Tubular Duplication of the Esophagus in a Newborn, Treated by Thoracoscopy

Igor Khvorostov1  Alexey Gusev2,3  Abdumanap Alkhasov2  Sergey Yatsyk2  Elena D’yakonova2,3

1 Department of Pediatric Surgery, Volgograd State Medical University, Volgograd, Russian Federation
2 Department of Pediatric Surgery, Federal State Autonomous Institution “National Medical Research Center for children health” MH RF, Moscow, Russian Federation
3 Department of Pediatric Surgery, RUDN University, Moscow, Russian Federation


Abstract

We present a case of tubular esophageal duplication in a 3-day-old female newborn (38 weeks, 2,500 g) without concomitant abnormal development. Esophageal duplication was diagnosed based on the clinical picture, direct laryngoscopy, esophagography and computed tomography. The duplicated esophagus was resected by thoracoscopy leaving the orthotopic esophagus in place. Isolation from the pharynx was performed via a separate cervical incision. After a follow-up period of 20 months, the child returned to normal growth and development.

Introduction

Congenital tubular esophageal duplication is a rare congenital anomaly in which the second esophagus with mucus, submucous, and muscular membranes, corresponding to the esophagus, is adjacent to the true esophagus without a common wall. This condition causes dysphagia, nausea, vomiting, retrosternal pain or respiratory problems (stridor and recurrent pneumonia). Presentation typically occurs during the newborn period. Tubular esophageal duplication represents approximately 10% of all foregut duplications.1,2 The incidence of this malformation is estimated to be 1:8,200, with male sex predominance 2:1.3 It is subgrouped into three types as follows: (1) cystic (the most common), (2) tubular, and (3) diverticular. Very few cases have been reported and described in newborns.4–8

Case Presentation

We report the case of a 3-day-old female (38 weeks and 2,500 g) presented to our surgical clinic from the maternity hospital with a history of respiratory distress, salivation, and dysphagia. The newborn was delivered via cesarean section to a primigravida mother. There was no visible additional malformation. Esophageal duplication was diagnosed based on the clinical picture, direct laryngoscopy, esophagography and computed tomography. The duplicated esophagus was resected by thoracoscopy leaving the orthotopic esophagus in place. Isolation from the pharynx was performed via a separate cervical incision. After a follow-up period of 20 months, the child returned to normal growth and development.

Keywords

► esophageal duplication
► newborn
► thoracoscopy
esophagus (►Fig. 2). A Nelaton’s catheter was inserted into the double esophagus before the operation for better visualization of the tubular duplication esophagus during thoracoscopy. The infant underwent a right thoracoscopy. The procedure was performed with three ports. The initial 5-mm trocar was inserted just below the right scapula tip and a 5-mm camera was inserted as well. A 3-mm port was placed in the fifth intercostal space posterior to the tip of the scapula. Another 3-mm trocar was inserted at the right midaxillary line along the scapula margin (►Fig. 3). The esophageal duplication tubular sac was removed from the surrounding tissue all the way from the main esophagus using a coagulation hook and monopolar coagulation. There was no common wall between the tubular and the main esophagus. The dissection of the doubled esophagus was continued cranially to the level of the superior chest aperture. Then, a left cervical neck incision was made and the tubular esophagus was removed. The child was left intubated for 5 days. Feeds via the nasogastric tube were commenced up until the sixth postoperative day. Recovery of the child was uneventful and the patient was discharged on the postoperative day 16. Histopathological examination showed a gastric-type mucosa with a well-developed submucosa, muscularis propria, and serosa. A contrast esophagogram via gastrografin swallowed 2 weeks after surgery showed a normal esophagus (►Fig. 4). After a follow-up period of 20 months, the child returned to normal growth and development.

Discussion

Pediatric tubular duplication of the esophagus is a rare congenital anomaly. The incidence of congenital esophageal duplication is estimated to be 1:8,200, with male sex predominance (male:female = 2:1).\textsuperscript{3} Cystic duplications are far more common, with tubular duplications accounting for less than 10% of cases.\textsuperscript{10} Tubular duplications without communication to the normal esophagus are more common than cystic duplications. Classically, these patients suffer from gastrointestinal and respiratory symptoms like nausea, vomiting, dysphagia, respiratory distress, or aspiration pneumonia. Diagnosis can be made by esophageal contrast study, with several duplications discovered incidentally on X-ray of the chest. A CT can be useful in making a diagnosis by showing a second tubular structure adjacent to the esophagus. In this case, the diagnosis was made via upper endoscopy. It allowed delineating the detailed anatomy, the extent of duplication, presence and length of a common wall, and a possible communication to the normal esophagus or trachea. It is well documented that a surgical excision is a viable option for the duplication of the esophagus.\textsuperscript{4,11} In our patient, the thoracoscopic approach was successful. This report shows that although extremely rare, a tubular esophageal duplication should be considered as a differential diagnosis in newborns with feeding intolerance. After surgical excision of the additional esophagus, prognosis is favorable in most cases.\textsuperscript{9,12}
Conflict of Interest
None declared.

References
10 Trappey AF III, Hirose S. Esophageal duplication and congenital esophageal stenosis. Semin Pediatr Surg 2017;26(02):78–86

Fig. 3 Patient positioning and placement of trocars: 1, camera; 2 and 3, working ports.

Fig. 4 Follow-up contrast esophagram 1 year after surgery showing a normal position of the orthotopic esophagus.