Minimally invasive resection of congenital tracheobiliary fistula with thoracoscopic cholangiography

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A 7-year-old girl was seen for severe asthma, chronic cough, and recurrent episodes of pneumonia. A carinal pouch was noted projecting inferiorly with tree-in-bud opacities on computed tomography (CT) scan (Figure 1). Bilious fluid was noted from within that tract on bronchoscopy (Figure 2), but contrast injection under fluoroscopic guidance did not demonstrate a fistulous connection to another structure (Figure 3). Esophagoscopy and esophagram were normal. After multidisciplinary discussion, the family elected for thoracoscopic resection of symptomatic presumed carinal diverticulum. Written consent to publish was obtained from the patient’s guardian.

With the patient in left lateral decubitus position, the fistula was identified at the carina (Video 1). This was confirmed with intubation of the fistula using a bronchoscope. The fistula was isolated at the level of the carina and divided using a 5-mm stapler (JustRight; Bolder Surgical), with care taken not to narrow either bronchus while ensuring a large pouch was not left behind. The fistula was then dissected along the chest wall with bilious fluid emanating from the tract. A cholangiogram catheter was placed within the fistula, and cholangiogram (8 mL of 50:50 diluted Omnipaque; GE Healthcare) confirmed the lack of anomalous common bile duct anatomy. The fistula was resected using a 30-mm Endo GIA tan load stapler.

CENTRAL MESSAGE
Congenital tracheobiliary fistula is a rare congenital anomaly. We present a patient treated with thoracoscopic resection with confirmation of biliary anatomy using thoracoscopic cholangiography.
Chest tubes were removed on postoperative day 3 (anterior) and postoperative day 4 (posterior), and the patient was discharged home later that day.

Final histopathology confirmed tracheobiliary fistula. At 2-week follow-up, she had complete resolution of symptoms and a normal radiograph of the chest. CT scan at 6 months showed surgical clips at the level of the fistula with no further pulmonary disease or bronchiectasis.

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DISCUSSION

Congenital tracheobiliary fistula is a rare congenital anomaly in which there is a patent connection between the trachea and the biliary tract. This is most commonly diagnosed in the neonatal period secondary to respiratory distress or recurrent pulmonary infection.1 Our patient, however, presented later in life, which made the diagnosis more challenging. While most fistulas, such as that seen in our patient, occur at the carina, it can also originate from the right main (30%) and left main bronchus (10%).2 Diagnosis is made using bronchoscopy and fistulography, with findings of yellow-stained tracheal mucosa and usually trifurcation at the trachea.

Another important consideration is that there is 36.8% incidence of concurrent common bile duct anomaly in these patients.2 In these select cases, the fistula may be the only outlet for bile outflow; therefore, the biliary system needs to be interrogated to ensure that there is normal bile duct drainage into the duodenum. Dynamic contrast imaging using fluoroscopy was employed in our patient to better delineate the anatomy before operative intervention, with no evidence of other anomalous biliary drainage. Had a coexisting atresia or hypoplasia of the common bile duct been noted with the fistula acting as the sole source of drainage for the left lobe of the liver, the patient would have required either hepatic resection with Roux-en-Y decompression, or mobilization of the fistula into the abdomen with a fistula enteric anastomosis. If unrecognized, and inadvertently ligated, this could result in hepatic atrophy of the affected segment, anomalous biliary drainage, cholelithiasis, and late malignant degeneration.3 Magnetic resonance cholangiopancreatography or hepatobiliary iminodiacetic acid can be of benefit to assess these.

FIGURE 2. Fistula tract located at the carina with bilious fluid within.

FIGURE 3. Dynamic bronchoscopy with fluoroscopic injection of contrast without evidence of anomalous biliary anatomy to the liver.

VIDEO 1. This video depicts the successful thoracoscopic resection of a tracheobiliary fistula in a pediatric patient. Before dividing the distal fistula at the level of the diaphragm, a thoracic cholangiogram was performed to confirm the biliary anatomy. Video available at: https://www.jtcvs.org/article/S2666-2507(22)00367-4/fulltext.
Long-term sequelae of a congenital tracheobiliary fistula include parenchymal injury from repetitive bile pneumonitis, as well as concern for malignant transformation. As a result, surgical resection of the fistula through right thoracotomy is the recommended treatment. A minimally invasive thoracoscopic approach has been associated with shorter hospital stay, less postoperative pain, and reduced rate of surgical-site infection for other indications in the pediatric population, and, therefore, is an attractive alternative to thoracotomy in this particular population. To date, there have only been 2 other reports of thoracoscopic resection for a congenital tracheobiliary fistula in a pediatric patient,\(^{4,5}\) one of which was 3.5 years old and the other a neonate. However, we report a new technique for intraoperative confirmation of biliary outflow, using intraoperative thoracoscopic cholangiography at time of thoracoscopic resection which is essential to avoiding significant hepatic morbidity.

This is a unique case of a tracheobiliary fistula in an older pediatric patient with symptomatic disease. The patient was successfully treated with thoracoscopic resection and ligation of the fistula with confirmation of biliary anatomy using thoracoscopic cholangiography.

References

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