Ten Years of Management of Esophageal Atresia in Benin: State of the Art, Experiences and Needs for Families of Children with Stoma

Mahunakpon Vihotogbe Leon Samuel Boris Gogan[1], Amoussou Sedjro Clotaire Romeo Houegban[1,*], Codjo Serge Metchihoungbe[2], Naziha Khen-Dunlop[3], Modoukpe Grace Immaculee Sergine Dossou[2], Aldrico Pautin Covi[2], Michel Armand Fiogbe[2] and Antoine Seraphin Gbenou[1]

[1]*Department of Pediatric Surgery, Mother and Child Teaching Hospital Lagune, University of Abomey-Calavi, Benin*

[2]*Department of Pediatric Surgery, National University Hospital Center "Hubert Koutoukou Maga", University of Abomey-Calavi, Benin*

[3]*Department of Pediatric Visceral Surgery - Hôpital Necker-Enfants malades - AP-HP, Paris, France*

Abstract: *Background/Purpose:* Esophageal atresia (EA) is a congenital defect in the thoracic esophagus associated or not with a tracheoesophageal fistula (TEF). It is associated with high mortality in low-income countries such as Burkina Faso, Madagascar and Ghana. The purpose of this study was to present the recent management of newborns with EA/TEF in Benin and to identify the needs of families whose children have survived.

Method: Over a period of 10 years, 54 newborns with EA/TEF operated on in the 2 largest university hospitals in Benin were included. Two groups were identified. Group A (n=33) included newborns in whom a primary repair had been performed. Group B (n=21) involved newborns who underwent staged repair. Staged repair involved cervical oesophagostomy + gastrostomy (n= 7) or upper oesophageal pouch suction + gastrostomy (n=2) or cervical esophagostomy + stoma of the lower esophageal pouch (n=12). Ten parents from group B were interviewed for the needs survey. The significance level was defined as $p < 0.05$.

Results: There were 31 male newborns. The median for gestational age was 37 gestational weeks (range: 35 - 38). Polyhydramnios was observed in 4 cases. The mean birth weight was 2365 g. (range: 1000 g - 3500 g). The mean age at diagnosis was 3.48 days (range: 24 hours - 19 days). Surgery was performed at day 5 on average with extremes from day 2 to day 17. The surgery lasted an average of 2h12 min and the overall mortality rate was 74.07%. Mortality rate in group A was 100% and 33.33% in group B. There was a strong statistically significant difference in survival between the two groups. ($p=19.10^{-8}$). Sepsis was the most common cause of death. Stress, fear and anxiety of losing the child were the difficulties frequently encountered at bedtime and every day. Four parents had to stop their activities to be available to take care of their child. All the parents had expressed the need for psychological support.

Conclusion: Pending an improvement of the technical platform, staged repair remains an essential option for the survival of patients. The creation of a framework for discussion with families authorizing social actions and quality medical support is desired.

Keywords: Esophageal atresia, Staged repair, Mortality, Family needs, Low-income countries.

INTRODUCTION

Esophageal atresia/tracheoesophageal fistula (EA/TEF) is a condition resulting from abnormal development before birth of the esophagus. During early development, the esophagus and trachea begin as a single tube that normally divides into the two adjacent passages between four and eight weeks after conception. If this separation does not occur properly, EA/TEF is the result. In EA, the upper esophagus does not connect (atresia) to the lower esophagus and stomach. Almost 90 percent of babies born with esophageal atresia also have a tracheoesophageal fistula (TEF), in which the esophagus and the trachea are abnormally connected, allowing fluids from the esophagus to get into the airways and interfere with breathing. A small number of infants have only one of these abnormalities. Its pathogenesis remains poorly understood to date [1]. Its overall incidence ranges from 1/2500 to 1/4500 births worldwide [2-5]. In Africa, its prevalence remains underestimated due to the lack of malformation surveillance registries in the different countries but also due to the lack of awareness of diagnosis, chronic under-equipment of health facilities and poor quality of follow-up of pregnant women [6, 7]. The progression is inexorably towards death if nothing

*Address correspondence to this author at the Department of pediatric surgery, Mother and Child Teaching Hospital Lagune, University of Abomey-Calavi, Benin. Home address: 11 rue Saint Marc, 67000 Strasbourg, France;
Tel: +33758410328; E-mail: houegbanromeo@gmail.com

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is done immediately after birth. The first cases of EA that survived after surgical management were reported by Leven and Ladd in 1939 [8, 9]. By this time, the repair approach was in debate with the possibility of a multistage repair using a subsequent esophageal replacement. Cameron Haight [10] performed the first successful one-stage repair of EA in 1941 using an extrapleural approach in a 12-day-old female infant. From this first experience came other successful repairs with a tremendous improvement in the survival rate. Waterston *et al*. [11] reported a 57% survival rate in 113 infants treated in the early 1950s. For Spitz [12], mortality was under 15% in the mid-1980s. Advances in surgical techniques and peri-operative neonatal intensive care have significantly improved the survival of patients with EA in recent decades. The main causes of poor prognosis remain chromosomal aberrations and complex cardiac malformations [13-15]. In developing countries, mortality remains high. It is 76 to 90% in sub-Saharan Africa [6, 16, 17] and 40% in Mahgreb [18]. Its management is still limited by the precariousness of technical platforms and remains a great anesthetic challenge per and postoperatively.

As in these countries, in Benin mortality related to this condition is high and approaching 100% when a primary repair is performed. In this context, the choice of a staged repair with subsequent esophageal replacement is made, pending subsequent improvement of the technical platform. With the survival of the first cases thanks to this therapeutic option, there is a need to consider an organization that allows a better support of the parents and these children, as in many European countries, while working on requirements for a management in accordance with the international standards. The purpose of this study was to present the recent management of cases of EA in Benin. On the other hand, based on the parents' experience, the authors had identified the needs of these families whose children survived and were carriers of a cervical esophagostomy and gastrostomy.

PATIENTS AND METHOD

Over a 10-year period (July 1, 2012 to June 30, 2022), 75 infants born with EA were treated at the Centre Hospitalier de la Mère et de l'Enfant Lagune (CHUMEL) in Cotonou and at the Centre National Hospitalier et Universitaire Hubert Koutougou Maga in Cotonou (CNHU-HKM). These are the two largest hospitals and universities in Benin. Of these infants, 56 had surgical management by a senior lecturer or a senior pediatric visceral surgeon and were included in this study. Two groups were identified. Group A included infants managed by extrapleural ligation of the TEF and primary repair. Group B included infants who underwent staged repair. All cases were type III according to Ladd's classification. Type III includes an upper and lower esophageal pouch associated with an inferior TEF. The length of the gap between the two pouches was not measured. Waterston's classification was used to distinguish three risk groups characterized by birth weight, preoperative pneumonia, and associated congenital malformations.

Group A consisted of 35 infants performed by primary repair after extrapleural ligation of the TEF. Group B included 21 infants, seven of whom had extrapleural ligation of the TEF (using thoracotomy approach) with a Fontan gastrostomy and cervical oesophagostomy. Two infants had a gastrostomy with permanent suction of the upper esophageal pouch, and twelve infants had a cervical esophagostomy and stoma of the lower esophageal pouch.

Parents from group B whose infants survived to hospital discharge (n=14) were included for the family needs study: their infants had a gastrostomy and a cervical esophagostomy. Informed consent was not obtained in four cases, hence excluded. Two neonates from group A were excluded because of insufficient information in the medical records relevant to the study. The total sample consisted of 54 children and 10 parents. Data investigated were: sex, gestational age, history of polyhydramnios, age at presentation, birth weight, type of atresia, associated congenital anomalies, presence of pneumonia at presentation to our unit, Waterston classification, age at surgery, type of surgery, anesthetic and resuscitation procedure, surgical complications, length of hospital stay, patient outcome, and experience and needs of families of children with gastrostomy and cervical esophagostomy. The parameters investigated, as well as the methodology, are synthesized in Figure **1**.

Preoperative care

All newborns had a chest radiograph. It showed the catheter curved in the proximal esophageal pouch. It also showed a subsequent aerated abdomen confirming the presence of the TEF (Figure **2**). A gentle suction of the upper esophageal pouch was performed in the newborn in the prone position associated with antibiotic prophylaxis. The other associated procedures depended on the clinical condition and associated

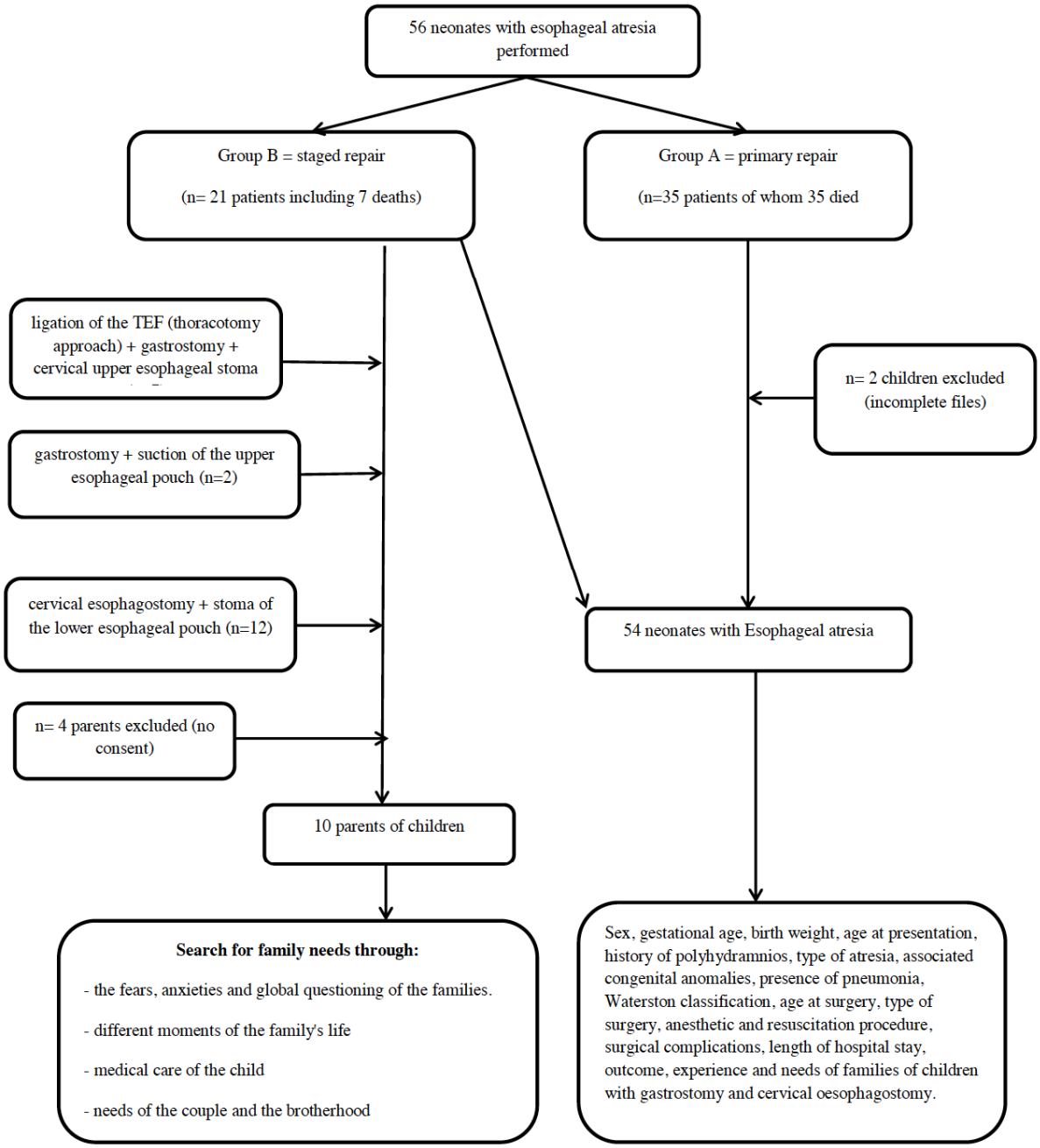

Figure 1: Study flowchart.

malformations. A cardiac ultrasound was carried out in all newborns.

Surgical Technique

Surgical management was variable and consisted of:

- A thoracotomy and then ligation of the TEF with primary repair. A Ch 6 trans-anastomotic catheter was left in place. Thirty-three children were treated with this technique.

- A cervical esophagostomy associated with fistula ligation (using abdominal approach) and lower esophageal pouch stoma [19]. Of the 12 infants operated on by this technique, 7 survived to discharge. Of those alive, two underwent colonic replacement at 3 ½ years and 1 year, respectively. The esophageal replacement was performed during the mission organized by a French *non-governmental organization* held regionally at the Hôpital Mère Enfant Dominique Ouattara in Bingerville, Côte d'Ivoire. The team was composed of senior French and regional lecturers and surgeons/anesthetists.

- A ligation of the TEF (using thoracotomy approach) followed by a Fontan gastrostomy and a cervical stoma of the upper esophageal pouch. All children in this group survived. One child in this group had a colonic replacement at 1 year of life in Bingerville.

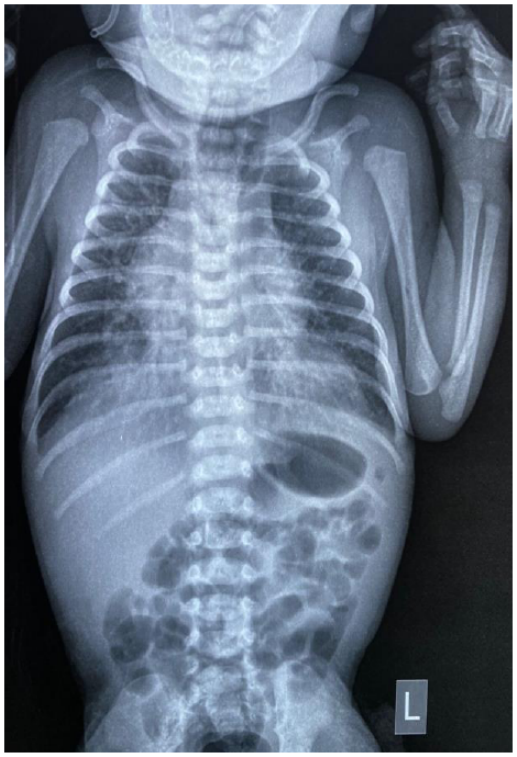

Figure 2: Chest radiograph showing type III esophageal atresia of the Ladd's classification.

Statistical Analysis

Data were collected using a survey form and entered into an Excel database. R software version 4.2.1 was used to analyze the results. Differences between groups were analyzed using the chi2 test. The level of significance was defined as $p < 0.05$.

RESULTS

There were 31 male newborns. The median for gestational age was 37 gestational weeks (range: 35 - 38). Polyhydramnios was observed in 4 cases. The mean birth weight was 2365 g. (range: 1000 g - 3500 g). The mean age at diagnosis was 3.48 days (range: 24 hours - 19 days). Associated malformations were diagnosed in 6 newborns. Four neonates had anorectal malformations, one neonate had anorectal malformations and hypospadias, and one neonate had spinal, limb and cardiac malformations as part of a VACTERL syndrome.

Surgical Treatment

Surgery was performed at day 5 on average with extremes from day 2 to day 17. Twenty-eight newborns had aspiration pneumonia. Surgery lasted an average of 2 hours and 12 minutes, and 15 children required a blood transfusion due to anemia. Table **1** shows the Waterston prognosis classification for the two groups and the survival rate.

Anesthesia/Resuscitation Procedure

Sevoflurane was the most used anesthetic gas (90%), followed by halothane in 10% of cases. They were associated with propofol for anesthetic induction and for intraoperative maintenance of anesthesia. The majority of newborns (72.3%) were intubated with a 2.5 mm intubation probe. The others were intubated with a size 3 one. Two peripheral venous routes were taken for the block but only one was used in most cases. The second was a backup one. No child had a central venous route. Ringer lactate and isotonic saline were the fluids used for surgical management. Extubation was immediate in 83.5% of cases in the operating room. The rest of the infants were extubated within a mean time of 45.56 h in the intensive care unit, with extremes of 24 h and 82 h.

Outcome

All infants from group A had an immediate postoperative course with complications leading to death. In group A, the mean age at the time of death was 6.78 days (range: 4-7 days). The most frequent complications were sepsis and persistent respiratory distress (Figure **3**). In Group B, immediate postoperative period was uneventful in 47.61% of

Table 1: Waterston Prognosis Classification and Survival Rate

	Group A (n=33)			Group B (n=21)			p=0.43
Waterston prognosis group	A	B	C	A	B	C	
Number of children	8	11	14	1	11	7	
Survival (%)	00	00	00	100	81.81	57.14	

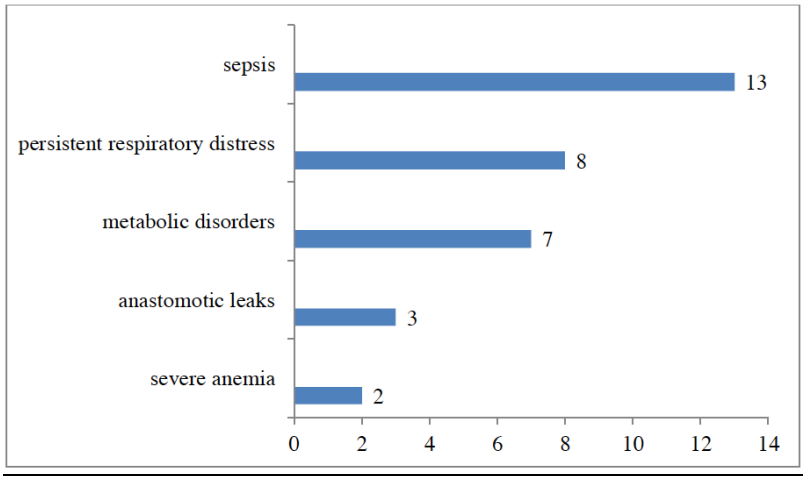

Figure 3: Early postoperative complications in children from group A (primary repair).

cases (10 out of 21). We recorded one case of hydropneumothorax after drainage, 6 cases of aspiration pneumomia and 2 cases of sepsis. There were two deaths due to respiratory and cardiac arrest.

Far from the initial surgery (more than six months postoperatively), five deaths were recorded. Two of the infants who underwent cervical esophagostomy, fistula ligation (using abdominal approach) and lower esophageal pouch stoma died after the initial surgical management, due to medical conditions (severe malaria in one child; malnutrition and severe pneumonia in another one). Two children succumbed to sepsis during hospitalization on the pediatric ward. Of the three children who underwent colonic replacement, one died during the esophageal replacement mission in Bingerville in the immediate postoperative period and a second after the treatment of a hiatal hernia. The overall number of deaths was 7 (33.33%) in group B with a statistically significant difference between the two groups ($p=19.10^{-8}$) Postoperative course is detailed in Table **2**.

Feeding was performed through the trans-anastomotic catheter or by the gastrostomy. In the cases of primary anastomosis, feeding was started at day 4 postoperatively on average (range: 3 - 7 days). The newborns in group B were fed as soon as the clinical condition allowed it through the gastrostomy. The average time to refeeding was 3 days (range: 2 - 4.5 days. No child received parenteral nutrition. The average length of hospitalization was 6.79 days (range: 4 - 17 days).

Family's Experience

Ten parents of children were interviewed to investigate the experience and needs of families whose children have a gastrostomy and cervical esophagostomy. Various difficulties were expressed. Qualitative analysis of the findings reveal at bedtime, getting up, and every day, the difficulties were stress, fear, and anxiety about losing the child in 80% of cases. "Every day that passed was a challenge won." confessed Marceline, a 26-year-old primiparous mother.

Table 2: Postoperative Course of All Operated Newborns

	Number of cases (n=54)	Colonic replacement performed (n=3)	Awaiting colonic replacement (n=11)	Died (n=40)	
Group A (n=33) **Primary repair**	33	-	-	33	$p=19.10^{-8}$*
Group B or staged repair (n= 21)					
Cervical oesophagostomy + gastrostomy	7	1	6	0	
Upper oesophageal pouch suction + Gastrostomy	2	-	-	2	
Cervical esophagostomy + stoma of the lower esophageal pouch	12	2	5	5	

*significant value.

Socially, four out of ten parents had to stop their activities to be available to take care of their child. Isabelle [a 35-year-old seller of adulterated petrol], Francine [a 38-year-old hairdresser's trainee], Violette and Rogatienne [two farmers of 23 and 31 years old], had to interrupt their work temporarily. For all parents, it was difficult to bear the crooked looks of others, the inappropriate comments. Rogatienne reported "He is not a child but a burden." The infant of Damienne [a 30-year-old general practitioner] has often been called a witch child by her roommates. Marceline had complained that some people think that it is definitely a child born of an illegitimate union. "This child is the fruit of lovemaking in bizarre sexual positions". Firmine told us. The condition was unknown, making it very difficult to explain it to outsiders and to eat when going out. The lack of funding for care by the government was mentioned by all parents.

The challenges at mealtime are represented in this social context where the disease is not known, by the difficulty of finding a hidden place at each meal so as not to disturb people and not to attract attention. For eight parents, it was also very hard to find a suitable diet to be administered through the catheter and to allow normal growth and weight gain. It was a nutritional challenge.

Family's Needs

Firmine [a 28-year-old female teacher] wrote: "The condition is not well known by the medical community so that it is quite difficult to manage in case of illness." The non-existence of a dedicated team for the follow-up was deplored by four parents and sometimes made the care complicated. Within the family, this situation is a source of general stress and this has repercussions on the family and on the harmony that must exist. In addition, it generates tension in the siblings (jealousy because he is more loved and source of attention, anxiety). The fitting of the stoma, the management of irritations, leaks or the accidental removal of the gastrostomy catheter are stressful situations outside the big cities and make any travel difficult and worrying. These observations were made by Genevieve, Agathe and Theodora respectively. All the parents had expressed the need for psychological support because 70% of them reported: "we have traumas that affect us for life".

DISCUSSION

Surgical Procedure and Outcome

Esophageal atresia (EA) is the type of condition that is life-threatening if no adequate action is taken [16]. Since the first successful repair more than 50 years ago, much has changed in terms of treatment and outcome [20]. Over the decades, the overall survival rate has steadily improved due to advances in surgical techniques and preoperative and postoperative medical care [21, 22]. Neonatal surgical mortality in general has decreased because of increased understanding of the condition and specialized training of neonatal surgeons, pediatric anesthesiologists, and neonatal intensive care providers. Advances in machine monitoring technology, nutrition, and antibiotics have also made survival and early definitive surgery more achievable [23]. In low-income countries, respiratory complications remain an important survival factor with a high rate of pneumonia at presentation and a lower level of neonatal support [24]. This explains the high mortality from EA in developing countries. In our series, the overall mortality was 74.07% with 51.85% of newborns presenting with a aspiration pneumonia on admission. This mortality is comparable in Sub-Saharan Africa to Fall *et al*. [25] in Senegal (72%), Bandre *et al*. [6] in Burkina Faso where the mortality was 76%. Randriamizao *et al*. [26] in Madagascar had also found similar mortality to ours (76.5%). It was, however, higher than that of Osei-Nketiah *et al*. [27] in Ghana (59.7%) and significantly higher than that reported by Tönz *et al*. [20] in Switzerland (14%) and by Gottrand *et al*. [28] in France, where it rarely exceeds 5-10% in recent years. Improved surgical and perioperative care, early diagnosis, and a decrease in pneumonia in developed countries explain these results [29, 30]. In our centres, more than half of newborns were seen at an advanced age of life with pulmonary complications as in most African series. In addition, the challenges of lack of well-equipped neonatal intensive care units and total parenteral nutrition are factors identified in previous studies as responsible for poor outcome of neonatal surgical emergencies in sub-Saharan Africa [16, 31, 32].

While primary repair of EA is mainly the preferred option for management [24, 33-36], mortality is still high for primary anastomosis in sub-Saharan Africa raising several questions about its management. Hence the needs to further enhance management in resource-limited settings. Several countries, such as Benin, have adopted a strategy of repairing these cases of EA. This is the case in Nigeria, which in 2013 had adopted primary anastomosis as a treatment strategy [7, 17] in neonates seen without feeding within 72 h of life and without thoracic signs. For patients presenting after 72 h of life and those with important thoracic signs, a staged repair was used. This consisted of a gastrostomy performed after ligation of the fistula

combined with a cervical esophagostomy. These children received an esophageal replacement within six months of the initial procedure. As a result of this procedure, the initial mortality was reduced, especially in neonates admitted with low birth weight, prematurity and/or pneumonia. In Ghana such an approach was also taken because of the high mortality associated with primary repair. However, when the results were evaluated, there was no significant difference in mortality between primary anastomosis and multistage reconstruction [27]. Although this study does not show a significant difference in mortality reduction, it is important to note in our series a reduction in mortality related to this multistage procedure. Mortality was once 100% in group A and 26.31% (5 deaths out of 19 operated) in group B. In Australia, because of the poor outcomes in the 1960s, a policy of management of EA was also developed. All patients were managed with early gastrostomy and then medically stabilized before thoracotomy and definitive repair a few days later. As outcomes improved, the rigid policy gradually relaxed and gastrostomy was performed only for long defect EA or as an option for treatment of anastomotic leaks [37]. This is a possible treatment option and better than the current practice in our country because there is no substitute for a native esophagus and it should be preserved whenever possible. However, in the absence of a sufficient number of medical aspirators and adequate means of resuscitation, it remains an option to be considered less.

The lack of postoperative neonatal respiratory support explained the rather high level of extubation on the table in our setting. This was similar to the practice reported in Madagascar by Randriamizao *et al* [26] where extubation in the operating room, as soon as the patient woke up after the anaesthesia, was performed because of the lack of postoperative neonatal respiratory support. Oxygen weaning was completed when the 21% oxygen saturation was greater than 92%, without respiratory distress [26]. These children, extubated on a table or not, had experienced sepsis, respiratory distress and metabolic disorders as postoperative complications. Sepsis remains a frequent postoperative complication in developing countries. Vukadin *et al*. [38] in Serbia had found sepsis and anastomotic stricture as frequent complications. In addition, there was a significant association of sepsis with mortality ($p<0.002$). In Nigeria, Ekenze *et al*. [17] had shown that sepsis was associated in 40% of cases with mortality. It is followed by pneumonia with undernutrition disorders in our context, for Mouafo Tambo *et al*. [16] in Cameroon and for Dave *et al* [39] in India.

Needs of Families

Addressing the needs of families of children with a gastrostomy in the case of EA is part of the quality of care and meets a medical but also social requirement. It is an approach aimed at improving the care of these children in its entirety in order to resolve a frustration, a lack. Several European societies working against some rare diseases have already considered the issue of the needs of families whose child has an EA. This is done through surveys or group sessions with parents or through exchanges and family meetings. However, it appears that focusing on this in a child with a gastrostomy and a cervical esophageal stoma in the context of EA is somewhat original. From discussions with the parents in our sample, the challenges at bedtime, wake-up, and each day were represented by stress, fear, and anxiety about losing their child. This is similar to the data of the l'Association Française de l'Atrésie de l'Œsophage (AFAO) which found among the problems that arose at bedtime, getting up and during the night; anxiety, stress and fear of separation. To resolve their anxieties, parents suggested in this survey the need for psychological support, more frequent exchanges with other parents, positive feedback and local meetings [40]. The creation of a framework for dialogue is essential. Several associations exist. Apart from AFAO in France, there is the KEKS (*Kreis für Eltern von kindern mitSpeiseröhrenmißbildungen*) in Germany, VOKS in the Netherlands (*Vereniging van Ouders van Kinderen met een Slokdarm Afsluiting*) and ABeFA (*Association Belge des Familles touchées par l'Atrésie de l'Œsophage*) united in the European Federation EAT (Esophageal Atresia and Tracheo-esophageal fistula support groups), TOFS (Tracheal Oesophageal Fistula Support) in the United Kingdom and OARA (Oesophageal Atresia Research Auxillary) in Australia. They contribute to better overall management of the condition by promoting cooperation between patients and their parents, patient organizations, and health care providers [41]. The creation of such an association or support group in our context will furthermore popularize the condition and allow for a better understanding and its management; thus addressing the needs related to social stigma expressed by the parents. One of the problems mentioned by parents is the unavailability of a dedicated team for the follow-up of the child. The lack of a customized health card, the lack of qualified health care provider and the unequal distribution of available health resources explain this state. In developed countries like France, a personalized emergency card specific to EA is given in

the context of emergency hospitalization. It allows the team to be contacted in case of need and is also useful in the case of a school trip, a consultation, etc. For each child, a hospital referral specialist is identified, to ensure the link with the town's health care providers (in particular the general practitioner), for telephone advice and to help with first-line diagnostic and therapeutic management [42, 43]. In Benin, follow-up was done by a pediatric surgeon monthly for the first six months and then quarterly. Children were reviewed in case of surgical problems and referred to a medical specialist if needed. Some preventable deaths occurred among the children in follow-up due to the absence of a team dedicated and the non-availability of a qualified primary care team in the patient's home town. The establishment of a multidisciplinary team remains an objective to be aimed at and will make it possible to resolve this situation.

In Benin, despite persistent barriers to care, the management of EA can be improved and should be a concern of health care providers and leaders. Primary repair using thoracotomy or thoracoscopy approach remains the reference treatment for type III EA with a short gap. However, until the technical platform and the organization of care in our community improve, staged repair remains an essential option for survival. The creation of a framework for discussion with the families authorizing social actions and quality medical accompaniment, and government funding of care will improve the quality of care administered to these children and their families.

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Nil

LEVEL OF EVIDENCE

Level IV

ABBREVIATIONS

EA: esophageal atresia, TEF: tracheoesophageal fistula

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