarian Surgery, International Cooperation

1. Introduction

Cleft lip and/or palate (CL/P) are an embryonic syndrome which consists of cleft lip, cleft lip and palate, and cleft palate, and which accounts for 65% of all the congenital malformations of the head and the neck [1]. Ethnic and geographical variations show a high prevalence between 0.55 and 2.50/1000 births in Mongolian populations, between 0.69 and 2.35/1000 in Caucasians; a lower prevalence of between 0.18 and 0.82/1000 in Afro-Americans is reported [2]. In Africa, the prevalence is also lower, estimated at 0.5/1000 in Nigeria [3]. The etiology of this malformation is complex and includes both genetic and environmental factors. Data on orofacial clefts actually are sparsely available in respect of Africa, and this is even truer in Burkina Faso where there are no published studies on CL/P dedicated to the pediatric subpopulation. It is a recent study which revealed that CL/P constituted 70.6% of all congenital malformations of the face and the neck in Ouagadougou [4]. In Burkina Faso, care of orofacial clefts is difficult, and to alleviate the governmental inadequacies, local, European and Canadian Non-Governmental Organizations (NGO) organize humanitarian campaigns to bring the specialized care of surgery to the patients. This study is aimed to describe the epidemiological, clinical aspects and management of CL/P among children seen within the framework of humanitarian missions of pediatric plastic surgery in Ouagadougou in Burkina Faso.

2. Materials and Method

2.1. Study Site

Burkina Faso is a landlocked country in the Sahelian region in western Africa which covers 272,967 km². It is populated with 14,017,262 inhabitants of whom more than 77% live in rural areas. The distribution of the population by age group shows that children 0 - 14 years, number 6,499,211. The population is in 49% under 15 years old. Economically, Burkina Faso is one of the poorest countries in the world, the gross national income is of US \$1560; 44.46% of the population live with less than US \$ 1/day. The sanitary statistics indicate a life expectancy at birth of 57 years; a rate of malformations of 4.1%; anemia has 88% prevalence in children under 5 years, 58% in pregnant women, and 50% in breast-feeding women [5]. The country is subdivided into 45 provinces including the Province of Kadiogo (12.3% of the population) situated in the center of the country in which the capital, Ouagadougou (10.5% of the population), is located. The public sanitary organization included three levels of care; the highest is constituted by the University Teaching Hospital (UTH) which is the reference level for specialized care. There are four UTHs in the country (3 in Ouagadougou and 1 in Bobo-Dioulasso, the 2nd largest city). This study took place at Clinique El Fateh-Suka (CFS) which is situated in Ouagadougou. It is a private hospital created by "Fondation Suka", a NGO the major objective of which is the improvement of the health of the mother and child in Burkina Faso. The partnership between "Fondation Suka", Canadian NGO "Missions Sourires d'Afrique", and international NGO "Smile Train", permits a Canadian medical and surgical team travelling to Ouagadougou to operate upon children presenting with oral or maxillofacial anomalies (*i.e.*, CL/P, aftereffects of Noma disease or burns), and to transfer the skills in the field of the pediatric plastic surgery to local surgeons. Burkina Faso still lacks a register of orofacial clefts.

2.2. Type, Population and Period of Study

It was a retrospective descriptive study of all the patients seen during the three missions of pediatric plastic sur-

gery in November 2007, November 2010 and February 2014.

2.3. Criteria of Inclusion and Noninclusion in the Study

All patients aged 0 - 14 years at the time of the current surgical mission who consulted for a CL/P were included in the study. Patients, who were more than 14 years old, and those who did not have a CL/P, were excluded.

2.4. Procedures

Before the arrival of the Canadian team and the beginning of surgery, a national media campaign (radio, television, newspapers and posters) was held for publicity among the population. The children who lived in rural areas arrived with their guardians (generally the mothers) at Ouagadougou either by public transportation bus, or by the bus chartered by a local collaborator NGO. Upon their arrival, the children were accommodated in the premises of this NGO before being transported to CFS where the surgeries were performed. Before these surgeries, the patients were examined and blood analyses were performed. Then they were prioritized for surgery on the basis of the diagnosis, the operating risks (age, weight, health...) and expected functional results (e.g., capacity to re-educate the language). The medical files were completed; the consent forms filled up, and the photos of patients were taken.

For the surgical operations, the techniques used were those of Millard [6] for cleft lip repair and of Wardill [7] for cleft palate repair.

All the patients were examined again at the end of the campaign by the surgical team. After the departure of the Canadian team, the local team gave assurance for the follow-up of the patients.

2.5. Operational Definitions

As defined by Zandi and Heidari [8], we used the following abbreviations and criteria for cleft type classification: CL = isolated cleft lip (excludes isolated cleft palate and cleft lip with cleft palate), CP = isolated cleft palate (excludes isolated cleft lip and cleft lip with cleft palate), CLP = cleft lip with cleft palate associated (excludes CL, CP), CL +/- P = cleft lip with or without cleft palate (includes CL, CLP, excludes CP), and CL/P = cleft lip and/or cleft palate (includes CL, CP, CLP, no exclusions).

2.6. Data Collection and Analysis

We extracted data from the clinical files and from the anesthetics dossiers of the patients. A proforma allowed collecting demographic data (age, sex, residence), clinical presentation (type, side and seat of the cleft, associated anomalies), and surgery (type of surgery, peri- and postoperative outcome). Then data were entered and analyzed using Epi-info7TM software (Center for Diseases Control, Atlanta, GA, USA).

2.7. Ethical Considerations

Prior to operation, the surgical procedure was explained to the responsible (father or mother or a legal guardian) of the child. In case of acceptance, a consent form was co-signed by this responsible and a member of the surgical team. This study was approved by the Ethics Committee of CFS.

3. Results

Three campaigns of pediatric plastic surgery allowed to examine 185 children aged 0 - 14 years presenting with CL/P, of whom 54 were seen in 2007, 65 were seen in 2010 and 66 were seen in 2014.

Among the children under 15 years old, the frequency of CL/P was 185/6,499,211 demonstrating a rate of 0.03/1000 population.

Table 1 shows the distribution of children by place of residence.

On the whole, there were 100 boys (54%) and 85 girls (46%), demonstrating a sex ratio of 1.18:1. Among 182 cases of CL +/- P, there were 98 boys and 84 girls, giving a sex ratio of 1.17:1. Among the 3 cases of CP, there were 2 boys and 1 girl.

The average age of children was 2.4 ± 3.2 years [0 - 12 years]; there were 6 newborns (8.7%). **Table 2** shows the distribution of the children by age group.

Table 1. Distribution of children presenting with cleft lip and/or palate by residence, Burkina Faso in 2007, 2010, 2014.

Residence	Frequency	Percentage (%)
Province of Kadiogo	62	33.5
Province of Séno	31	16.8
Province of Kouritenga	21	11.3
Province of Sanmatenga	18	9.7
Province of Yagha	15	8.1
Province of Oudalan	10	5.4
Province of Sanguié	7	3.8
Province of Gourma	5	2.7
Province of Yatenga	5	2.7
Province of Houet	4	2.2
Province of Kéné Dougou	4	2.2
Province of Comoé	2	1.1
Republic of Côte d'Ivoire	1	0.5
Total	185	100.0

Table 2. Distribution of children presenting with cleft lip and/or palate by age group; Burkina Faso in 2007, 2010, 2014.

Age group (years)	Frequency	Percentage (%)
<1	69	37.3
1 - 4	79	42.7
≥5	37	20.0
Total	185	100.0

Of the total number of clefts, 182 (98.4%) were CL +/- P of which 92 (49.7%) were CLP and 90 (48.7%) were CL. Unilateral CL +/- P frequency was 138 (74.6%), and involved the left side in 49.2% of cases and the right side in 25.4% of cases. Bilateral CL +/- P were 41 (22.2%) cases, while there were 3 (1.6%) cases of median CL +/- P. In this group of CL +/- P, there were 92 CLP; 63/92 (68.5%) were unilateral CLP of which 68.2% concerned the left side and 31.8% concerned the right side; there were 29/92 (31.5%) bilateral CLP. Among CL +/- P, there were 90 CL, unilateral CL represented 83.4% of cases of which 64.0% concerned the left side and 36.0% the right side; bilateral CL represented 12/90 (13.3%) of cases. There were three CP (1.6%).

According to the type, laterality and severity, the most frequent cleft was Unilateral Left Cleft Lip (ULCL) (25.9%) followed by ULCL and Palate (ULCLP) (23.2%) and Bilateral Cleft Lip and Palate (BCLP) (15.7%).

Among boys, ULCLP (28.0%) and ULCL (20.0%) were the most frequent clefts, whereas among girls, ULCL (33.0%) and Unilateral Right Cleft Lip (URCL) (18.8%) were the most frequent clefts.

Table 3 shows the distribution of the children by sex, type, laterality and severity of clefts.

Of the 185 CL/Ps, 146 (78.9%) were isolated clefts while 39 (21.1%) were associated with other congenital malformations.

A total of 160 surgeries were performed in 150 of 185 (81.1%) children consisting of 135 cheilorrhaphy and 25 palatorrhaphy. The postoperative period was uneventful. For the 35 nonoperated children (18.9%), the main reason for nonoperation was the young age and/or the small weight of the child; no newborns were operated upon.

Table 4 shows the distribution of the nonoperated children according to the reason for nonoperation.

Table 3. Distribution of children by sex and type, laterality and severity of clefts, Burkina Faso in 2007, 2010, 2014.

Sex	Type, laterality and severity of the cleft							Total	
	BCL	MCL	BCLP	URCLP	ULCLP	URCL	ULCL		CP
Girls	4	3	11	7	15	16	28	1	85
	(4.7)	(3.5)	(13.0)	(8.2)	(17.6)	(18.8)	(33.0)	(1.2)	(100.0)
	[33.3]	[100.0]	[37.9]	[35.0]	[34.9]	[59.3]	[58.3]	[33.3]	[45.9]
Boys	8	0	18	13	28	11	20	2	100
	(8.0)	(0.0)	(18.0)	(13.0)	(28.0)	(11.0)	(20.0)	(2.0)	(100.0)
	[66.7]	[0.0]	[62.1]	[65.0]	[65.1]	[40.7]	[41.7]	[66.7]	[54.1]
Total	12	3	29	20	43	27	48	3	185
	(6.5)	(1.6)	(15.7)	(10.8)	(23.2)	(14.6)	(25.9)	(1.6)	(100.0)
	[100.0]	[100.0]	[100.0]	[100.0]	[100.0]	[100.0]	[100.0]	[100.0]	[100.0]

The percentages in parenthesis refer to the lines; those in brackets refer to columns. BCL: Bilateral Cleft Lip; MCL: Median CL+/-P; BCLP: Bilateral CL+/-P; URCLP: Unilateral Right CL+/-P; ULCLP: Unilateral Left CL+/-P; URCL: Unilateral Right CL; ULCL: Unilateral Left CL; CP: Cleft Palate.

Table 4. Distribution by reason of 35 nonoperated cleft lip and/or palate cases, Burkina Faso in 2007, 2010, 2014.

Reason	Frequency	Percentage (%)*
Too young/low weight	20	57.1
Polymalformation/complexity of the cleft [†]	6	17.1
Severe anemia	5	14.3
Current infection	5	14.3
Severe malnutrition	2	5.7
Miscellaneous (cleft too short, lack of time)	2	5.7

*Total exceeds 100% because a child could combine several reasons for not being operated; [†]Tessier #3, 4, 7, 11.

4. Discussion

4.1. About Clefts Epidemiology

In this study, a low frequency of CL/P in children aged 0 - 14 years in Burkina Faso was observed. Unfortunately, given the limitations of the study, the prevalence of clefts could not be ascertained. If a registry existed, it would not only fill this gap but it would also constitute a database that would improve the level of knowledge available on birth prevalence of CL/P and the associated international, geographical, ethnic and cultural variations [9].

According to the residence, the province of Kadiogo was the most represented by one-third of the patients. The reason is that the population of this province had more easy access to the information about the surgical campaigns, besides having less distance to travel when compared with patients who came from other provinces. But overall, there were many more children who came from other provinces which are very distant from the capital, Ouagadougou, and in which the populations for a great part are rural and poor. This trend is also observed in other developing countries where the specialists hardly meet except in referral hospitals [10]-[12]. For all these reasons, many children usually do not have access to the specialized health services and they remain home. To receive benefits from the NGOs and avail of the humanitarian surgery, they can arrive up to the capital and receive adequate care.

On average, we have seen the children after their second anniversary. This finding is similar to that of Diakité in Mali a nearby country sharing the same climatic, social, demographic and economic characteristics as Burkina Faso. This author reported 133 children presenting with CL/P who had an average age of 2.5 years. He also

found a distribution of the age at presentation which superimposed our results [13]. In developing countries, among the reasons that are put forward to explain the long time before consultation and/or surgery, we can quote the ignorance, the mystic and religious faiths, the social cultural heaviness, the inaccessibility, and the nonavailability of specialized health services; but also, and especially, the financial constraints, because most of the parents of the children presenting with clefts are poor [10] [14]. Where the children were seen earlier, it is the information of the populations on clefts and the free access of the treatment that explain the precocity of the diagnosis [15]. In Africa, the discovery of a CL/P is almost always a surprise for everybody including caregivers and parents, because the diagnosis is generally carried out in the delivery ward, with a malformed newborn. Contrary to the developed countries where prenatal diagnosis is possible [16], this is rarely (1%) made in Africa [13]. The causes of orofacial cleft underdiagnosis are, on one hand, the inaccessibility of the ultrasound facility for many pregnant women, and, on the other hand, the expertise of the technician, drawing attention to look for a cleft in the fetus is sometimes prompted only by a detail such as the presence of the anomaly in the mother [13]. In reality, in our context of a country with limited resources, these diagnostic limits do not, in particular, concern CL/P but generally all congenital malformations.

Our findings match the data of the literature relative to the predominance of males in overall CL/P and in CLP, in particular [2] [3] [13] [16]-[18]. However, other authors report a global ascendancy of girls compared with boys [19]. In our study, there were two boys and one girl presenting with CP, but the size of this sample was too small, as it did not allow discussing our results meaningfully with that of the other studies. According to certain authors, the difference of prevalence of clefts between the sexes would be due to the differences of sex hormones, speed of growth of tissues and organs, and mortality *in utero* between male and female fetuses [20]. However, considering that our statistics are based on patients who presented themselves for surgery rather than the number of children born with clefts, there may be a selection bias.

4.2. About Clefts Clinical Presentation

We noted ascendancy in CLP cases, followed by CL cases; CP cases were the least frequent. This distribution of the clefts in our study is in accordance with most of the data found in the literature [2] [3] [18] [21]. This order can, however, be upset and shows ascendancy in the CL [22] or CP cases [19] [23]. In this study, unilateral clefts were the most frequent compared with bilateral clefts, as well among CLP as CL; the left side was the most frequently affected, corresponding with the data in the literature [2] [16]-[18] [24]. According to the laterality and severity, ULCL were predominant followed by ULCLP, then BCLP, URCL, URCLP, BCL and, lastly, CP. Probably because of the different mode in recruitment of the patients, our findings are different from those of Franco *et al.* [12] in Brazil who found this sequence: ULCLP (27%)-CP (15.6%)-ULCL (13%)-URCL-URCLP-BCLP (each 7%)-BCL (2%). We found median clefts in the frequency of 1.6% closed to that of 1% found by Doray *et al.* [16], whereas, Aljohar *et al.* [23] found a threefold higher frequency of this type of cleft.

In this study, the frequency of 22.2% of bilateral clefts was higher than that found in the literature where it varies from less than 10% [12] [18] [25] to 17% - 19% [8] [13]. As previously mentioned, our data were based on patients who went for surgery encouraged in a certain way, and this could induce a selection bias. In addition, children with bilateral clefts present a most important surgical challenge, and it is possible that these children are not operated out of the humanitarian missions contrary to unilateral clefts which are frequently supported by local teams.

The frequency of 78.9% of isolated clefts found in this study was in compliance with the literature, which shows an ascendancy (more than 70%) of isolated CL/P [9] [17] [26]. However, in reality, this frequency of isolated clefts should be reviewed downward for the benefit of a more increased part of syndromic clefts because even seemingly isolated; 10% - 20% of the clefts are associated with other congenital malformations [9] [21].

4.3. About Care of Clefts

The majority of the children underwent surgery; cheilorrhaphy was frequently more practiced than palatorrhaphy which is in line with previous African studies [11] [15] [27]. As previously mentioned, the reasons for this may be the higher number of cleft lips than palates, as well as that cleft lip repair precedes palate repair [15]. In this study, all the interventions were successfully performed, and no peri- and postoperative negative outcomes were reported. This confirms that surgery of CL/P is satisfactory [15] [27] [28]; complications, such as desertion of suture, infection, keloid scars, narrowness of the mouth, are rarely reported [13], but oronasal fistulas can be

tackled as postoperative complications [15] [22]. Factors which influence the surgical outcome of clefts are in relation to the severity of the cleft, the race and particular characteristics of the patient, the experience and expertise of the surgeon, the technique used, the time of the intervention, and the postoperative care [15] [29] [30]. In Burkina Faso, the care of children with CL/P constitutes a challenge for the patients and their families. Indeed, the services of otorhinolaryngology or maxillofacial surgery are concentrated in the four tertiary hospitals located in the two main cities which are far from the rural areas where the majority of patients live. We also know that in these referral hospitals, the specialists are in insufficient numbers and are underequipped. Furthermore, the poorest populations cannot financially support the surgical operation in the absence of free access to the care, or the absence of an affordable and attractive system of cost sharing for them. So, many clefts cases escape treatment, and the humanitarian surgery constitutes an opportunity for the patients to benefit from appropriate care. In such a context, we understand that the priority is in the surgery, the other components of the care (orthodontics, otorhinolaryngology, speech pathology, psychology...) being relegated back. Besides, at the moment, the recommendation for integrated CL/P care by a multidisciplinary healthcare team is a gamble in the African countries [31]; so much so, the specialists are too insufficient and are underequipped.

4.4. About Study Limitations

This study, however, presents limits which lie in its hospital-based and retrospective characteristics. Nevertheless, the results can be of use as a basis to investigations of higher scale at the hospital level than at the national level, to better assess the morbidity of CL/P, to approach their etiologies and risk factors, and to implement a specific program which addresses CL/P in Burkina Faso.

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