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Redo Thoracotomy and Esophageal Re-Anastomosis for Management of Complex Anastomotic Esophageal Stricture in an Operated Case of Esophageal Atresia with Tracheoesophageal Fistula

Minakshi Bhosale[1]*, Vegunta Harshendra[2], Pooja Patat[2], Raghuveer Pratap[2], Shivaji Mulgir[3]

[1]Professor & HOD, (MS, DNB, MCh - Paediatric Surgery), Department of Paediatric Surgery, B J Government Medical College; PUNE - 411 001
[2]MCh Resident, (MS - General Surgery), Department of Paediatric Surgery, B J Government Medical College; PUNE - 411 001
[3]Assistant Professor, (DNB, MCh - Paediatric Surgery), Department of Paediatric Surgery, B J Government Medical College; PUNE - 411 001

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***Corresponding author:** Minakshi Bhosale
Professor & HOD, (MS, DNB, MCh - Paediatric Surgery), Department of Paediatric Surgery, B J Government Medical College; PUNE - 411 001

Abstract | **Case Report**

Anastomotic esophageal stricture (AES) is among the most frequent complications after repair of esophageal atresia (EA) with tracheoesophageal fistula (TEF), occurring in 30-60 % of children. Presentation in a patient with AES causes significant clinical concern, substantially affecting quality of life of the child as well as the care-givers. We report case of a toddler who presented at the age of fourteen months with symptoms secondary to complex AES following repair of EA with TEF in the neonatal period. She had complete dysphagia both for solids as well as liquids. After preoperative optimization, she underwent redo thoracotomy and end-to-oblique esophageal re-anastomosis. Postoperative course was uneventful. Timely intervention enabled preservation of the native esophagus and complete functional recovery.

Keywords: Anastomotic Esophageal Stricture (AES), Redo Thoracotomy, Esophageal Atresia (EA), Tracheoesophageal Fistula (TEF), Dysphagia, Gastroesophageal Reflux Disease (GERD), Anastomotic Leak.

INTRODUCTION

Esophageal atresia (EA) with Tracheoesophageal Fistula (TEF) is a common congenital digestive tract anomaly occurring in approximately 1 in 2,500 to 1 in 4,500 live births worldwide [1,2]. It requires surgical repair in the neonatal period. Improved surgical techniques, as well as advances in preoperative and postoperative neonatal intensive care, have raised overall survival rates to over 90 to 95%. However, post-repair management is frequently complicated by lifelong gastrointestinal and respiratory morbidity [1-3].

Surgical repair of EA with TEF primarily involves division of fistula and primary end-to-end esophageal anastomosis. Anastomotic esophageal stricture (AES) is the most frequent late structural complication, with an incidence ranging from 30 to 60% [1,2,3]. Development of AES is multifactorial, but mainly attributed to tension on the anastomosis, gastroesophageal reflux disease (GERD) and anastomotic leak in early postoperative period [4]. Decreased luminal diameter from AES leads to postprandial esophageal stasis, feeding intolerance, recurrent tracheobronchial aspiration, failure to thrive, and severe lower respiratory tract infections (LRTIs) [1,2,5,6].

This manuscript presents a clinical case report including the presentation, preoperative optimization, surgical approach, and outcome of a toddler with complex AES.

CASE PRESENTATION

A fourteen-month-old female child presented to our hospital, a tertiary care center in Maharashtra, India with progressive dysphagia, postprandial non-bilious vomiting, recurrent bouts of aspiration, and LRTIs. She was second born, delivered by normal delivery at term and had birth weight of 3.2 Kg. Antenatal history was

non-contributory except for diagnosis of polyhydramnios at 28 weeks gestational age. Baby had frothing after birth and was diagnosed to have Gross Type C EA with TEF. She underwent primary repair of tracheoesophageal fistula via right thoracotomy on day of Life 3 at a district hospital. Baby received post-operative care in NICU. Dye study done on pod 7 was suggestive of leak which was managed conservatively. Initial NGT feeds were increased progressively to oral feeds. Baby was discharged at the age of one and half months when she was on full feeds. Feeding in propped up position and recommended prophylactic PPI therapy (such as Lansoprazole) was not advised upon discharge. Baby was symptom-free until 6 months of age, when she started having bouts of cough during feeds and non-bilious vomiting after feeds. Symptoms worsened to complete dysphagia both for solids and liquids over period of time. She required multiple hospitalizations for treatment of aspiration pneumonia. On presentation to our hospital her general condition was moderate. She was conscious but irritable. She was dehydrated, undernourished and had weight of 5.8 kg. Her HR was 116/ min, RR was 28/min and SPO2 was 94% on room air. Auscultation revealed bilateral wheezing and reduced air entry on the right side. A healed right posterolateral thoracotomy scar was visible.

Her hemoglobin was 8.5 gm, WBC Count was15,400/cmm, platelet count was 260,000/cmm

RFT, LFT, and coagulation panels were normal. Chest radiograph revealed right upper lobe atelectasis (collapse) with consolidation. Bilateral patchy perihilar and interstitial air-space opacities were noted and were more prominent on the right side.

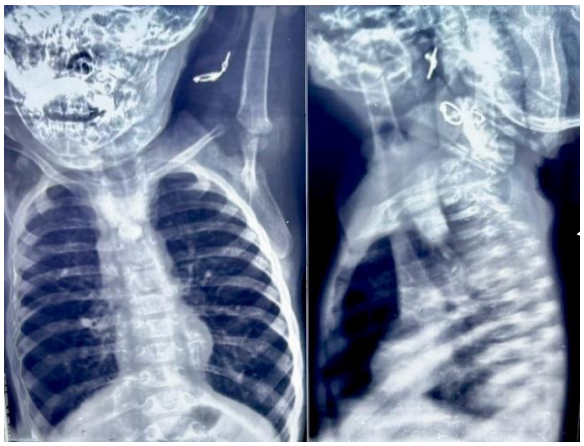

Fig. 1: Contrast esophagogram (AP and lateral View) showing dilated upper esophagus with stricture at T_{3-4} level

An upper GI contrast esophagogram revealed dilated upper esophagus with severe narrowing at T3/T4 level, suggestive of a high-grade anastomotic stricture at prior anastomotic site (Fig 1). Child was initially posted for endoscopic dilatation under anesthesia. However, the smallest available video-endoscope (Fujifilm EG-580NW2, distal end diameter 5.8 mm) or even the smallest guidewire could not be negotiated across the stricture confirming the diagnosis of a complex stricture.

Persistent feeding intolerance with recurrent vomiting over a prolonged period (nearly 6-8 months) had caused nutritional impairment and failure to thrive. Intravenous fluid infusion was started for correction of dehydration. Proton pump inhibitor therapy was initiated. Intravenous Augmentin and oral Tamiflu was given for management of aspiration pneumonia as per advice of the pediatrician. Blood transfusion was given for correction of anemia. Total parenteral nutrition was given for pre-operative nutritional optimization. Six-hourly nebulization and aggressive chest physiotherapy were initiated. After pre-op nutritional rehabilitation and weight gain to 1.6 kg, surgical intervention was planned as per existing ESPGHAN and NASPGHAN Guidelines. Redo right posterolateral thoracotomy was done entering the thoracic cavity via 5th intercostal space (intra-pleural approach), going through a virgin area just below the prior scar site, deliberately avoiding entry through the prior incision. Yet there was significant scarring and extensive tissue adhesions, making mobilization of the esophagus extremely difficult. It resulted in blood loss of approximately 100 cc which was replaced on table. Upper esophageal pouch was 10-12 mm in diameter; whereas the diameter of lower esophageal pouch was 6-7 mm. Stricture segment was 5-6 mm long with extensive, dense fibrotic scar tissue surrounding the old anastomotic site causing marked luminal occlusion (Fig 2). No attempt was made to excise the dense fibrotic stricture segment since it was closely adherent to the right bronchus. Adjacent healthy, esophageal tissue was mobilized, leaving the fibrotic stricture segment in-situ. Lower pouch was obliquely cut distal to the stricture. A tension-free single-layer end-to-oblique esophageal anastomosis was performed using 4-0 Polyglactin, simple, interrupted sutures over nasally placed trans-anastomotic no 9 infant feeding tube (IFT) taking care to include the mucosa in every suture (Fig 3). Incision was closed in layers after insertion of no 16 G Romson's intercostal drain tube. Operative time was 4 hours. Patient tolerated the procedure well, was extubated and was put on BCPAP O2 supplementation for first 24 hours, which was gradually tapered. IV Meropenem was

given for 3 weeks post-operatively. Nebulization and aggressive chest physiotherapy were restarted. Anti-reflux therapy i.e., Lansoprazole was started through NGT from early post-operative period. On post-operative day 4 enteral nutrition via the trans-anastomotic IFT was initiated and gradually advanced to full feeds by post-operative day 6. Contrast esophagography done on post-operative day 10 confirmed luminal continuity without obvious anastomotic leak (Fig 4). Oral feeds were re-introduced in upright position and gradually advanced. There was no vomiting, choking, or dyspnea by dysphagia. The trans-anastomotic tube was removed on post-operative day 11. ICD was removed on post- operative day 12. Her weight was 8.2 Kg at the time of discharge. She was discharged on oral antibiotics, multivitamins and PPI (Lansoprazole) supplementation with advice to continue the PPI (Lansoprazole) supplementation at least for 6 months post-operatively. Small, frequent, thickened feeds in an upright position were advised.

The child is on regular follow up. Her PPI supplementation was discontinued after 6 months. Follow-up evaluation at 1 year demonstrated complete resolution of her symptoms. The patient is tolerating both liquids and solid diet without dysphagia or cough. Growth curves demonstrate sustained weight gain.(Fig2)

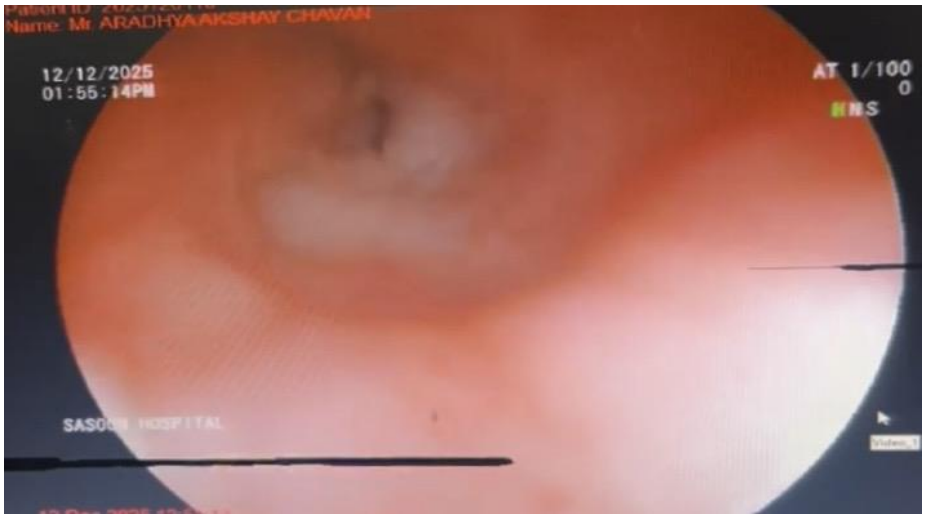

Fig. 2: Anastomotic stricture causing marked luminal occlusion

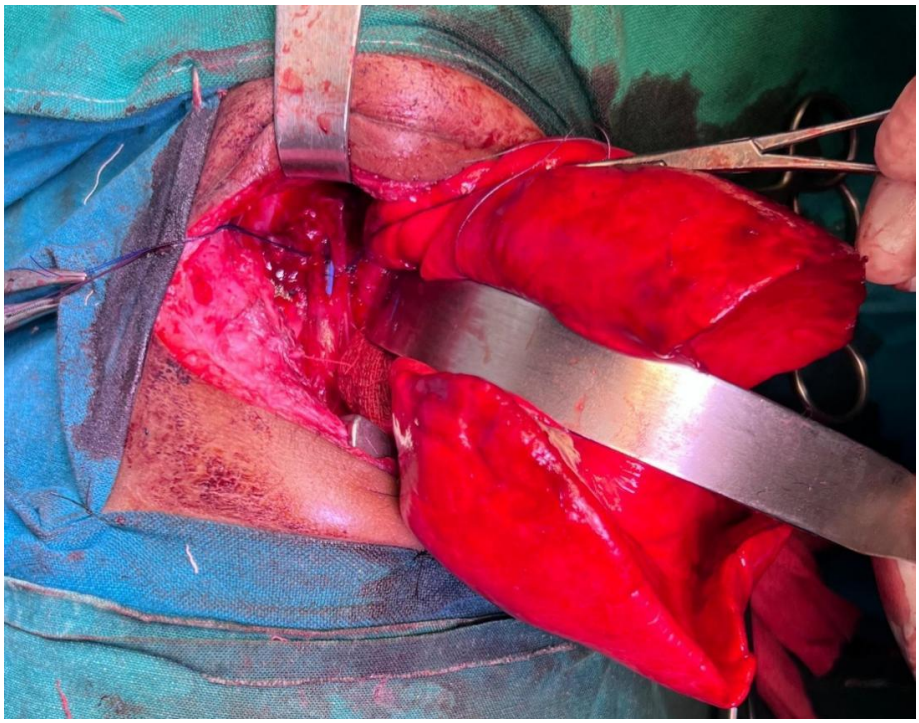

Fig. 3: Intra-operative photograph showing infant feeding tube passing across the esophagus after completion of posterior layer of anastomosis

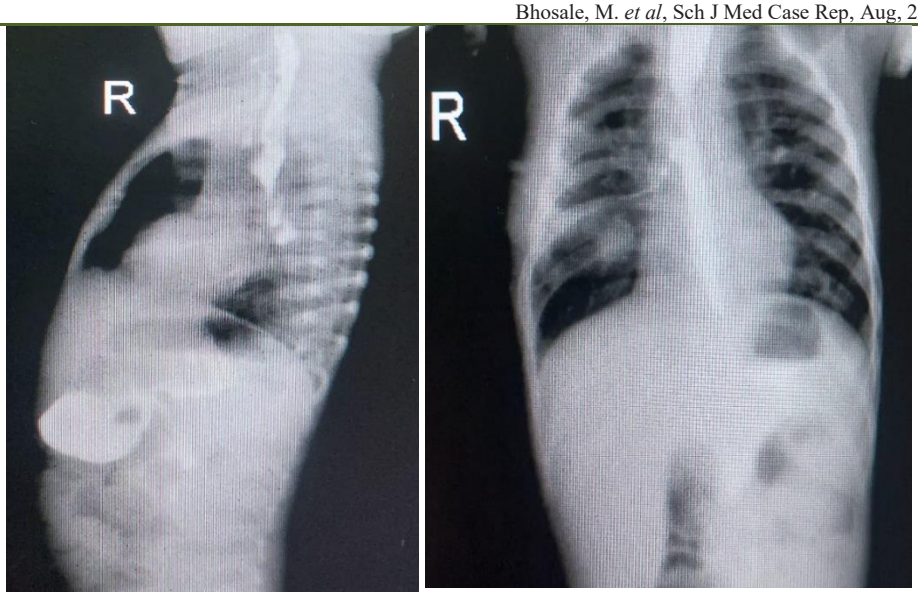

Fig. 4: Post-operative dye study suggestive of luminal continuity. Contrast is seen entering the stomach

DISCUSSION

AES after primary repair of EA and TEF remains a major management challenge [1–3]. Expertise of the operating surgeon, suture material used, type of anastomosis, tension on the anastomosis, degree of ischemia, anastomotic leak, and presence of GERD are the main factors related to development of AES. minimally invasive endoscopic interventions, including balloon or bougie dilation, stent placement, incisional therapy, intralesional steroid injection, and topical application of Mitomycin C are initial treatment modalities of choice [7]. However, complex strictures with thick, mature fibrotic rings do not respond to dilations and require surgical intervention [8,9,10].

Our case presented at the age of fourteen-months with cough during feeds, dysphagia, recurrent episodes of aspiration pneumonia requiring hospitalizations, and failure to thrive. Her symptoms had progressively increased over period of 6-8 months. This presentation is consistent with the typical clinical course of post-EA anastomotic strictures. She had complete dysphagia for solids for over 6 months and also for liquids since last 1 month indicating near total luminal obstruction. Contrast esophagogram showed dilated upper esophagus with abrupt cut off at T3/T4 level. Her AES was very dense and even the smallest sized endoscope or it's guidewire couldn't be negotiated. Also, since length of the stricture could not be assessed at this stage, the exact surgical plan could not be decided. We had to go in with the plan to perform esophageal replacement with an interposition graft if length of stricture was long or if adequate mobilization failed. Hence, surgery was scheduled after aggressive pre-operative optimization over 3-4 weeks, so that the child who had respiratory and nutritional compromise would tolerate the supramajor surgery under general anesthesia. Our patient underwent redo right thoracotomy, and end-to-oblique esophageal re-anastomosis. Since the stricture segment was fibrotic and closely adherent to the right bronchus, it was deliberately left in-situ. Esophageal preservation and complete functional recovery was possible in our case.

This child had undergone primary repair of EA with TEF in the neonatal period at a district hospital where the anastomosis was probably done by untrained personnel. She had prolonged anastomotic leak in the post-operative period almost for 4-6 weeks. Moreover, she was not advised any PPI therapy on discharge from the hospital. Improper surgical technique, inadequate mobilization, tension on the anastomosis, anastomotic leak, and unmanaged gastroesophageal reflux disease (GERD) might be the predisposing factors for stricture formation in this child. The dense fibrotic stricture had resulted in profound nutritional failure and respiratory compromise requiring, aggressive preoperative stabilization.

CONCLUSION

AES is a significant post-repair complication of EA and TEF. When stricture is dense or refractory to conservative measures, leaving the fibrotic stricture segment in-situ and end-to-oblique re-anastomosis of mobilized esophageal pouches is a safe and curative treatment modality. It enables esophageal preservation and complete functional recovery. Aggressive preoperative nutritional optimization and correction of respiratory infection is of paramount importance.

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Conflict of interest - None to declare

Contribution Details:

1) Contributor 1: Conception and design, critical revision and final approval of the manuscript

2) Contributor 2: Acquisition and interpretation of data, drafting of manuscript
3) Contributor 3: Conception and design, drafting of the manuscript
4) Contributor 1: Conception and design, drafting of the manuscript
5) Contributor 1: Acquisition and interpretation of data, drafting of manuscript

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