Case Report

Thoracoscopic Repair of Type C Oesophageal Atresia in a Nigerian Hospital: A Report of Two Cases

Arua Obasi Igwe, Lakmeel Kannachamkandy[1], Ebere Lilian Agwu[1], Temitope A. Lawal[1], Temilolu O. Nwankpele[1], Arinzechukwu O. Mosanya[2], Nikoloz Dzagnidze[2], Augustine Jeremai Olugbemi[3], Ibukun Abidoye[4], Andriy Danyltsiv[5]

Department of Surgery, Paediatric Surgery Unit, Evercare Hospital Lekki, [1]Department Paediatrics and Neonatology, Evercare Hospital Lekki, [2]Department of Surgery, General Surgery Unit, Evercare Hospital Lekki, [3]Department of Surgery, Cardiothoracic Surgery Unit, Evercare Hospital Lekki, [4]Department of Radiology, Evercare Hospital Lekki, [5]Department of Anaesthesia and Critical Care, Evercare Hospital Lekki, Lekki, Lagos State, Nigeria

Abstract

Management of oesophageal atresia (OA) with tracheoesophageal fistula (TOF) in Nigeria and the West African subregion has no doubt been a very demanding task for paediatric surgeons, not necessarily due to lack of skills, but due to the significant demand on neonatal intensive care, which in our region, is often fitted with the poor infrastructure needed to make this a success. Furthermore, the use of open thoracotomy has increased this demand resulting in a significant number having severe morbidities and significant mortality rates. Hence, in our subregion, there is still a slow progression to meet up with the evolving trend of the management of this complex condition in the developed world. Following the first documented successful thoracoscopic repair of OA with TOF since 2000, there has been a progressive evolution and refinement of this technique, such that thoracoscopic repair is fast becoming the gold standard for the repair of all types of OAs, including long-gap anomalies. This article reports our experience with the first two cases of thoracoscopic repair of OA with TOF in the West African subregion.

Keywords: Children, oesophageal atresia, thoracoscopy, tracheoesophageal fistula

Introduction

Oesophageal atresia (OA) is a congenital anomaly characterised by interruption of the oesophagus, creating a proximal and a distal end, which may or may not be connected with the trachea as a tracheoesophageal fistula (TOF). It forms part of the VACTERL (Vertebral anomalies, Anal atresia, Cardiac defects, Tracheo-Esophageal fistula, Renal anomalies, Limb abnormalities) anomalies; hence, may occur together with any.

This anomaly occurs in about 1 in 2500–3000 live births, with a slight male preponderance of 1.26:1.[1]

The mainstay of treatment has been surgical reconnection of the two ends or oesophageal replacement depending on the type. The surgical technique can be open or thoracoscopic depending on the surgeon's experience and expertise.

In Nigeria, the mortality rate has remained very high mainly due to late presentation, and poor facilities for adequate pre- and post-operative management.[2,3]

Received: 30-10-2022 **Revised:** 20-03-2023 **Accepted:** 06-04-2023 **Available Online:** 14-03-2024

Access this article online

Quick Response Code:

Website: https://journals.lww.com/AJPS

DOI: 10.4103/ajps.ajps_145_22

Up till now, to the best of our knowledge, open thoracotomy is the only primary procedure performed in West African countries for these children to create oesophageal anastomosis, and this, however, has had very high mortality and morbidity rates. We, hereby, present the first two cases of primary repair done using the thoracoscopic technique in a private multi-specialist hospital in Nigeria.

Case Reports

Case 1

Our first case was a 3.5 kg 3-day-old male neonate delivered in an external facility by spontaneous vaginal delivery at 38 weeks, who was noted to have difficulty with breathing, repeated choking and cyanotic and apnoeic spells. An attempt

Address for correspondence: Dr. Arua Obasi Igwe, Evercare Hospital Lekki, Lekki, Lagos State, Nigeria.
E-mail: aruaobasi@yahoo.com

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

For reprints contact: WKHLRPMedknow_reprints@wolterskluwer.com

How to cite this article: Igwe AO, Kannachamkandy L, Agwu EL, Lawal TA, Nwankpele TO, Mosanya AO, *et al.* Thoracoscopic repair of type C oesophageal atresia in a Nigerian hospital: A report of two cases. Afr J Paediatr Surg 2024;21:129-33.

to pass a nasogastric (NG) tube failed, with the tube coiled up in the upper oesophagus; hence, a diagnosis of OA with TOF was suspected.

He was subsequently transferred to a nearby teaching hospital, where he was initially resuscitated; however, was said to continue to have recurrent apnoea and three episodes of cardiac arrest requiring cardiopulmonary resuscitation, and was subsequently referred to us due to a lack of facility for advanced neonatal intensive care, including mechanical ventilation. On presentation, he was resuscitated and placed on mechanical ventilation in the neonatal intensive care unit (NICU).

The chest X-ray showed a right upper lobe atelectasis, the NG tube at the upper pouch which suggests a proximal atresia and gas in the stomach which confirmed a distal fistula [Figure 1]. 2D echocardiography (ECHO) done did not show any significant cardiac anomaly.

After 24 h, a repeat X-ray showed re-expansion of the right upper lung [Figure 2]. He was subsequently taken to the operating room the same day for thoracoscopic repair after informed consent was obtained from the parents.

Surgical technique

The patient was transferred to the operating room on a transport ventilator with main tracheal intubation. He was placed in a modified prone position with a 30° right tilt.

After routine skin preparation and draping, a Veress needle was introduced into the chest through the 4th intercostal space below the tip of the scapula, and the lung was collapsed with CO_2 insufflation at the rate of 1 L/min with a maximum pressure of 6 mmHg. A 5-mm step trocar was placed in the 4th intercostal space below the tip of the scapula; a 3-mm trocar was placed at around the 3rd intercostal space, and a second 3-mm trocar at the 6th intercostal space, both at the posterior axillary line [Figure 3].

A 5-mm 30° telescope was introduced through the 5-mm port, and the azygos vein was identified as a landmark for dissection.

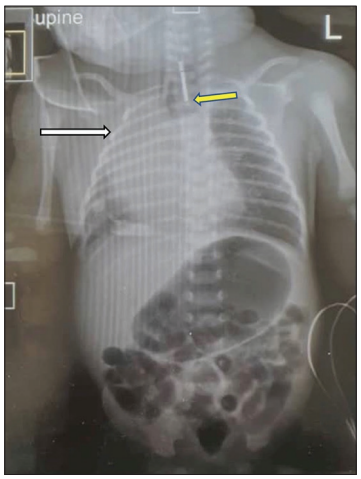


Figure 1: Radio-opaque tube (yellow arrow) in the upper pouch and distal bowel gas with right upper lobe atelectasis (white arrow). (inpatient X-ray from Evercare Hospital Lekki)

The lower oesophagus was mobilised first down to the trachea fistula while preserving the vagus nerves running along the wall, and the fistula ligated using polydioxanone (PDS) 4/0. Using a hitch stitch to elevate the distal oesophagus, the ligation of the fistula flush with the trachea was facilitated [Figure 4a].

The anaesthetist pushed down on the NG tube to facilitate the identification of the upper pouch. The upper pouch was mobilised using blunt and sharp dissection with minimal electrocautery until an adequate length was achieved [Figure 4b]. The first stitch was placed on the posterior wall bringing the edges together. The tube NG was advanced into the stomach, and the anastomosis was completed using interrupted PDS 4/0 sutures on a round body needle. Figure 5 shows the completed anastomosis. A chest drain was placed, and the wounds were closed with Vicryl 4/0 sutures.

The patient was then transferred back to the NICU, where he continued on mechanical ventilation for 4 days post-surgery.

He was successfully extubated on the 4th day post-surgery, and was commenced on NG tube feeding, which he tolerated well. He was then taken for a fluoroscopic barium swallow study on day 6, which showed intact anastomosis without leak [Figure 6].

The chest drain was then removed, and oral feeds were commenced.

The patient had occasional respiratory distress while feeding, which was suspected to be due to tracheomalacia. This was managed conservatively using repeated saline and steroid nebulisation.

He was discharged on post-operative day 12 on full feeds. The patient has had up to eight clinic visits; at the last visit, he is 1 year and 2 months old with a weight of 11.2 kg and currently has no complications.

Case 2

A 2-h-old pre-term female neonate delivered at 34 weeks corrected gestational age (the Ballard score) presented with

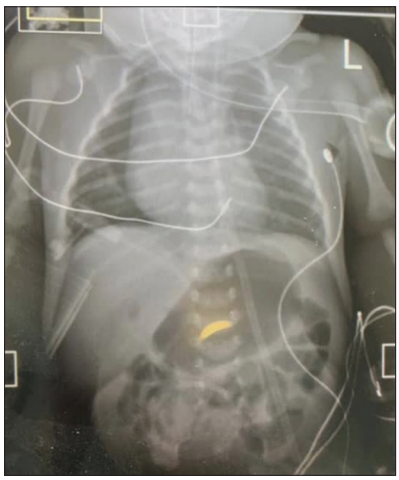


Figure 2: Re-expanded right lung (inpatient X-ray from Evercare Hospital Lekki)

a prenatal ultrasound of severe polyhydramnios and absence of stomach bubble suggestive of OA with or without TOF. Weight on admission was 1.65 kg. She also had micrognathia and low-set ears. She was immediately admitted to the neonatal intensive care and placed on continuous suctioning of the upper pouch using a size 8 Fr Replogle tube, prophylactic antibiotics and glucose-containing intravenous fluids. She was also noted to have recurrent apnoeic episodes which necessitated elective mechanical ventilation. A plain X-ray done showed a spontaneous pneumoperitoneum (possibly resulting from gastric perforation secondary to vigorous ambu-bagging from the referring centre), NG tube coiled in the upper pouch suggesting proximal OA and the presence of distal bowel gas confirming distal TOF [Figure 7].

A 2D echocardiogram done showed a patent ductus arteriosus and no other significant cardiac anomalies.

On the 2nd day of life, she was taken to the operating room following informed consent from the parents.

Surgical technique

Anaesthesia, positioning and port placement were similar to Case 1 [Figure 3]. The distal fistula was located at the level of the carina, and the distance between the two oesophageal ends was about three vertebral bodies [Figure 8].

A limited dissection of the distal fistula was done and was ligated flush with the trachea [Figure 8]. The upper pouch was mobilised up to the root of the neck, and using the sliding knot technique, the two ends were approximated with 4/0 PDS sutures [Figure 9].

The anastomosis was done in a standard fashion using an interrupted 4/0 PDS suture over a size 8 Fr NG tube. A chest drain was placed, and the wounds were closed in a standard fashion.

On the 1st day post-operation, due to the worsening of the pneumoperitoneum, a bedside peritoneal drain was placed, and effluents were noted to be serous only.



Figure 3: Semi-prone position with the right side elevated. The patient is placed transversely at the head of the table. Black circle o 5 mm port at the 4th intercostal space below the tip of the scapula, white circles ◎; 3 mm ports at the 2nd intercostal space and 6th intercostal spaces posterior axillary line. Blue drawing showing the position of the trachea, and the red drawing showing the anticipated anatomy of the OA and TOF (intra-operative picture from Evercare Hospital Lekki). OA: Oesophageal atresia, TOF: Tracheoesophageal fistula

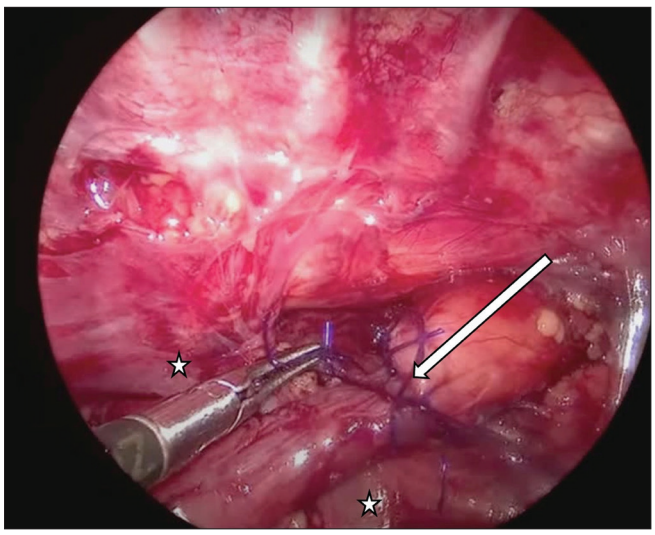


Figure 5: Completed anastomosis (white arrow) with the azygous vein (white stars) preserved (intra-operative picture from Evercare Hospital Lekki)

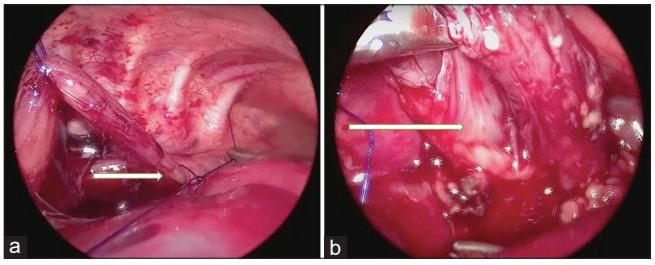


Figure 4: (a) A hitch stitch to facilitate the ligation of the fistula (white arrow) flush with the trachea; (b) (white arrow) mobilisation of the upper pouch (intra-operative pictures from Evercare Hospital Lekki)

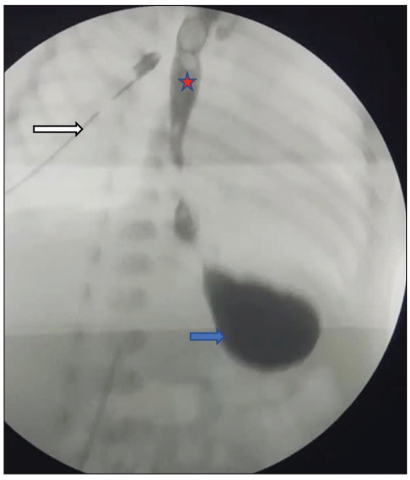


Figure 6: Barium Swallow; red star (oesophagus), white arrow (chest tube), stomach (blue arrow). (inpatient X-ray picture from Evercare Hospital Lekki)

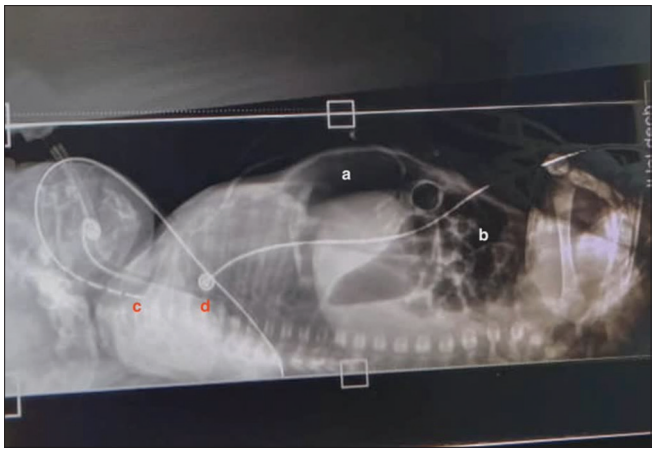


Figure 7: a: Pneumoperitoneum; b: bowel gas; c: Replogle tube in the upper pouch; d: endotracheal tube (inpatient X-ray picture from Evercare Hospital Lekki)

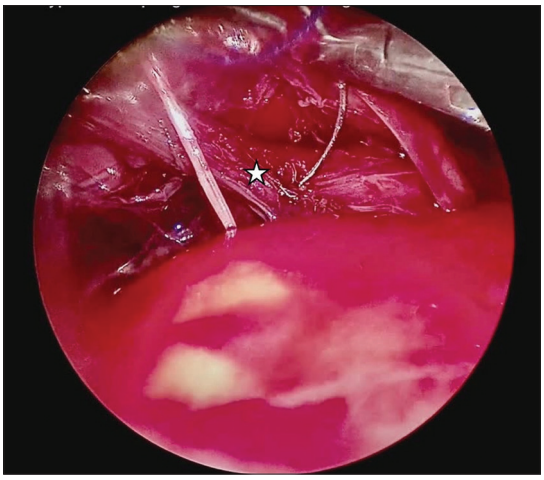


Figure 8: (White star) ligating distal fistula (intra-operative picture from Evercare Hospital Lekki)

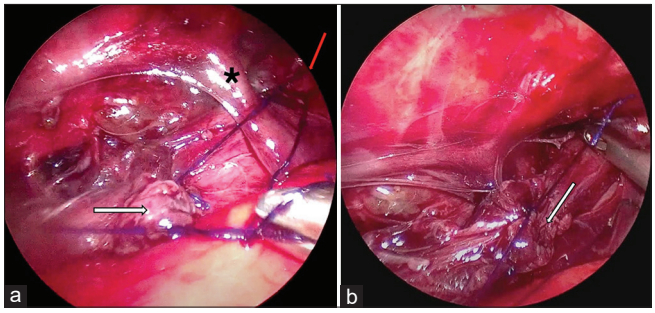


Figure 9: (a) Using sliding knot to approximate the proximal (red arrow) and distal (white arrow) oesophagus about 2 vertebra bodies apart with the azygous vein (black Asterix) intact, (b) completed anastomosis (white arrow); (intra-operative pictures from Evercare Hospital Lekki)

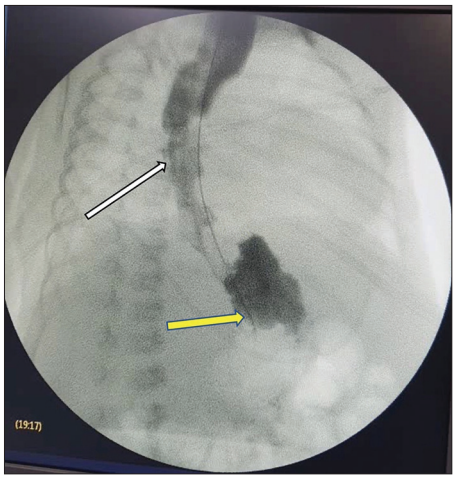


Figure 10: Barium swallow showing the normal movement of contrast from the oesophagus (white arrow) to the stomach (yellow arrow) without anastomotic leak (inpatient X-ray picture from Evercare Hospital Lekki)

Mechanical ventilation was maintained electively for about 7 days after surgery to allow for adequate healing, taking note of the significant tension of the anastomosis.

The peritoneal drain was removed on post-operative day 5, and there was no recurrence of pneumoperitoneum and no evidence of peritonitis or intestinal obstruction.

On the 10th day, a barium swallow was done under fluoroscopy, which confirmed oesophageal continuity without anastomotic leak [Figure 10].

The chest drain was then removed, and oral feeding was started. NG tube feeding was, however, maintained for up to 20 days, and oral feeds were gradually increased as tolerated.

Other post-operative challenges included recurrent apnoeic episodes requiring resuscitation and placement on intranasal oxygen.

The patient was discharged home on the post-operative day 32.

The patient has had four clinic visits, weighing 3.5 kg at the last clinic visit at the age of 3 months and tolerating feeds.

Discussion

Since the first thoracoscopic repair of OA was described by Dr. Steven Rothenberg and his team in 2000, there seems to be a paradigm shift in the approach of OA and TOF in developed countries. In 2005, the first series and multicentre study were reported.[4] The study noted a wide adoption of the technique by several centres and a significant success rate with only a few conversions to open thoracotomy. The study also noted that even patients with other major anomalies, including cardiac anomalies, successfully underwent thoracoscopy.[4]

Since then, several centres, especially in developed climes, have adopted thoracoscopic technique, and the advantages have since become more obvious, including better visibility for both surgeon and assistant, and elimination of long-term musculoskeletal deformity associated with thoracotomies.[5]

In a bid to reduce post-operative morbidity associated with lateral thoracotomies, Bianchi *et al*.,[6] in 1991, documented the

use of axillary skin crease incision for the repair of OA with TOF and patients with patent ductus arteriosus with excellent outcomes vis-a-vis excellent aesthetics and attendant reduced post-operative chest wall morbidity. This technique has since been adopted by some surgeons and has shown comparable surgical outcomes with lateral thoracotomies while achieving better cosmesis and reduced chest wall morbidity. However, this technique is still limited by poor visibility of the operating field as is the case with lateral thoracotomies.

Comparative studies, done by Borruto *et al*.[5] and Yang *et al*.,[7] confirmed the feasibility of the thoracoscopic technique and noted the reduced morbidity relating to the chest wall incisions.

In the West African subregion, the current practice has been open thoracotomy for primary repair. In fact, a majority of patients still have cervical oesophagostomy with distal oesophageal banding and gastrostomy.[3,8,9] They are subsequently planned for oesophageal replacement at a later date for those who survive. In 2014, Bode and Ademuyiwa.,[8] while describing a reverse gastric tube oesophageal substitution for the staged repair of OA and TOF, concluded that staged repair may be the only hope of survival of these patients in our setting, where patients typically present late and with inadequate support facilities such as neonatal intensive care and parenteral nutrition. However, comparative studies have demonstrated no significant improvement in mortality rate following staged or primary repair but attributed the high mortality rate to inadequate perioperative patient care due to a lack of appropriate facilities and trained neonatal critical care personnel.[3,9]

The cases presented are the first two cases in our facility, and to the best of our knowledge, the first thoracoscopic repair was done in West Africa. Based on the prognostic criteria and classification, both fell into Waterson class C due to severe pneumonia and atelectasis in Case 1 and pre-maturity with low birth weight in Case 2. Based on this classification, in our setting, these patients are not candidates for primary repair; however, we were able to offer them primary thoracoscopic repair with good outcomes. This was only possible due to extreme collaboration with neonatologists, neonatal intensivists and anaesthesiologists and a well-equipped NICU for both pre- and post-operative management.

In both cases, the availability of mechanical ventilation was a key to their survival due to the peculiarities of the patients at the time of presentation. Case 1 had aspiration pneumonitis with right atelectasis and subsequent post-operative pneumonia, while Case 2 had several episodes of apnoea of pre-maturity, pneumoperitoneum and a long gap with anastomosis under significant tension; hence, mechanical ventilation was indicated in both.

For both cases, the azygous vein was preserved, and anastomosis was done above it; this is because the surgeon believes that the azygous vein creates a natural separation between the fistula and the anastomosis, thus reducing the need to place any synthetic material or pleural flap to reduce the risk of anastomotic leak or recurrent fistulisation. Sharma *et al*.,[10] in 2007, suggested that preservation of the azygos vein may prevent early post-operative oedema of the oesophageal anastomosis by maintaining the venous drainage and thus may form an additional protective factor against anastomotic leaks.

Conclusion

We, hereby, conclude that thoracoscopic repair of OA and TOF is feasible and safe in developing countries given appropriate infrastructure, expertise and teamwork. It is rapidly becoming the gold standard for the management of all forms of OA and TOF in many institutions in the developed world, including the long-gap anomalies. Our index cases have demonstrated that mechanical ventilation is critical in reducing the morbidity and mortality associated with OA management in high-risk children.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Rothenberg SS. Esophageal Atresia and Tracheoesophageal Fistula Malformations. In: Holcomb III GW, Murphy JP, St Peter SD. Holcomb and Ashcraft's Pediatric Surgery. 7th Edition: Elsevier Health Sciences; Edinburgh. 2019. p. 437-58.
2. Nwosu JN, Onyekwulu FA. Oesophageal atresia and tracheooesophageal fistula: A 12 years experience in a developing nation. Niger J Med 2013;22:295-8.
3. Adebo OA. Oesophageal atresia and tracheo-oesophageal fistula: Review of a 10-year personal experience. West Afr J Med 1990;9:164-9.
4. Holcomb GW 3rd, Rothenberg SS, Bax KM, Martinez-Ferro M, Albanese CT, Ostlie DJ, *et al.* Thoracoscopic repair of esophageal atresia and tracheoesophageal fistula: A multi-institutional analysis. Ann Surg 2005;242:422-8.
5. Borruto FA, Impellizzeri P, Montalto AS, Antonuccio P, Santacaterina E, Scalfari G, *et al.* Thoracoscopy versus thoracotomy for esophageal atresia and tracheoesophageal fistula repair: Review of the literature and meta-analysis. Eur J Pediatr Surg 2012;22:415-9.
6. Bianchi A, Sowande O, Alizai NK, Rampersad B. Aesthetics and lateral thoracotomy in the neonate. J Pediatr Surg 1998;33:1798-800.
7. Yang S, Wang P, Yang Z, Li S, Liao J, Hua K, *et al.* Clinical comparison between thoracoscopic and thoracotomy repair of gross type C esophageal atresia. BMC Surg 2021;21:403.
8. Bode CO, Ademuyiwa AO. Reverse gastric tube oesophageal substitution for staged repair of oesophageal atresia and tracheo-oesophageal fistula. Afr J Paediatr Surg 2014;11:366-70.
9. Osei-Nketiah S, Hesse AA, Appeadu-Mensah W, Glover-Addy H, Etwire VK, Sarpong P. Management of oesophageal atresia in a developing country: Is primary repair forbidden? Afr J Paediatr Surg 2016;13:114-9.
10. Sharma S, Sinha SK, Rawat JD, Wakhlu A, Kureel SN, Tandon R. Azygos vein preservation in primary repair of esophageal atresia with tracheoesophageal fistula. Pediatr Surg Int 2007;23:1215-8.