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Abstract

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

Design: Retrospective.

Patients: Clinical files of patients operated for CLP during humanitarian missions from October 2008 to May 2013 in Y, the capital of X, 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 was 1.05. Distribution of cases was as follows: Cleft lip (CL) 109/201
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. Two patients died of laryngospasm in the immediate perioperative course. 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. 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 3 local surgeons were trained to deal with primary cases.

**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. The incidence is reported to be from 0.4% to 1.49% in African countries. Cleft lip and palate (CLP) is not only a physical deformity; it represents a psychological and social burden on affected children and their families. 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.

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. 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 local capacity or to improve the long-term standard of CLP care in the country. In October 2008, a retired French pediatric plastic surgeon brought “Sourire de l’espoir” (smile of hope) missions to X. These missions were sponsored by “Chaine 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 training in plastic surgery 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 “Chaine 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 Y, the capital of X.

Context and Planning of the Missions

From October 2008 to May 2013, XX organized biannual missions in Y. Professor XX served as the mission leader and trainer. The local team of “Chaine de l’Espoir” conducted communication campaigns to the population through local TV, radio, and schools. Non-medical staff within the NGO registered all patients with CLP. A patient list was compiled, and all patients were scheduled for clinics on the first day of the mission. Both the local team and the French team of Professor XX conducted the initial screening of the preselected patients. Preoperative investigations included a Complete Blood Count and ABO blood group analysis. 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. Anesthesiologists assessed all patients scheduled 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.

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 over 15 years), sex, 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 without medical insurance and with income lower than the minimum wage in Togo, given that no social security-supported hospital treatment was available during that specific period.

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 postoperative outcomes included peri-operative complications mainly recorded by anesthesiologists. In the postoperative course, we assessed the infection rate, as well as complications such as fistula and duration of hospital stays.

**Surgical Management During the Missions**

All procedures, both in adults and children, were performed under general anesthesia with oro-tracheal intubation. The Tennison-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 repairing clefts of the palate. Patients with CLP were operated on in two stages when they were under six months old and in a one-stage procedure when they 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. These rating criteria were used in the patients’ files by the operating team.

**Inclusion and Exclusion Criteria**

The study included clinical files containing all the assessed parameters and excluded clinical files with missing or incomplete assessed parameters.

**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 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 March 2013. A total of 271 preregistered patients were screened during the consultations, out of which 220/271 patients with CLP underwent surgery. During each mission, an average of 22 surgical procedures (range: 8 to 34) were performed. Additionally, 38/271 patients underwent surgery for other diseases, mainly pediatric surgery cases such as congenital hand abnormalities, mandibular fistula post-osteomyelitis, and cases of vascular abnormalities. Four patients received primary repair for lateral clefts (Tessier numbers 3, 4, and 7). Two CLP patients unfit for anesthesia and 7 CLP patients in the last mission could not receive surgery during the missions. Among patients operated on for CLP, the clinical files of 19 patients were either unavailable or lacked sufficient information to meet the inclusion criteria for the study. A total of 201/220 (91.36%) files were included for the analysis during the mission period. Table 1 displays the number of patients operated on in each mission from March 2008 to May 2013.

Table 1.
Distribution of patients during ten Smile missions in X
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 3 months to 50 years. Among the patients, 82/201 (40.80%) were above 5 years old (school age in $X$), and out of these, 28/82 (34.15%) were not attending primary school, while 54/82 (65.85%) were attending school. The sex ratio was 1.05.

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 their parents. Socio-economic conditions were unfavorable for 185/201 (92.04%) patients, while 16/201 (7.96%) patients lived in favorable conditions.

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

**Clinical Data**

The malformation was diagnosed at birth in 184/201 patients (91.54%); two of these patients presented with CP. In 17/201 (8.46%) patients with CP, the diagnosis was made during the growth period when the patients had started acquiring language. Concerning the anatomic-clinical form, 109/201 patients (54.23%) presented a cleft of the primary palate equivalent to CL, 53/201 patients (26.37%) had a cleft of the secondary palate marked as G2 equivalent to CP, and
39/201 patients (19.40%) showed a cleft of both the primary and secondary palate equivalent to 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. There were associated malformations in 10/201 (4.98%): palpebral ptosis (2 patients), Pierre Robin syndrome (2 patients), Van der Woude syndrome (1 patient), hydrocephalus (1 patient), ear malformations (2 patients), unilateral clubfoot with forearm agenesis and ventricular septal defect (1 patient), and bilateral clubfeet (1 patient). Among patients with associated malformations, only one with Van der Woude syndrome was treated for the associated malformation during the primary cleft repair.

### Table 2. Anatomic-clinical types and forms of the 148 clefts lip

<table>
<thead>
<tr>
<th></th>
<th>Right side</th>
<th>Left side</th>
<th>Bilateral</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cleft of primary palate</td>
<td>44(40.37)</td>
<td>58(53.21)</td>
<td>7(6.42)</td>
<td>109(100.00)</td>
</tr>
<tr>
<td>Cleft of primary and secondary palate</td>
<td>13(33.33)</td>
<td>20(51.29)</td>
<td>6(15.38)</td>
<td>39(100.00)</td>
</tr>
<tr>
<td>Total</td>
<td>57(38.52)</td>
<td>78(52.70)</td>
<td>13(8.78)</td>
<td>148(100.00)</td>
</tr>
</tbody>
</table>

Chi-square=0.81; ddl=2; p=0.8

**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 cheiloplasty. The Millard technique was applied to bilateral clefts in 5/148 (3.38%) cases. The Kriens and Sommerlad procedure was the palatoplasty technique used in all 53 patients with CP and 36/39 patients with CLP. Three patients with CLP could not have palate repair during the missions. Eleven patients had a revision surgery of the palate. Among these 11 patients, 9 patients were previously operated on in other missions. The other 2 patients experienced complications from the “Sourire de l’Espoir” missions in X. 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%). We recorded two deaths that occurred 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 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. During the missions, 2/82 (2.44%) patients were diagnosed with oronasal fistula. The complication was treated using a revision intravelar veloplasty for one patient and a tongue-pedicled flap for the other. Two patients with velopalatal insufficiency were treated with pharyngoplasty.
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

<table>
<thead>
<tr>
<th>Surgical repair</th>
<th>Cleft lip</th>
<th>Cleft lip and palate</th>
<th>Cleft palate</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>T repair</td>
<td>92</td>
<td>1</td>
<td>0</td>
<td>93</td>
</tr>
<tr>
<td>T+S repair</td>
<td>0</td>
<td>19</td>
<td>0</td>
<td>19</td>
</tr>
<tr>
<td>T+S repair in two steps</td>
<td>0</td>
<td>7</td>
<td>0</td>
<td>7</td>
</tr>
<tr>
<td>O repair</td>
<td>15</td>
<td>1</td>
<td>0</td>
<td>16</td>
</tr>
<tr>
<td>O+S repair</td>
<td>0</td>
<td>3</td>
<td>0</td>
<td>3</td>
</tr>
<tr>
<td>M repair</td>
<td>2</td>
<td>1</td>
<td>0</td>
<td>3</td>
</tr>
<tr>
<td>M+S repair</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>2</td>
</tr>
<tr>
<td>S repair</td>
<td>0</td>
<td>5</td>
<td>53</td>
<td>58</td>
</tr>
<tr>
<td>Total</td>
<td>109</td>
<td>39</td>
<td>53</td>
<td>201</td>
</tr>
</tbody>
</table>

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

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, with 48/201 patients treated. The fifth and the sixth missions were the phases of “dumb help”. Forty-two patients were operated on during this phase. 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.
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 XX and their 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 \cite{24,25}. Adult patients continue to live with the malformation that is ideally treated in early childhood. Several factors contribute to the delayed surgical care, including low awareness levels among the population, particularly those in remote areas, and delayed diagnosis in patients with CP. Sometimes, a poor anesthesia standard contributes to delaying the first surgery \cite{24,25}. Growing up with the malformation has a negative impact on the social lives 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 \cite{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 \cite{25,26,27,28}. As a registry of malformations does not 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 X.

Concerning the treatment, an array of operative techniques is in use for the primary surgical repair of CLP around the world. XX and his teammates 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 training curve and more surgical skills \cite{9,10,25}. A careful dissection of the muscular plane added to repair layer by layer in all these methods improved the functional and cosmetic results on the lip. 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 \cite{29,30}. However, safety guidelines have been issued even in HICs aiming at reducing perioperative 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 \cite{31,32,33}. Palate repair is best performed between 10-14 months \cite{29,30}. A hemoglobin concentration of 10 g/dl increases the safety of anesthesia in children operated for CLP. When children are diagnosed late, they become old enough to afford a long anesthesia time. The closure of the palate becomes an emergency in such patients; a one-step approach is the rule. Some patients treated during missions of XX in X shared this situation. Despite following all these guidelines, complications of the surgical repair of CLP are numerous. The deadliest are laryngospasm. In this study, we have recorded two deaths among 201 patients operated on due to laryngospasm. Although laryngospasm is not always lethal, it carries substantial consequences for the family and care team \cite{26,34,35}. In Nigeria, a team has reported one death among 500 CLP operations owing to laryngospasm \cite{26}. During the Smile Train missions in India, laryngospasm was the most common (40.9%) complication but resulted in no deaths. During humanitarian missions in limited resource settings, anesthesiologists should be involved not only in the preoperative investigation but also in the postoperative phase. In X, a 2005 study reported a perioperative mortality rate of 2.05% for all surgical procedures. Unfortunately, 93% of them were reported as avoidable \cite{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 perioperative complications that can occur during surgery for CLP, such as excessive bleeding and other anesthesia complications [39], were not observed in our study. In a later postoperative course, we have seen a surgical site infection in only one patient. Oronasal 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, notwithstanding the expertise and long experience of the mission leader (XX), may have accounted for this low rate of this complication. Additionally, most patients were regularly reviewed during missions and educated about postoperative care and exercises to improve phonation.

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 X, even though it regards a representative sample of the population of patients with CLP in X. 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 X. 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 XX since the trainees are empowered to address the local primary needs of CLP. Local surgeons should be aware of the increasing cosmetic demands of the patients for a better quality of life. They should also prepare for new challenges in the nowadays well-codified schedule of the treatment of CLP.

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