LETTER TO THE EDITOR

Congenital bronchoesophageal fistula repaired by bronchoscopy-assisted fistulectomy in a neonate

To the Editor,

Congenital bronchoesophageal fistula (BEF) is a rare disease with equal frequency in males and females [1]. It is usually diagnosed in infancy when associated with esophageal atresia, or later in adult life if there is isolated BEF [2]. Braimbridge and Keith [3] classified congenital BEF into four types. Type II BEF is most common in adults, but rare in neonates [2]. Here, we report a neonate who presented with type II BEF, which was difficult to diagnose prior to surgery.

A male infant suffered from tachypnea after birth. Nasal continuous positive airway pressure was immediately administered. However, the sudden onset of pneumothorax and right upper lung pneumonia occurred on the Day 5 after admission. Intubation and chest tube insertion were performed immediately, and his condition improved. We removed the chest tube 2 days later and extubated the patient successfully on Day 15. However, sudden onset of desaturation recurred on hospitalization Day 21 after feeding was started orally. He was intubated once again. An extensive right upper lobe infiltrate persisted through the next 14 days of treatment in spite of enhanced chest percussion therapy (Fig. 1A). An esophagogram revealed no tracheoesophageal abnormality and no gastroesophageal reflux (Fig. 1B). Tracheobronchoscopy (TBS) showed an abnormal orifice found at the right main stem bronchus from which secretion leaked continuously, suspected as BEF (Fig. 1C). Additional multi-detector-row computer tomography (MDCT) was arranged to validate the presence of this lesion, but the result was normal. Due to the persistent symptoms, surgery was arranged.

During thoracotomy, an appropriately-sized flexible TBS (Olympus BF Type N20, Japan) with an outer diameter of 1.8 mm was used; passage through a 3.5-mm ID tracheal tube via a connector required the use of intermittent positive pressure ventilation (IPPV) to ensure adequate ventilation. Under illumination of the flexible tracheobronchoscope from the orifice at the right main bronchus, a 10-mm long fistula between the right main bronchus and the esophagus was clearly identified. The fistula was then divided and ligated. Postoperatively, the patient’s symptoms resolved completely.

The presentation of BEF is nonspecific, including chronic pneumonia, coughing, and hemoptysis [2]. This case was a neonate with isolated BEF presenting with several pulmonary complications. The first complication was the difficulty to wean from the ventilator. Although the pneumothorax was quickly resolved, the endotracheal tube was hard to wean until 10 days later. The second aspect was the recurrent pneumonia, especially when the patient started oral feeding, and this persisted despite treatment. The feeding history, recurrent pneumonia, and distended stomach hinted at a tracheoesophageal abnormality.

Barium esophagogram is the most sensitive tool to diagnose tracheoesophageal anomalies and MDCT is another noninvasive tool to investigate the disease [4]. However, the esophagogram may fail to demonstrate the disease in some situations. Firstly, the length and diameter of the fistula is relatively short and small in a neonate [5]. Secondly, the fistulous tract extended upwards from the esophagus and barium could not pass through the fistula because of the gravity. Thirdly, an esophageal fold or membrane flap may create a check valve to mask the fistula. In other words, MDCT cannot definitely detect the disease because of tiny lesions or the respiratory motion artifacts, especially in neonates [2].

Several reports have used intraoperative flexible TBS in repairing tracheoesophageal fistula [5], but there has been no such report for BEF. During the operation, we used a flexible tracheobronchoscope, which provided a transillumination to aid BEF localization and facilitate surgical identification. However, IPPV is mandatory during TBS through the tracheal tube to compensate for the increase in airway resistance. Oxygen desaturation, pneumothorax, and...
life-threatening gastric hyperinflation have been previously reported [5], although none of these occurred in our case.

In conclusion, pediatricians should be aware of BEF in newborns when unusual pulmonary complications occur. Even in the absence of evidence of BEF in barium esophagogram and MDCT, TBS is still helpful to uncover this rare disease in neonates. Intraoperative flexible TBS identification and probing of BEF is invaluable for successful surgical repair.

References


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