Original Article

Successful Management of Oesophageal Atresia in Cameroon, Sub-Saharan Africa

Olivier Kopong Fola[1], Bonaventure Jemea[2,3], Jean-Emile Emmanuel Bayiha[4], Bernadette Ngo Nonga[1,3]

Departments of [1]Surgery, [2]Anesthesia and Intensive Care Unit, University Hospital Centre of Yaounde, University of Yaounde I, [3]Department of Surgery and Subspecialities, Faculty of Medicine and Biomedical Sciences, University of Yaounde I, [4]Department of Surgery, Yaounde Central Hospital, University of Yaounde I, Yaounde, Cameroon

Abstract

Background: Oesophageal atresia (EA) is the most common congenital anomaly of the oesophagus. Despite improvement of survival observed over the previous two decades in developed countries, the mortality remains very high and the management greatly challenging in resource-poor settings such as Cameroon. We report our experience of management of EA in this environment, with a successful outcome. **Materials and Methods:** We prospectively assessed patients diagnosed with EA and operated in January 2019, at the University Hospital Centre of Yaounde. Records were reviewed for demographics, history and physical examinations, radiological findings, surgical procedures and outcomes. The study has received approval from the Institutional Ethics Committees. **Results:** In total, six patients (three males and three females, sex ratio, 0.5; mean age at diagnosis, 3.6 days; range, 1–7 days) were assessed. A past history of polyhydramnios was found in one patient (16.7%). All patients were classified Waterston Group A at diagnosis, with Ladd-Swenson type III atresia. Early primary repair was performed in four patients (66.7%) and delayed primary repair in two patients (33.3%). Operative repair mainly involved resection of the fistula, suture of trachea and oesophagus end-to-end anastomosis, followed by interposition of vascularised pleural flap. Patients were followed up 24 months. With one late death, the survival rate was 83.3%. **Conclusion:** Improvement has been achieved in the outcomes of neonatal surgery in Africa in the past two decades, but EA-related mortality remains relatively too high. Using simple techniques and available, reproducible equipment can improve survival in resource-poor settings.

Keywords: Oesophageal atresia, resource-poor setting, successful management, Cameroon

Introduction

Oesophageal atresia (EA) is a congenital malformation defined by the discontinuity of the oesophagus occurring in 2.4 in 10,000 births.[1] EA with or without tracheoesophageal fistula (TEF) is a foregut defect that is a major component of the VATER/VACTERL association.[2] The current definition of the VATER/VACTERL association is the presence of at least three of the following: Vertebral defects, anal atresia, cardiac defects, TE fistula, or renal and limb anomalies in the absence of a specific genetic diagnosis.[2] EA can be isolated (in 50% of cases, being sporadic or showing a risk of intra-familial recurrence) or associated with Trisomy 21, Trisomy 18, 22q11 deletion or specific clinical entities such as CHARGE (Coloboma, heart defect, choanal atresia, growth and mental retardation, genital hypoplasia and ear anomalies), Feingold, Opitz, Pallister-Hall, Potter, Schisis, Goldenhar or G syndromes.[2] The history of EA commenced in the year 1670 with Durston's description of 'A narrative of a monstrous birth at Plymouth'.[3,4] Surgical correction was started only in 1920s and the first survivors were reported in 1939.[3] Thomas Gibson first described EA associated with TEF in 1697. The survival of infants born with EA, TEF or both has improved dramatically since Cameron Haight's first successful repair in 1941, after innumerable attempts by other surgeons.[5-7] The improvement of survival observed over the previous two decades is multifactorial and largely attributable to early surgical intervention and advances

Address for correspondence: Dr. Olivier Kopong Fola, Department of Surgery, University Hospital Centre of Yaounde, University of Yaounde I, Yaounde, Cameroon.
E-mail: dr.olivierfola@yahoo.com

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in neonatal intensive care, neonatal anaesthesia, ventilatory and nutritional support, antibiotics, surgical materials and techniques.[8] Indeed, mortality is currently limited to those cases with coexisting severe life-threatening anomalies worldwide.[8] This improvement in survival remains remarkable in developed countries with overall rates which have risen from 4.6% in 1972-76 to 100% nowadays.[9-13] However, the overall mortality still remains very high in third world settings, like in Africa, with rates ranging mainly between 80 and 100%.[14-19] Thus, management and outcome of EA are still challenging in resource-poor settings because of lack of appropriate infrastructures, surgical materials and supplies; lack of health-cover system for most of the population, suitable staff and approaches that are adapted to the African context. Very few studies have been carried out on EA in Cameroon. Hence, here we report our experience on the management of EA with a favourable outcome in a very resource-poor setting.

Materials and Methods

Patients

We carried out a prospective study from January 2019 to December 2020 at the University Hospital Center of Yaounde, Cameroon. Patients with EA, operated on in our centre in january 2019 were registered in a predefined computerised database containing data on demographics, presenting symptoms, past medical history (antenatal visits and congenital anomalies), radiological findings, pre-operative evaluation, operative procedure, post operative morbidity, post operative mortality and outcomes. All patients were operated on by the same team.

Preoperative workup

Complete blood counts, renal function tests, serum electrolytes, HIV testing, coagulation profile, blood type and crossmatching.

Procedure

Primary repair

Surgery was performed under general anaesthesia. A regular single-lumen endotracheal tube was used to achieve non-selective lung intubation without inflation of the balloon. Patients were then placed in the lateral decubitus position, on the opposite side of the minithoracotomy. Ceftriaxone and metronidazole were used for antibiotic prophylaxis during surgery. We made an 8 cm small skin incision for posterior muscle-sparing minithoracotomy to enter the chest cavity through the right 5th intercostal space. We then proceeded to exploration. All the patients had a type III atresia, with distal TEF, according to Ladd and Swenson classification.

We mainly proceeded with resection of the fistula, suture of trachea by transfixing suture, permeabilisation of the upper pouch of oesophagus, and end-to-end anastomosis of the esophagus under less tension, by interrupted sutures, with a 12 French nasogastric tube used as transanastomotic tube and pushed far toward the duodenum [Figure 1]; followed by interposition of vascularised pleural flap on trachea (after closure of the fistula).

Delayed primary repair

Surgery was performed under general anaesthesia. A regular single-lumen endotracheal tube was used to achieve non-selective lung intubation without inflation of the balloon. Patients were then placed in the lateral decubitus position, on the opposite side of the minithoracotomy. Ceftriaxone and metronidazole were used for antibiotic prophylaxis during surgery. We made a 9 cm skin incision for posterior muscle-sparing minithoracotomy to enter the chest cavity through the right 5th intercostal space. We then proceeded to exploration. All the patients had a type III atresia, with distal TEF, according to Ladd and Swenson classification.

We mainly proceeded with resection of the fistula, suture of trachea by transfixing suture, permeabilisation of the upper pouch of oesophagus, and end-to-end anastomosis of the esophagus under less tension, by interrupted sutures, with a 12 French nasogastric tube used as transanastomotic tube and pushed far toward the duodenum; followed by interposition of vascularised pleural flap on trachea (after closure of the fistula).

A retrosternal gastric pull-up was performed in an infant; the cervical oesophagus was mobilized through the neck, the stomach was mobilised through laparotomy, the left gastric artery was ligated, and pyloromyotomy was done. The stomach was brought to the chest through the transhiatal route. A single-layer oesophageo-gastric retrosternal anastomosis was performed under the same conditions described above, with the stomach calibrated on the native oesophagus.

Post-operative drainage system

A simple surgical drainage system consisting of a hose and a manifold connected to the chest tube [Figure 2] was used in the post-operative period in each case. A formal system for post-operative pleural drainage is not readily available in Cameroon and our hospital was not equipped with wall suction devices. None of our patients were placed on respirator during the post-operative period.

Post-operative management

Patients were transferred to the surgical intensive care unit where they received standard post-operative care including intravenous fluids, analgesics and post-operative antibiotics. Then, they were transferred to the paediatric department by post-operative day 2. Chest physiotherapy was initiated as soon as possible. We aggressively managed any post-operative pain. Patients were fed early, by post-operative day 1 through the nasogastric tube. The chest tube was removed on post-operative day 5. A chest radiograph was performed before and after the removal of the thoracic drain. The nasogastric tube was removed 7–10 days post-operative. The study was approved by the ethics committee of the Faculty of Medicine and Biomedical Science and the National Ethics Committee of Cameroon and conformed to the tenets of the Declaration of Helsinki. Written informed consent was obtained from patients whose images are enclosed. Data analysis was performed using Epi info 7 (Centre for Disease Control and Prevention, Atlanta, GA).

Results

We analysed the records of six patients (3 males and 3 females, sex ratio, 0.5; mean age at diagnosis, 3.6 days; range, 1–7 days). Four new-borns and two infants were concerned. A past history of polyhydramnios was found in one patient (16.7%). The average number of antenatal visits was 4 (range, 4–6). The mean gestational age was 37.8 weeks (range, 37–38 weeks). The mean birth weight was 2992 g (range, 2780–3300 g). All patients had been referred from peripheral hospitals across the country. Post-natal diagnosis was made average 4.2 days after birth (range, 1–8 days). No associated malformation was found.

The main features at presentation were hypersalivation (6; 100%), regurgitation (6; 100%) bronchial congestion (2; 33.3%) and abdominal bloating (1; 16.7%). All the patients were classified Waterston group A at diagnosis. A type III atresia was found in all the patients, according to Ladd and Swenson classification.

Early primary repair was performed in four patients (66.7%), neonates and delayed primary repair in two patients (33.3%), infants. The delayed primary repair was due to the fact that the two infants presented to our institute only at those advanced ages. In fact, two infants were operated, aged 3 and 4 months, respectively with a feeding gastrostomy performed at birth in both.

Surgery took place on average 2.4 days after admission (range, 1–4 days). The HIV serology was negative in all patients. The pre-operative American Society of Anaesthesia score was in class III and with Altemeier class 2, in all patients. Surgical procedures took place under general anaesthesia with non-selective lung intubation. A posterior muscle sparing minithoracotomy was systematically performed on the right fifth intercostal space. Incision' sizes were 8 cm for neonates and 9 cm for infants. We mainly proceeded with resection of the fistula, suture of trachea by transfixing suture, permeabilisation of the upper pouch of oesophagus, and end-to-end anastomosis of the esophagus under less tension, by interrupted sutures, with a 12 French nasogastric tube used as transanastomotic tube and pushed far toward the duodenum [Figure 1]; followed by interposition of vascularised pleural flap on trachea (after closure of the fistula). A retrosternal gastric pull-up was performed in an infant, with a single-layer esophageo-gastric retrosternal anastomosis. No intraoperative complications were encountered. Intraoperative bleeding did not exceeded 20 mL. The duration of surgery was between 60 and 90 min.

At the end of surgery, a chest tube was systematically put in place, 12 French for neonates and 14 French for infants. Incubators were available in two cases [Figure 2]. When incubators were not available, patients were warmed using simple devices with hot water bottles wrapped in towels [Figure 3]. A simple surgical drainage system [Figure 2] was used in the post-operative period. A formal system for post-operative pleural drainage is not readily available in Cameroon and our hospital was not equipped with wall suction device.

Respiratory physiotherapy was performed by stimulating crying and dorsal clapping. None of our patients were placed on the respirator during the post-operative period. Oral feeding was initiated at day one post-operative through the nasogastric tube. Chest tubes were removed at day five post-operative. Nasogastric tubes were removed 7–10 days post-operative, before discharge. Skin sutures were removed on the post-operative day 12. One case of severe pneumonia was observed in an infant in the early post-operative period. This pneumonia was successfully managed by subsequent resuscitation including oxygen supply and broad-spectrum antibiotics such as imipenem. The intensity of post-operative pain was difficult to assess. The EVENDOL scale was used

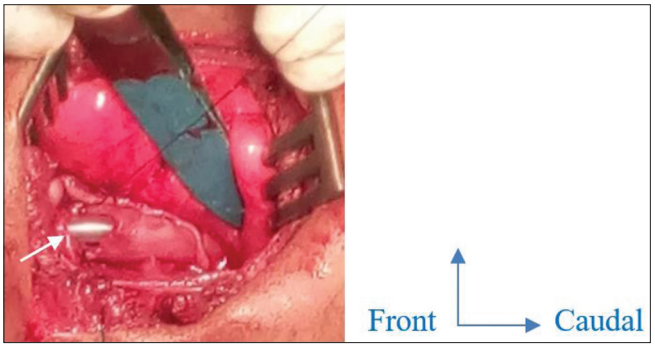


Figure 1: Minithoracotomy and esophagus end-to-end anastomosis using a 12 French nasogastric tube as stent (white arrow)

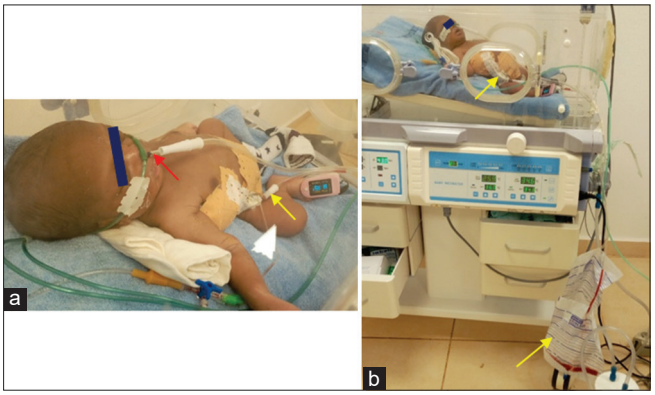


Figure 2: Newborn in the post-operative period. (a) Nasogastric (red arrow) and chest tube (yellow arrow) are visible. (b) Simple surgical drainage system (yellow arrows)

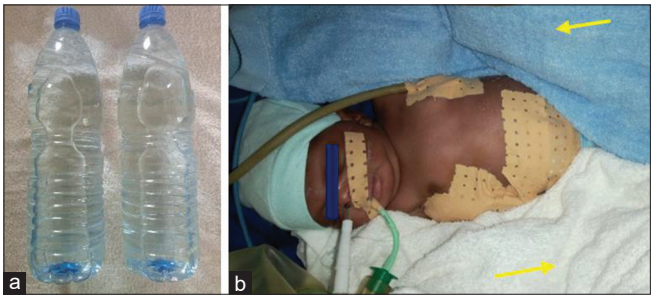


Figure 3: Usage of simple devices with hot water bottles (a) wrapped in towels (b) to warm babies (yellow arrows)

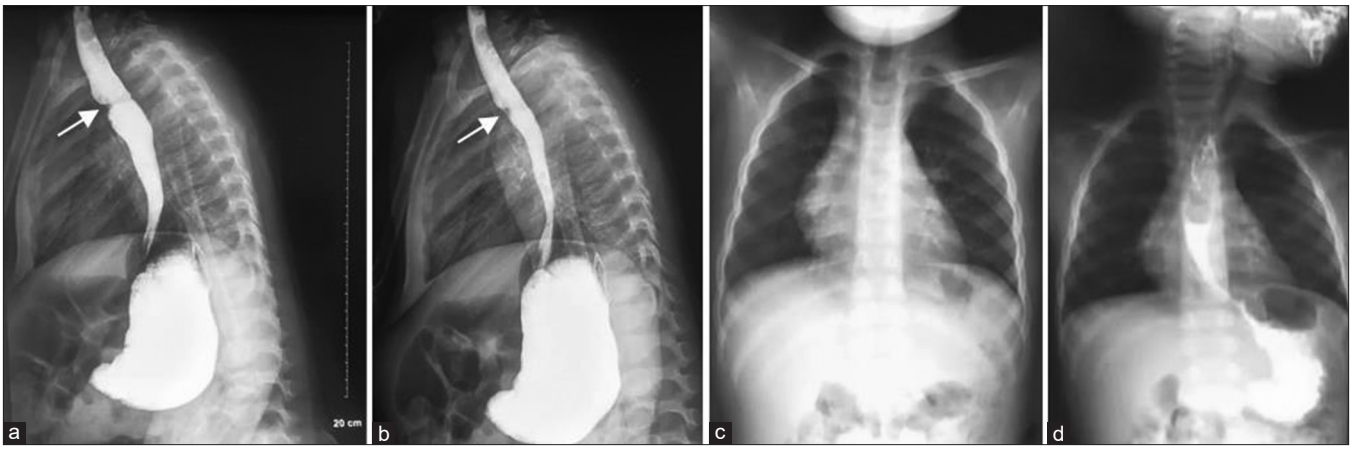


Figure 4: Water soluble contrast swallow test (a and b) showing a patent oesophageal anastomosis (white arrow), 18 months after surgery. Two years later, the anastomosis is still patent (c and d)

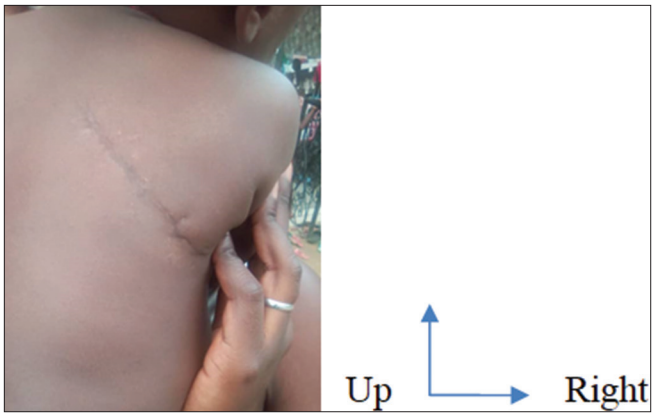


Figure 5: Healthy patient with esthetic scar, thirteen months after surgery for EA

in children and pain was rated at 2/15 at most. New-borns and infants were generally relieved by paracetamol. At discharge, patients had no residual pain. Patients were followed up 24 months. Patients' growth curves were ideal. Delayed post-operative water soluble contrast swallow tests showed patent oesophageal anastomosis [Figure 4]. One of the infants died 7 months after surgery from severe community-acquired pneumonia. The survival rate was 83.3% at 2 years, with healthy patients and aesthetic scars [Figure 5].

Discussion

Techniques of TEF closure and oesophageal anastomosis, as well as early feeding, are crucial for survival after EA surgery. The European Reference Network for Rare Inherited Congenital Anomalies (ERNICA) in its latest recommendations of August 2020, emphasised the particular benefits of TEF closure by transfixing suture, oesophageal anastomosis by interrupted sutures, and initiation of feeding 24 hours post-operatively, as well as routine insertion of transanastomotic tube or maximum duration of thoracoscopy of 3 hours, in the improvement of outcome.[20] Our patients were fed early, at day 1 post-operative. They were properly warmed up with our makeshift means. We performed TEF closure by transfixing suture. We proceeded with end-to-end anastomosis of the oesophagus under less tension, by interrupted sutures, with a nasogastric tube used as transanastomotic tube, pushed far toward duodenum, as well as a pleural patch on trachea after closure of the fistula. All that greatly influenced survival and permitted families of patients to avoid wasting money with parenteral nutrition, which is very expensive and not readily available in our settings.

The clinical features of EA are polymorphic and lack of symptoms does not exclude the presence of the pathology. Polyhydramnios is a condition often associated with EA. The overall mortality still remains very high in third world settings, like in Africa, with rates ranging mainly between 80 and 100%.[14-19] Major complications and causes of death worldwide include severe pneumonia with atelectasia, sepsis, anastomotic leak, congestive heart failure, gastroesophageal reflux, anastomotic stricture, wound infection, recurrent TEF, hypothermia and hyponutrition.[9,15,21] Anastomosis under tension and tracheomalacia are identified risk factors for anastomotic complications.[22]

Advances in diagnostics techniques and perioperative care have greatly improved the outcome of neonatal surgery. Surgeons should carefully perform anastomosis under less tension to prevent anastomotic complications in the primary repair of EA/TEF.[22] The technique of suture when performing anastomosis is also determinant. Interrupted sutures are more and more recommended worldwide to prevent inherent complications, compared to disrupted sutures sometimes ischaemic on a non-well vascularised oesophageal wall.[20] Routine insertion of transanastomotic tube is strongly recommended, despite some cases of associated increase in oesophageal stricture formation described.[20-24] Oesophageal end-to-end anastomosis is technically relatively more easy to perform than end-to-side anastomosis. At term, there is no significant differences between end-to-end and end-to-side anastomosis, despite end-to-side repair has been reported to be associated with reduced rate of stricture and gastro-oesophageal reflux disease requiring operation compared with end-to-end

repair.[25,26] Operative repair involving resection of the fistula, suture of trachea and oesophagus followed by interposition of large vascularised pleural flap has demonstrated no evidence of recurrence at long-term follow-up.[27]

Non-selective lung intubation without inflation of the balloon was systematically performed. Insuflation of the balloon in new born and infants may compromise tracheal wall capillary blood flow, possibly leading to mucosal ischaemia, ulcerations, tracheomalacia, tracheal stenosis, and even tracheal rupture or tracheo-oesophageal fistulae.[28,29]

Minithoracotomies are ideal approaches to face associated musculoskeletal problems following classic thoracotomies. Although associated with a longer operative time, thoracoscopic repair has the advantages of an earlier time to extubation, first oral feeding and shorter hospital stay.[24] There is an overall non-statistically significant difference in morbidity comparing thoracoscopic approach with conventional open repair of EA.[24,30,31] Minithoracotomies remain interesting approaches in resource-limited settings where thoracoscopy is not avaliable, in this era of minimally invasive surgery.

We used a simple surgical drainage system [Figure 2] in the post-operative period. It is generally recommended to use a low pressure wall suction to be able to drain the blood from the pleural cavity. Prior studies carried out in our institute shown that simple gravity was enough to provide good drainage with the same results.[32,33] A costless simple drainage system can be efficient. The major problems encountered in our daily practice are lack of infrastructure with proper equipment and poverty, since there is no health cover system. Despite this unfavorable context, a successful management of EA can be done with simple principles and means.

Conclusion

Improvement has been achieved in the outcomes of neonatal surgery in Africa in the past two decades, but EA-related mortality remains relatively too high. Using simple techniques and available, reproducible equipment can improve survival in resource-poor settings.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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