Comment

Sequestration is thought to occur when an accessory lung bud migrates abnormally during differentiation from the caudal foregut together with its blood supply, which becomes the aberrant supplying artery [1]. Extralobar sequestration is associated with one or more congenital anomalies in about 50% of patients, commonly including diaphragmatic hernia, communication with foregut structures, or cardiopulmonary malformations. Only about 14% of intralobar sequestrations are associated with additional anomalies [3]. Despite the presence of both extralobar and intralobar sequestrations in this patient, no such anomalies were found.

Sequestration is identified most frequently among the infant and pediatric populations, either during workup for respiratory distress or recurrent infection, or as an incidental finding on imaging studies performed for unrelated reasons. As with this patient, adults most frequently present with recurrent pneumonia or chest pain, or both. Symptomatic sequestrations are predominately intralobar [4].

The case presented represents an extremely rare variation of pulmonary sequestration. Subphrenic extralobar sequestration is an unusual finding, comprising 1.5% to 2.5% of reported cases. The initial presentation of these lesions is most commonly an incidentally imaged abdominal mass, and when the radiographic evaluation cannot exclude malignancy, the mass is resected [5, 6]. Duplicated intralobar and extralobar sequestration is an even rarer variant, with 9 reported patients with coexisting ipsilateral intralobar and extralobar sequestrations and 3 with an intralobar and contralateral extralobar sequestration [3, 7, 8]. Our patient presented with sequestrations on both sides of the diaphragm. Derivation of the arterial supply from the celiac trunk is reported in 1% of sequestrations [3].

Principles of management for pulmonary sequestration in the adult patient were highlighted in this case and include careful preoperative evaluation of functional pulmonary reserve and precise characterization of aberrant arterial supply to prevent uncontrolled intraoperative hemorrhage (conventional angiography is not necessary if computed tomography or magnetic resonance angiogram adequately define the anatomy) [2, 4].

Symptomatic sequestrations should be resected, and many experts believe that all intralobar sequestrations should be surgically treated due to the high rate of infection associated with these lesions. Subphrenic sequestrations have been reported primarily in the pediatric population, with resection undertaken to definitively exclude pediatric renal neoplasm in nearly all cases. Given that the typical clinical course of an asymptomatic extralobar sequestration is benign, conservative management is a reasonable option for management of an asymptomatic lesion in the adult patient if a definitive diagnosis of extralobar sequestration can be reliably established radiographically [1, 3].

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Three-Stage Reconstruction of the Airway and Alimentary Tract in a Case of Tracheal Agenesis

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In the few surviving cases of tracheal agenesis, infants have not been capable of oral intake because the esophagus was used as a substitute for the trachea. We performed a three-stage reconstruction of the airway and alimentary tract in an infant with tracheal agenesis. This procedure involved a double cervical esophagostomy followed by an anastomosis of the upper mid-esophagus and carinal trachea. Finally, the esophagus was reconstructed by an anastomosis of the cervical esophagus to the lower esophagus. This novel procedure may become a highly effective surgical treatment for some infants in critical condition due to tracheal agenesis.

(Ann Thorac Surg 2010;89:2019–22)
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Tracheal agenesis is a rare, congenital abnormality in which the tracheal segment between the cricoid cartilage and the carina is absent or severely stunted. This fetal malformation was first described by Payne [1] and classified by Floyd and colleagues [2]. This classification is based on the degree of distal tracheobronchial development and its fistulous connection to the esopha-

Accepted for publication Nov 4, 2009.

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Published by Elsevier Inc

0003-4975/$36.00
doi:10.1016/j.athoracsur.2009.11.021

Ann Thorac Surg
2010;89:2019–22

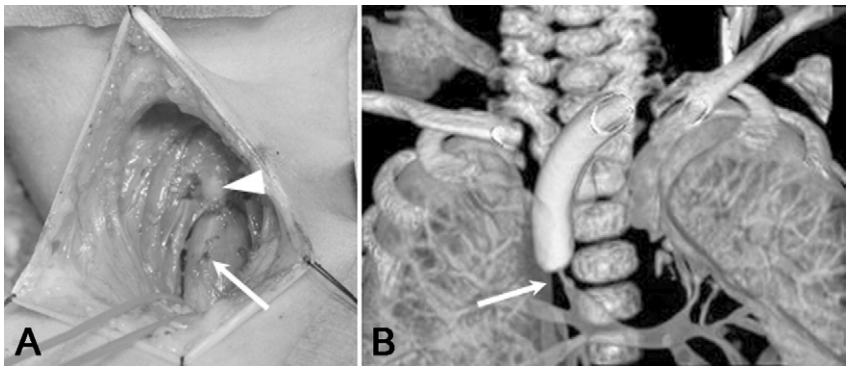

Fig 1. (A) The airway was disrupted at the end of the cricoid cartilage (arrowhead) and there was no trachea in front of the esophagus (arrow). (B) Three-dimensional reconstructed computed tomographic scan at 5 months of age demonstrated a narrow tracheoesophageal fistula (arrow) between the cannula and carinal trachea.

gus. Several surgical approaches have been attempted, although a definitive treatment strategy has not been established. Fonkalsrud and colleagues [3] initially reported a case in which the patient survived for 6 weeks after reconstructive surgery that involved trachealizing the esophagus and dividing the distal esophagus. However, in most cases, the patients are unable to be weaned off mechanical ventilation. Recently, a few cases of spontaneous respiration and long-term survival have been reported [4, 5]. Even in these cases, however, the infants were incapable of oral intake because the esophagus was used as a substitute for the trachea, with the exception of a case in which the esophagus was reconstructed by colonic interposition [6]. We planned a three-stage reconstruction of the airway and the alimentary tract during the neonatal period.

A female infant with a history of hydramnios was born at 35 weeks gestation (weight, 2,240 g). She had respiratory distress develop immediately after birth, but this condition improved after esophageal intubation. Endotracheal intubation was unsuccessful, which suggested the absence of the trachea and the existence of a tracheoesophageal fistula. She was suspected to have tracheal agenesis and was transferred to our hospital. The diagnosis of tracheal agenesis was confirmed by flexible fiberscopic examination. Distal esophageal banding with gastrostomy was performed 6 hours after birth. The diagnosis of Floyd's type I tracheal agenesis was made as the tracheal bifurcation with an opening to the right tracheal bronchus that was visualized at the anterior wall of the esophagus through a tracheoesophageal fistula. Ultrasonography revealed an association with tetralogy of Fallot. A plan for the three-stage reconstruction of the airway and the alimentary tract was developed, and the first step of the operation was performed the next day.

A horizontal skin incision was made above the sternal notch. The larynx was disrupted at the end of the cricoid cartilage and no trachea was detected in front of the esophagus (Fig 1A). A proximal cervical esophagostomy was created on the right anterior chest wall to divert salivary secretions. A distal cervical esophagostomy was created as the entrance of the newly established airway. A tracheostomy cannula was positioned in the esophagus connecting the infant to the ventilator (Fig 2). A three-dimensional reconstructed computed tomographic scan demonstrated the presence of a stenotic tracheoesophageal fistula requiring repeated balloon dilation (Fig 1B). The patient was momentarily free from mechanical ventilation. However, the tracheoesophageal fistula was not rigid and easily collapsed, which resulted in unstable respiration. Consequently, airway reconstruction was attempted at 8 months of age.

After a right thoracotomy through the fourth intercostal space, the mid-esophagus, tracheoesophageal fistula, and carinal trachea were isolated. The lower mid-esophagus was divided below the tracheoesophageal fistula, which was resected using cardiopulmonary bypass. Anastomosis of the upper mid-esophagus to the carinal trachea was performed, and the end of the lower mid-esophagus was closed and left in the thoracic cavity. A ringed, expanded polytetrafluoroethylene graft (diameter, 20 mm; length, 20 mm) was placed with radial traction sutures around the esophagus as an external supportive stent [6] to prevent esophageal collapse (Fig 3). The distal esophageal banding was released to avoid strangulation and the cervical esophagus was elongated 1

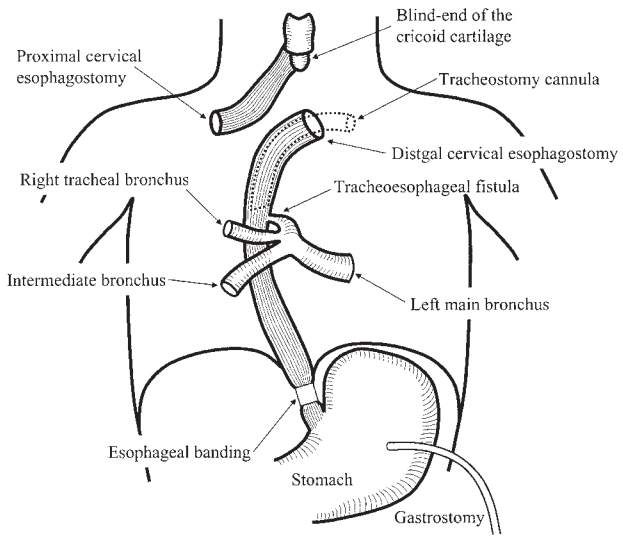

Fig 2. Diagram of the airway and alimentary tract after a double cervical esophagostomy and esophageal banding.

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