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Chest tube-related complications after a staged thoracoscopic repair of a long-gap esophageal atresia: A case report

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ABSTRACT

Background: Long-gap esophageal atresia remains a challenging condition. There are no standard guidelines on the optimal surgical approach or the optimal management strategy for the potential postoperative complications. We report a case of long-gap EA treated by staged thoracoscopic repair in which the chest drain caused a complication.

Case report: A female with prenatal diagnosis of polyhydramnios and a small stomach suspicious for esophageal atresia was delivered by cesarean section at 33 weeks of gestation. The orogastric tube remained coiled in the upper esophagus, confirming the diagnosis of esophageal atresia. X-rays showed a gasless abdomen, suspicious for a lack of a distal tracheo-esophageal fistula. On the second day of life, the patient underwent a tracheoscopy, which confirmed the absence of a tracheo-esophageal fistula. A gastrostomy was done at the same time. At 1 month of age the patient underwent a fluoroscopic study, and the esophageal gap was found to be 3.5 vertebral bodies. At 2 months of age she underwent a thoracoscopic placement of internal traction sutures. Five days later she underwent a thoracoscopic esophageal anastomosis, which was under moderate tension. A chest tube was left in place. Seven days after the operation a routine esophagram showed that the chest tube had slipped into the esophagus through the anastomosis. We pulled the chest tube back under direct endoscopic vision. There was also a tight stenosis at the anastomosis. We placed a *trans*-anastomotic tube for feedings and a naso-esophageal tube to suction the secretions of the upper esophagus. Forty-for days later we repeated the esophagram and confirmed the persistence of the tight stenosis. The patient underwent serial dilations every 2 weeks for 3 months and a Nissen fundoplication, after which the stricture completely resolved.

Conclusion: Although rare, the chest tubes used after the repair of an esophageal atresia can cause complication at the anastomotic site.

1. Introduction

The treatment of esophageal atresia (EA) has improved over the years, reaching a survival rate of 90–95 % [1]. However, treating long-gap esophageal atresia (LGEA) remains challenging due to its infrequent occurrence, the lack of standardized surgical guidelines, and its high morbidity, and the high incidence of complications [2]. While esophageal traction via external or intrathoracic stitches has shown promising outcomes, there is a lack of conclusive data [3–5]. The most frequent complication is the stenosis of the anastomosis, which occurs in 50 %–80 % of cases of LGEA repair. Factors that contribute to the development of a stenosis are tension

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on the anastomosis, ischemia at the ends of the esophageal pouches, a 2-layer anastomosis, the use of silk stitches, the occurrence of an anastomotic leak, the presence of gastroesophageal reflux [6].

We report a case of pure long-gap EA treated by thoracoscopic internal traction and delayed anastomosis that developed an unexpected complication.

This manuscript was prepared following the CARE guidelines.

2. Case report

A 30-year-old woman having an in vitro fertilization with embryo transfer was referred to our center at 21 weeks of gestation after a routine prenatal ultrasound had found a single umbilical artery, a small stomach, and polyhydramnios, which in combination were suspicious for esophageal atresia. A fetal magnetic resonance done at 33 weeks of gestation confirmed all the findings. The fetus was delivered by cesarean section at 33 + 6 weeks of gestation due to a bleeding placenta previa. The newborn female had a birth weight of 2180 g. Plain films of the chest and abdomen showed the naso-gastric tube coiled in the upper esophagus at the level of the second intercostal space, and a gasless abdomen. These findings were suspicious for a pure esophageal atresia, or an esophageal atresia with a proximal fistula. On day of life 2 after we did a tracheoscopy and confirmed that there was no fistula of any type, we created a gastrostomy at the same time. We kept a naso-esophageal tube to suction continuously the secretions accumulated in the upper pouch and started enteral feedings through the gastrostomy. At one month of age, we did a contrast study to measure the esophageal gap, which was still long (3.5 vertebral bodies; Fig. 1). At 2 months of age, she underwent a thoracoscopic placement of internal traction sutures according to the technique described by Patkowski [4]. Five days later we did a thoracoscopic end-to-end anastomosis under moderate tension and left a nasogastric *trans*-anastomotic tube and a chest tube. On postoperative day seven we did a routine contrast esophagram which showed that the chest tube had slipped into the esophagus through the anastomosis. The contrast was being collected by the chest tube, and no contrast was reaching the lower esophagus (Fig. 2). An esophagoscopy was done to pull back the chest tube under direct view. We could see with the esophagoscope that the anastomosis had developed a tight stenosis. We placed a *trans*-anastomotic tube to bypass the stenosis, and placed a naso-esophageal tube to continuously suction the upper esophagus. We placed a new chest tube under fluoroscopy and left it in place for 5 days. On postoperative day 44 we did an esophagoscopy and confirmed that there was still a tight stenosis. We did a dilatation at that time up to a 24 Fr size. We did subsequent serial dilatations, every 2 weeks, for 3 months, but the stenosis was still present. We then decided to do a Nissen fundoplication, after which the stenosis completely resolved (Fig. 3). She has not needed further dilatations afterwards.

3. Discussion

Different procedures such as gastric pull-up, gastric tube reconstruction, primary anastomosis, and esophageal replacement, have been tried over the last century to treat pure long-gap EA. Each of the techniques has its own set of potential complications and associated morbidities, and none of them is ideal. A delayed anastomosis was introduced as an option when it became apparent that in most cases the two esophageal pouches approximate spontaneously in 75 % of cases throughout the first 3 months of life. An international survey conducted in 2014 revealed that the most favored technique for long-gap EA was, in fact, a delayed primary anastomosis [7]. More recently, Patkowski et al. described the use of internal traction sutures followed by an anastomosis 5–9 days later and showed that in 85 % of cases of long-gap EA a primary anastomosis was possible [4,7,8]. This is the technique that we used in our case. While

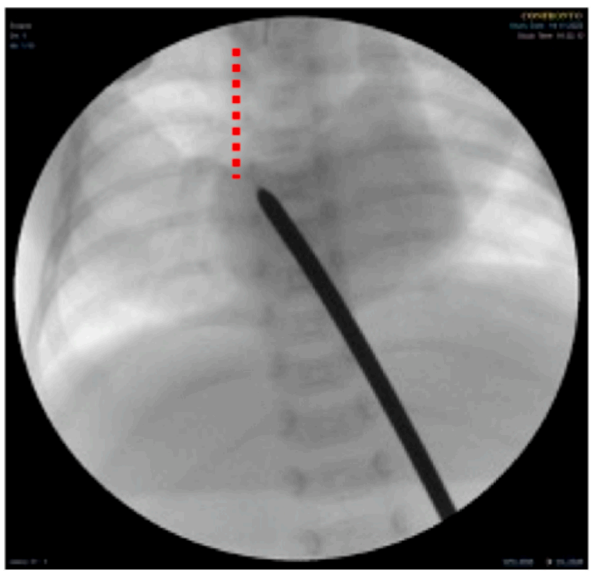

Fig. 1. A fluoroscopic study after 1 month of life showed that there was still a considerable gap between the two esophageal ends (red dotted line).

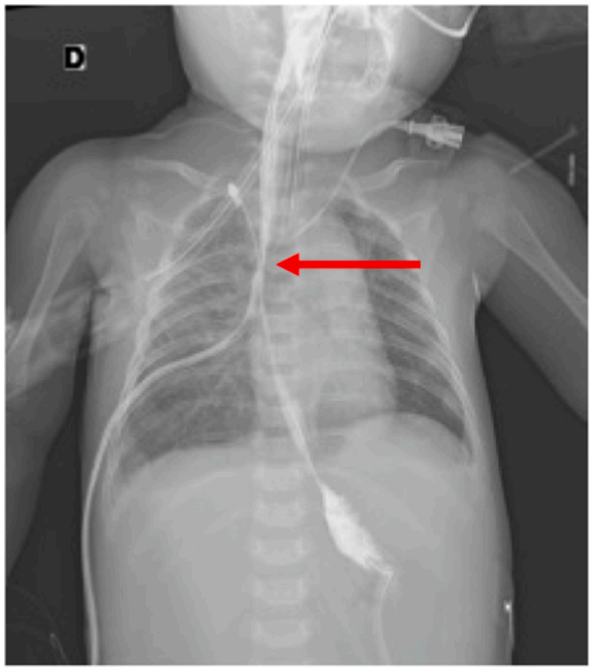

Fig. 2. Fluoroscopic contrast study on postoperative day 7 shows the thoracic drain inside the anastomosis (red arrow).

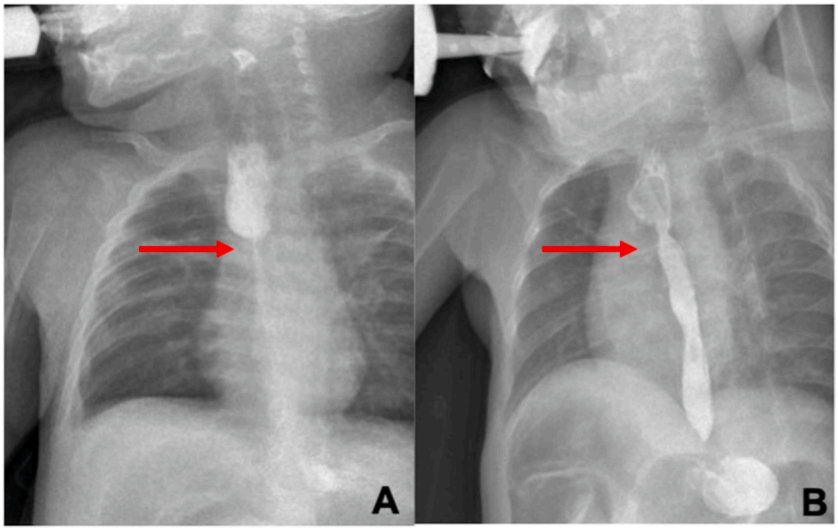

Fig. 3. Anastomotic stenosis: before dilations (A) and after dilation and Nissen fundoplication (B) (red arrows).

trans-anastomotic tubes are left by most pediatric surgeons regardless of the length of the gap, there is still debate regarding the benefits of leaving a prophylactic chest tube [9]. Recent reports showed that leaving a chest tube did not improve outcomes in patients with esophageal atresia but instead was associated with a longer length of stay and a higher rate of esophageal stricture formation. Chest tubes have been associated with a variety of complications [10–16]. The complication caused by the chest tube in our case (dehiscence and stricture of the anastomosis) is remarkably rare, and to the best of our knowledge has only been reported once [15].

Conservative management for anastomotic leaks is the preferred approach and includes chest-tube drainage, total parenteral nutrition, and broad-spectrum antibiotics [16–19]. It is well known that the occurrence of an anastomotic leak increases the likelihood of a subsequent anastomotic stenosis [20]. The most common treatment for anastomotic strictures is balloon dilation, although semi-rigid dilators are considered equally safe [6,19]. The first dilatation should be done at least 3 weeks after the anastomosis due to the increased risk of esophageal perforation. The interval between dilatations is variable in the literature, ranging from once per week to once per month [21]. Lastly, there is abundant evidence that patients with esophageal strictures benefit from anti-reflux procedures because the acid reflux plays a key role in the persistence of stenosis [6,22].

4. Conclusion

Although rare, the chest tube routinely used after the repair of an esophageal atresia can cause severe complications such as dehiscence and stenosis of the anastomosis.

5. Patient consent

"Consent to publish the case report was not obtained. This report does not contain any personal information that could lead to the identification of the patient."

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6. Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

CRediT authorship contribution statement

Donatella Di Fabrizio: Writing – review & editing, Writing – original draft. **Edoardo Bindi:** Visualization, Supervision, Conceptualization. **Alba Cruccetti:** Visualization. **Giovanni Cobellis:** Validation, Supervision, Conceptualization.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

References

[1] Sulkowski J.P, Cooper J.N, Lopez J.J, Jadcherla Y, Cuenot A, Mattei P, et al. Morbidity and mortality in patients with esophageal atresia. In: Surgery (United States). Mosby Inc.; 2014. p. 483–91.
[2] Bourg A, Gottrand F, Parmentier B, Thomas J, Lehn A, Piolat C, Bonnard A, Sfeir R, Lienard J, Rousseau V, Pouzac M, Liard A, Buisson P, Haffreingue A, David L, Branchereau S, Carcauzon V, Kalfa N, Leclair M.D, Lardy H, Irtan S, Varlet F, Gelas T, Potop D, Auger-Hunault M. Outcome of long gap esophageal atresia at 6 years: a prospective case control cohort study. J Pediatr Surg 2023 Apr;58(4):747–55. https://doi.org/10.1016/j.jpedsurg.2022.07.023. Epub 2022 Aug 1. PMID: 35970676.
[3] Foker J.E, Linden B.C, Boyle E.M, Marquardt C. Development of a true primary repair for the full spectrum of esophageal atresia. Ann Surg 1997;226:533–41.
[4] Bogusz B, Patkowski D, Gerus S, Rasiewicz M, Górecki W. Staged thoracoscopic repair of long-gap esophageal atresia without temporary gastrostomy. J Laparoendosc Adv Surg Tech 2018 Dec;28(12):1510–2. https://doi.org/10.1089/lap.2018.0188. Epub 2018 Jul 17. PMID: 30016196.
[5] Zani A, Cobellis G, Wolinska J, Chiu P.P, Pierro A. Preservation of native esophagus in infants with pure esophageal atresia has good long-term outcomes despite significant postoperative morbidity. Pediatr Surg Int 2016 Feb;32(2):113–7. https://doi.org/10.1007/s00383-015-3821-x. Epub 2015 Oct 31. PMID: 26520653.
[6] Manfredi M.A. Endoscopic management of anastomotic esophageal strictures secondary to esophageal atresia. Gastrointest Endosc Clin N Am 2016 Jan;26(1):201–19. https://doi.org/10.1016/j.giec.2015.09.002. PMID: 26616905.
[7] Zani A, Eaton S, Hoellwarth M.E, Puri P, Tovar J, Fasching G, Bagolan P, Lukac M, Wijnen R, Kuebler J.F, Cecchetto G, Rintala R, Pierro A. International survey on the management of esophageal atresia. Eur J Pediatr Surg 2014 Feb;24(1):3–8. https://doi.org/10.1055/s-0033-1350058. Epub 2013 Aug 9. PMID: 23934626.
[8] Borselle D, Davidson J, Loukogeorgakis S, De Coppi P, Patkowski D. Thoracoscopic stage internal traction repair reduces time to achieve oesophageal continuity in long gap oesophageal atresia. Eur J Pediatr Surg 2023 Dec 28. https://doi.org/10.1055/a-2235-8766. Epub ahead of print. PMID: 38154482.
[9] Sakuov Z, Dzhenalaev D, Ospanov M, Rustemov D, Lozovoy V, Erekeshov A, Otegen T, Patkowski D. Long-gap esophageal atresia repair using staged thoracoscopic internal traction: the first Kazakhstan experience. J Laparoendosc Adv Surg Tech 2022 Dec;32(12):1265–8. https://doi.org/10.1089/lap.2022.0244. Epub 2022 Nov 1. PMID: 36318795; PMCID: PMC9805844.
[10] Nguyen M.V.L, Delaplain P.T, Lim J.C, Golden J.M, Gayer C.P. The value of prophylactic chest tubes in tracheoesophageal fistula repair. Pediatr Surg Int 2020 Jun;36(6):687–96. https://doi.org/10.1007/s00383-020-04664-6. Epub 2020 May 6. PMID: 32377785.
[11] Aslanabadi S, Jamshidi M, Tubbs R.S, Shoja M.M. The role of prophylactic chest drainage in the operative management of esophageal atresia with tracheoesophageal fistula. Pediatr Surg Int 2009 Apr;25(4):365–8. https://doi.org/10.1007/s00383-009-2345-7. Epub 2009 Mar 17. PMID: 19290533.
[12] Gawad N, Wayne C, Bass J, Nasr A. A chest tube may not be needed after surgical repair of esophageal atresia and tracheoesophageal fistula. Pediatr Surg Int 2018 Sep;34(9):967–70. https://doi.org/10.1007/s00383-018-4307-4. Epub 2018 Jul 26. PMID: 30051148.
[13] Anand S, Singh A, Krishnan N, Yadav D.K. Whether prophylactic intraoperative chest drain insertion in esophageal atresia-tracheoesophageal fistula is an evidence-based practice or just a prejudice: a systematic review and meta-analysis. J Pediatr Surg 2022 Aug;57(8):1554–60. https://doi.org/10.1016/j.jpedsurg.2021.06.015. Epub 2021 Jul 8. PMID: 34284871.
[14] Fowler C.L. Subclavian artery compression from a chest tube after thoracotomy in a premature infant. Pediatr Radiol 1995;25(6):458–9.
[15] Johnson J.F, Wright D.R. Chest tube perforation of esophagus following repair of esophageal atresia. J Pediatr Surg 1990;25(12):1227–30.
[16] Bindi E, Torino G, Noviello C, Simonini A, Torre M, D'Agostino R, Cobellis G. Recurrent tracheoesophageal fistula secondary to clips migration after thoracoscopic esophageal atresia repair. Journal of Pediatric Surgery Case Reports 2020;62:101651. https://doi.org/10.1016/j.epsc.2020.101651.
[17] Dingemann C, Eaton S, Aksnes G, Bagolan P, Cross K.M, De Coppi P, Fruithof J, Gamba P, Husby S, Koivusalo A, Rasmussen L, Sfeir R, Slater G, Svensson J.F, Van der Zee D.C, Wessel L.M, Widenmann-Grolig A, Wijnen R, Ure B.M. ERNICA consensus conference on the management of patients with esophageal atresia and tracheoesophageal fistula: diagnostics, preoperative, operative, and postoperative management. Eur J Pediatr Surg 2020 Aug;30(4):326–36. https://doi.org/10.1055/s-0039-1693116. Epub 2019 Jul 2. PMID: 31266084.
[18] Zhao R, Li K, Shen C, Zheng S. The outcome of conservative treatment for anastomotic leakage after surgical repair of esophageal atresia. J Pediatr Surg 2011 Dec;46(12):2274–8. https://doi.org/10.1016/j.jpedsurg.2011.09.011. PMID: 22152864.
[19] Kulshrestha S, Kulshrestha M, Tewari V, Chaturvedi N, Goyal A, Sharma R.K, Sarkar D, Tandon J.N, Katyal V. Conservative management of major anastomotic leaks occurring after primary repair in esophageal atresia with fistula: role of extrapleural approach. J Indian Assoc Pediatr Surg 2020 May-Jun;25(3):155–62. https://doi.org/10.4103/jiaps.JIAPS_73_19. Epub 2020 Apr 11. PMID: 32581443; PMCID: PMC7302468.
[20] Ten Kate C.A, Tambucci R, Vlot J, Spaander M.C.W, Gottrand F, Wijnen R.M.H, Dall'Oglio L. An international survey on anastomotic stricture management after esophageal atresia repair: considerations and advisory statements. Surg Endosc 2021 Jul;35(7):3653–61. https://doi.org/10.1007/s00464-020-07844-6. Epub 2020 Aug 3. PMID: 32748272; PMCID: PMC8195894.

[21] Baird R, Laberge J.M, Lévesque D. Anastomotic stricture after esophageal atresia repair: a critical review of recent literature. Eur J Pediatr Surg 2013 Jun;23(3): 204–13. https://doi.org/10.1055/s-0033-1347917. Epub 2013 May 29. PMID: 23720206.
[22] Lu Y.H, Yen T.A, Chen C.Y, Tsao P.N, Lin W.H, Hsu W.M, Chou H.C. Risk factors for digestive morbidities after esophageal atresia repair. Eur J Pediatr 2021 Jan; 180(1):187–94. https://doi.org/10.1007/s00431-020-03733-1. Epub 2020 Jul 9. PMID: 32648144.