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Cleft Lip and Palate Repairs in Togo, “Sourire de l’Espoir” Humanitarian Missions’ Experience: A Retrospective Study of 201 Consecutive Cases

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Abstract

Objective: To describe “Sourire de l’Espoir” humanitarian missions on cleft lip and palate (CLP) repair and how it shaped the future of CLP repair in Togo.

Design: Retrospective.

Patients: Clinical files of patients operated on for CLP during humanitarian missions from October 2008 to May 2013 in Lomé, the capital of Togo, were reviewed in a nine-month postoperative assessment from June to December 2020. Demographics, clinical, and therapeutic patterns were assessed. We also assessed the Zwisch method used for the training of local surgeons during the missions. Statistical analysis was performed with Epi Info (CDC version 7.1.3.3-2013).

Results: CLP repair was performed in 201 patients during 10 missions. Patients’ mean age was 7.05 (SD 9.22) years (range: 3 months-50 years); the sex ratio (male/female) was 1.05. Distribution of cases was as follows: Cleft lip (CL)

109/201 (54.23%); cleft palate (CP) 53/201 (26.23%), and CLP 39/201 (19.40%). The Tennison-Borde-Bedouelle-Malek technique was used for the repair of CL in 119/148 patients (80.41%); the Onizuka technique (modified Millard technique) for the revision surgery of CL in 19 patients previously operated in other settings. The Kriens and Sommerlad palatoplasty technique was performed for primary and revision surgeries for all CP in 89/92 (96.73%) patients. The postoperative course was uneventful in 199/201 (99.00%) patients. One patient presented with an infection on the lip that healed with local wound care, and two patients with palatal fistula were treated by revision surgery. Two patients died in post-operative period.

Three surgeons of the local team were trained as per the Zwisch method: in the first two missions, show and tell (60/201 cases), the third and fourth missions, smart help step (48/201 cases); from the seventh mission onwards, passive assistance (51/201 cases) by the local surgeons with no help.

Conclusion: The “Sourire de l’Espoir” humanitarian missions provided primary and revision surgery to children and adults with CLP while three local surgeons were trained to build a local capacity.

Keywords: Cleft lip and palate, Humanitarian missions, Zwisch method, Togo.

Introduction

Cleft lip (CL) with or without cleft palate (CP) is a frequent congenital malformation^{[1][2]}. The incidence is reported to be from 0.4% to 1.49% in African countries^{[3][4][5]}. Cleft lip and palate (CLP) is not only a physical deformity; it represents a psychological and social burden on affected children and their families^{[6][7]}. In high-income countries (HICs), where diagnosis is made in the prenatal assessment, children are treated from the perinatal period to the end of their growth^[8]. In low-middle-income countries (LMICs), many children with CLP grow up with the malformation due to precarious common beliefs, financial conditions, limited access to existing low-equipped hospitals, and a scarcity of trained surgeons. For decades, non-governmental organizations (NGOs) provided humanitarian surgical missions to fill the gap in the management of CLP cases around Africa^{[9][10]}. In Togo, various humanitarian missions from different western countries offered sporadic surgical care to patients living with cleft lip and palate (CLP). The treatment was usually limited to the primary surgery. Local surgeons were barely associated with these missions. Consequently, in the post-mission period, no follow-up and no assessment of the results were carried out. There was no plan to build up a local capacity or to improve the long-term standard of CLP care in the country. In October 2008, a retired French pediatric plastic surgeon (Bernard Pavy) brought “Sourire de l’espoir” (smile of hope) missions to Togo. These missions were sponsored by “Chaîne de l’espoir” (Chain of Hope), a French NGO with branches in French-speaking African countries. Besides operating on children with CLP, the mission aimed to train local surgeons and create a local team capable of performing the primary surgery for patients living with CLP. Therefore, three general surgeons with some knowledge in plastic surgery procedures were identified to join the French team with the goal of being trained for the primary surgery of CLP.

This retrospective study aimed to describe the missions, how the local team was built, and the socio-demographic, clinical, therapeutic, and outcome aspects of the patients with CLP who were operated on.

Materials and Method

This retrospective study was based on the registers of the “Chaîne de l’Espoir” NGO and paper-based clinical records of the patients operated on for CLP from October 2008 to May 2013 during the “Sourire de l’Espoir” mission in Lome, the capital of Togo.

Context and Planning of the Missions

From October 2008 to May 2013, « Chaîne de l’Espoir » organized biannual missions in Togo. Professor Bernard Pavy served as the mission leader and trainer. The local team of “Chaîne de l’Espoir” conducted communication campaigns through local TV, radio, and schools. Non-medical staff within the NGO registered all patients with CLP. A patient list was compiled, and patients were scheduled for clinics on the first day of the mission. Both the local team and the French team conducted the initial screening of the preselected patients. Preoperative investigations included a Complete Blood Count and ABO blood group check. To be eligible for surgery, children needed to have a hemoglobin level of at least 10g/dl, weigh at least 4.5 kg, and have no significant findings that could compromise anesthesia. Adults with no other comorbidities were also eligible for surgery. The final surgical schedule was determined based on clinical presentation (anatomical type of cleft), age (more than 10 weeks old for clefts of the primary palate and more than six months for clefts of the secondary palate), and existing comorbidities (associated congenital or acquired comorbidity). A ten-day operative schedule was created based on the identified cases. Screening and surgeries were done in the Sylvanus Olympio Teaching Hospital but for some logistic reasons, operations during three missions were done in a private hospital (Martin Luther King) in Lome. All surgeries were done for free for all patients; local and foreign teams received no payment for the care.

Socio-demographic, Clinical, and Outcome Parameters

We extracted the following parameters from the clinical files and NGO registers: socio-demographic information including age (children under 15 years, and adults were above 15 years), gender, place of residence, and socio-economic conditions of the patients.

We considered patients or children living in favorable socio-economic conditions if they had parents with health insurance, resided in their own house, or held a high-income job. Patients with unfavorable economic conditions were those with no medical insurance and income lower than the minimum wage in Togo.

Clinical assessment included the clinical presentation of CLP based on the Kernahan and Stark’s classification^[11], as used in the clinical files. This classification describes CLP as clefts of the primary palate (equivalent to cleft lip or CL), clefts of the secondary palate only (cleft palate or CP), and clefts of both the primary and secondary palates (cleft lip and

palate or CLP).

Peri and post operative outcomes included peri operative complications mainly recorded by anesthesiologists. In the postoperative course, we assessed the length of hospital stay, and other complications as infection, wound break down and other non planned events. In the follow up, we searched for complications as fistula, phonation deficiency, asymmetry of the lips. Patients satisfaction was also recorded.

Surgical Management During the Missions

All procedures, both in adults and children, were performed under general anesthesia with oro-tracheal intubation.

The Tension-Borde-Bedouelle-Malek (T repair) ^{[12][13][14]} was used in the repair of CL; the Onizuka technique (O repair), a modified Millard technique (M repair) ^[15], was practiced in revision surgery on the lips. The Millard technique ^[16] was used for bilateral CL, and the Kriens and Sommerlad technique (S repair) ^{[14][17][18][19]} for CP repair. Patients with CLP were operated on in two stages when they were under six months age; the procedure was given in one stage when patients were older than six months. The training of local surgeons followed the Zwisch method ^[20].

Results were assessed as good when both the surgeon and the patients were satisfied, and no further revision surgery was required. Results were considered poor when a revision surgery was needed or recommended. In patients with secondary CP, results were described as good when there was no significant fistula, and phonation was clear enough to enable a decent standard of living. In patients with CL, good results entailed no conspicuous asymmetry or scarring on the lips.

Inclusion and Exclusion Criteria

To be included, files had to be of patients operated on for CLP during the missions, carrying enough information to allow analysis of parameters described above. Missing or incomplete files of patients operated on for CLP, files of patients operated on for other diseases during the missions, and files of patients not operated on were excluded.

Statistical Analysis

We used CDC (Centers for Disease Control) Epi Info version 7.1.3.3-2013 and Microsoft Excel 2013 for statistical analysis. We reported continuous variables as numbers and percentages. The Chi-square degree of freedom was provided if necessary. Statistical significance was indicated by a p-value < 0.05.

Ethical Considerations

The data for this study were collected following the Declaration of Helsinki^[21]. The local ethical committee of the Sylvanus Olympio Teaching Hospital approved this study. The patients or their families provided written informed consent for the procedure and data collection before the surgery.

Results

Socio-demographic Parameters

Ten humanitarian missions took place from October 2008 to May 2013. A total of 271 preregistered patients were screened during the consultations, out of which 220/271 patients with CLP underwent surgery. Among the other other 51 patients, 38/271 underwent surgery for other diseases, mainly pediatric plastic surgery cases such as congenital hand abnormalities, mandibular fistula post-osteomyelitis, and cases of vascular abnormalities. A total of four patients received primary repair of lateral clefts (Tessier 3, 4, and 7). Two CLP patients were unfit for anesthesia and 7 CLP patients who did not find a place in the schedule of the last mission and were operated on outside the missions. Among patients operated on for CLP, the clinical files of 19 patients were lost or lacked consistent information to allow inclusion in the study.

A total of 201/220 (91.36%) files was the final number included for the analysis in this study. During each mission, an average of 22 surgical procedures (range: 8 to 34) were performed. Table 1 displays the number of patients operated on in each mission from October 2008 to May 2013.

Table 1. Distribution of patients during ten Smile missions in Togo

Years and months of missions	Number
2008	
October	34
2009	
March	26
November	27
2010	
May	21
November	18
2011	
April	24
November	10
2012	
May	14
November	8
2013	
May	19
Total	201

Children accounted for 170/201 (84.57%), while adults made up 31/201 (15.43%). The average age of patients was 7.05 (SD 9.22) years, with a range from three months to 50 years. The sex (male/female) ratio was 1.05.

Among pediatric patients, 82/201 (40.80%) were above five years old (school age in Togo). Inside this group a total of 28/82 (34.15%) were not attending school, while 54/82 (65.85%) were attending school (financial and social reasons were reported).

Regarding social life, 169/201 (84.08%) patients reported being victims of mockery. A 2-year-old child with a cleft palate was abandoned on a farm by his parents pretending that the child was a source of curse. Socio-economic conditions were unfavorable for 185/201 (92.04%) patients, while 16/201 (7.96%) patients were considered in favorable conditions.

Concerning the place of residence, 111/201 (55.22%) patients lived in Lome, the capital, whereas 88/201 (43.78%) came from other regions of the country; 2/201 (1.00%) patients came from neighboring countries.

Clinical Data

Concerning the anatomic-clinical form, 109/201 patients (54.23%) presented a CL, and 53/201 patients (26.37%) had a CP. A total of 39/201 patients (19.40%) showed a CLP.

CL was left-sided in 78/148 patients (52.70%), right-sided in 57/148 patients (38.52%), and bilateral in 13/148 patients (8.78%). Table 2 summarizes the clinical presentation of CL cases. The malformation was diagnosed at birth in 184/201 patients (91.54%). Inside this group only two patients presented with CP. In 17/201 (8.46%) patients all presenting with CP, the diagnosis was made during the growth period when the patients have started acquiring language.

Table 2. Clinical presentations of CLP in the 148 novel cases

	Right side (n, %)	Left side (n, %)	Bilateral (n, %)	Total (n, %)
Cleft of primary palate	44(40.37)	58(53.21)	7(6.42)	109(100.00)
Cleft of primary and secondary palate	13(33.33)	20(51.29)	6(15.38)	39(100.00)
Total	57(38.52)	78(52.70)	13(8.78)	148(100.00)

Chi-square=0,81; ddl=2; p=0.8

Associated malformations were seen in 10/201 patients (4.98%) that encompassed palpebral ptosis (two patients), Pierre Robin syndrome (two patients), Van der Woude syndrome (one patient), hydrocephalus (one patient), ear malformations (two patients), unilateral clubfoot with forearm agenesis and ventricular septal defect (one patient), and bilateral clubfeet (one patient). Among patients with associated malformations, only one with Van der Woude syndrome was operated on for cleft repair.

Surgical Procedures and Postoperative Outcomes During the Missions

The Tennison-Borde-Bedouelle-Malek technique was performed in 119/148 (80.41%) patients, and the Onizuka procedure was used in 19/148 (12.84%) patients for revision cheiloplasty. The Millard technique was applied to bilateral 5/148 (3.38%) CL cases. The Kriens and Sommerlad procedure was used for 89 CP (44,28%) primary repair. A total of 11 had revision CP repair; among them, 9 patients were previously operated on in missions from other teams, and two were complications of the surgeries in the present mission surgery. Techniques used were the Kriens and Sommerlad procedure in 10 patients and a tongue flap in one patient.

In 39 patients with CLP, 24/39 patients had a one-step repair, and a two-step approach was used in 7/39 patients. Table 3 reports the distribution of the surgical techniques according to the cleft type. Figure 1 shows the preoperative and postoperative pictures of two patients operated on by the mission leader. The postoperative course was uneventful in 199/201 patients (99.00%).



Figure 1. Before and after surgery of a-b: 10-year-old boy presenting bilateral cleft lip and c-d: cleft palate operated by the trainer.

Table 3. Surgical repair technique used by type of cleft

Surgical repair	Cleft lip	Cleft lip and palate	Cleft palate	Total
T repair	92	1	0	93
T+S repair	0	19	0	19
T+S repair in two steps	0	7	0	7
O repair	15	1	0	16
O+S repair	0	3	0	3
M repair	2	1	0	3
M+S repair	0	2	0	2
S repair	0	5	53	58
Total	109	39	53	201

O Repair = Onizuka repair; O+S Repair = Onizuka + Kriens and Sommerlad repair; S Repair = Kriens and Sommerlad repair; M Repair = Millard repair; T+S Repair = Tennison-Borde-Bedouelle-Malek + Kriens and Sommerlad repair; T Repair = Tennison-Borde-Bedouelle-Malek repair; M+S Repair = Millard + Kriens and Sommerlad repair.

A total of 2 patients aged 2 and 4 year-old died in the immediate postoperative period, both after primary cheiloplasty. These deaths, as reported in the medical files, were attributed to laryngospasm that occurred after the patients were transferred to the general recovery room. The average duration of hospital stay was 3.13 (SD 1.42) days, with a range of 2-8 days. One patient operated on for a CL developed a superficial infection that healed with local wound care and oral antibiotics.

In the follow up, 2/82 (2.44%) patients were diagnosed with oronasal fistula (included in the cases of revision CP repair). Two patients with velo-palatal insufficiency were treated with surgical pharyngoplasty.

A number of a three local surgeons were trained according to the Zwisch model. The first 2 missions were “show and tell”; 60/201 patients were operated on by the mission leader. The third and fourth missions were the “smart help” step when 48/201 patients were operated on. The fifth and the sixth missions were the phases of “dumb help” in which 42 patients were operated on. From the seventh mission onwards, 51/201 patients received primary surgery for CLP performed by the local surgeons with no help. The mission leader made rounds to inspect the procedures. Two patients operated on by the local surgeons are shown in Figure 2.



Figure 2. Before and after surgery of a-b: Cleft lip on a 5-year-old girl, and c-d: before and after repair of a posterior cleft palate in a 1-year-old girl operated in the tenth mission by the local surgeons.

Discussion

Most humanitarian missions in African countries and other LMICs miss the opportunity to train local surgeons. The role and goals of these missions typically do not include such training. One distinctive aspect of the humanitarian missions organized by Bernard Pavy and his team was the inclusion of a training program for a local team while providing surgical care to patients. This approach offers the advantage of imparting basic technical skills to local surgeons and operative teams, enabling them to continue treating malformations after the missions conclude for financial or other reasons. The benefits of this approach have been reported in other countries beyond Africa. Building local teams contributes to improving the standard of care in a given area [9][22][23]. The standards of care for diseases like CLP are low in Africa. In this study, the average age was 7.05 years, ranging from 3 months to 50 years. In other African countries with similar levels of care, the average age ranged from 3.9 to 9.34 years [24][25]. Adult patients continue to live with CLP that is ideally treated in early childhood. Several factors contributed to the delayed surgical care, including low awareness level among the population, especially those in remote areas, delayed diagnosis in patients with CP, and available trained surgeons to take care of them. Sometimes, a poor anesthesia standard contributes to delaying the first surgery [24][25]. Growing up with the malformation has a negative impact on the social life of children. School-age patients may avoid attending classes to avoid mockery and various forms of stigmatization, as observed in this study.

Despite the prevalence of female gender being reported in the literature [23], we observed no significant difference between males and females in our study. However, the specific patterns associated with gender may be more elucidated in future studies for the clinical outcomes. We detected more patients with isolated CL and CP than patients with associated CLP. This presentation is opposed to what was reported in the literature where CLP is preponderant to isolated CL and CP [25][26][27][28]. As a registry of malformations does not yet exist in our country, it is hard to claim that this profile represents the entire patients' population. Future studies may provide a better understanding of the clinical patterns of clefts in Togo.

Concerning the treatment, an array of operative techniques is in use for the primary surgical repair of CLP around the world. In this study, we mainly used the Tennison Borde-Bedouelle-Malek method for unilateral lip repair. This technique, based on geometrical measurement and drawing, is easy to learn and reproducible by non-experienced surgeons. Although the Millard technique seems to be the most frequently used nowadays, it requires a longer learning curve and more surgical skills [9][10][25]. A careful dissection of the muscular plane added to repair layer by layer make all these techniques opposable in term of functional and results. Nevertheless, a randomized study may be welcome to compare the two methods in the hands of teams who have experience in both of them. There are numerous operative schemes that exist among teams who take care of CLP. Some surgeons may consider closing the lip as soon as possible. For most of the teams, primary palatoplasty should be performed before the patient starts learning a language [29][30]. However, safety guidelines have been issued even in HICs aiming at reducing peri operative complications. One of these guidelines follows the rule of 10's: the surgery is postponed until the child weighs 10 pounds and is 10 weeks old or more [31][32][33]. Palate repair is best performed between 10-14 months [29][30]. A hemoglobin concentration of 10 g/dl increases the safety of anesthesia in children operated for CLP. In our setting this scheme was not a big deal as the schedule gave priority to older patients that were numerous thus, not having place for patients that were at the border of these criteria; the closure of the palate becomes an emergency in such patients using a one-step approach in case of CLP. Old children and adults

were all able to afford a long anesthesia time. Possible complications are laryngospasm occurring intra or post-operatively. In this study, we have recorded two deaths among 201 patients operated on due to laryngospasm. This complication is not always lethal, especially when diagnosed intra-operatively but it carries substantial consequences for the family and care team [26][34][35]. In Nigeria, a team has reported one death among 500 CLP operations owing to laryngospasm [26]. During the Smile Train missions in India, laryngospasm was the most common (40.9%) complication but resulted in no deaths. In our case, the two children died in the recovery room may have escaped the vigilance of the team in this room that had to care for all the patients from all team. In Togo, a 2005 study reported a peri operative mortality rate of 2.05% for all surgical procedures. Unfortunately, 93% of them were reported as avoidable [36]. Recently, the improvement of ICU and anesthesia standards reduced this mortality to 1.03% [37]. In other LMICs, a prevalence of 19 to 51 deaths per 10,000 anesthetics has been reported, and in HICs, 20 per 10,000 [38] was the rate. Other common peri operative complications that can occur during surgery for CLP, such as excessive bleeding and other anesthesia complications [39], were not seen in our study. In a later postoperative course, we have seen a surgical site infection in only one patient. Oro-nasal fistula appeared in two patients; thus, the rate of this complication was 2.44%. This number is lower than what was reported by other teams (5-29%) [9][40][41]. The careful dissection and an appropriate muscle repair during the intravelar veloplasty, the long experience of the mission leader that was at the end his career, may have accounted for the low rate of this complication. Additionally, most patients were regularly reviewed during missions and educated about postoperative care and exercises to improve phonation as speech therapists were not available in the area in the time of these missions.

Biases and *Limitations*

The limitations of the present study are mainly related to its retrospective design. This study did not intend to provide the epidemiological data of CLP in Togo, even though it regards a representative sample of the population of patients with CLP in Togo. The classification of CLP and semantics used to describe the clinical presentation and postoperative complications were those used by the mission leader.

Conclusions

The experience developed throughout the ten “Sourire de l’Espoir” missions highlighted remarkable aspects of CLP in Togo. The missions provided good surgical care to patients of different ages, including adult patients often living in unfavorable socioeconomic conditions. Three Togolese surgeons acquired technical skills about Tennison-Borde-Bedouelle-Malek and the Kriens-Sommerlad veloplasty. This training represented one of the most valuable outputs of the missions of Togo since the trainees who are empowered to address basic local primary needs of CLP may gain experience and get into more complicated cases as the cosmetic demands of the patients increases for a better quality of life.

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