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PERINATAL/NEONATAL CASE PRESENTATION

H-type tracheoesophageal fistula in an extremely low birth weight premature neonate: appearance on magnetic resonance imaging

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Many diagnostic methods have been used to establish the diagnosis for a suspected H-type tracheoesophageal fistula (TEF). In case of a strong assumption of an H-type fistula, besides all standard diagnostic work-up tools a more aggressive combined approach is advisable. However, in a critically ill premature infant, conventional invasive investigations could not be performed as being potentially hazardous and not always easy to achieve. We describe the unique imaging features of an H-type TEF on magnetic resonance imaging (MRI). Our case demonstrates that MR images could be used for diagnosis, and localization of an H-type TEF could be detected safely and accurately in a sick preterm infant.

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Introduction

Tracheoesophageal fistula (TEF) without associated esophageal atresia is a rare congenital abnormality. The incidence of H-type TEF accounts for about 4% of tracheoesophageal abnormalities with an incidence of about 1 per 100 000 births.[1,2] The clinical diagnosis of an H-type TEF has been associated with the triad of paroxysms of coughing or cyanosis with feeding, gaseous distension of the gastrointestinal tract and recurrent pneumonia or bronchitis. Performance of an esophagogram and bronchoscopy are eventually diagnostic. Many other diagnostic techniques have been advocated in cases of H-type TEF such as scintigraphy, direct sagittal computed tomography (CT) scan and virtual bronchoscopy.[3–6] In spite of these, fistula identification can be elusive and difficult thus delaying the prompt diagnosis to early childhood or even to adulthood.[7,8] The rarity of the condition and concurrent problems as respiratory distress and gastroesophageal reflux may also postpone the detection of TEF in preterm newborns.[9] Also radiological investigations can be difficult to achieve and may yield increased risk of respiratory arrest especially in a low birth weight and sick infant.

Herein, we describe the imaging features of an H-type TEF on magnetic resonance (MRI) for the first time; and usefulness of MRI as a noninvasive technique in low birth weight and sick infant is discussed.

Case Report

A male infant who was born at 27 weeks' gestation with a birth weight of 880 g had severe respiratory distress requiring endotracheal intubation, surfactant therapy and mechanical ventilation. At a postnatal age of 3 days, the baby was extubated and supported by nasal continuous positive airway pressure. At the same time, the baby was started on breast milk feedings by orogastric tube, with gradually increasing volumes. Upon feeding episodes of apnea and aspiration pneumonia occurred which were attributed to gastroesophageal reflux. Repeated chest radiographs showed recurrent consolidations of the right upper lobe and gastric dilatation highly suggestive for aspiration pneumonia. Initially, a barium contrast rapid sequence cine-esophagogram and esophagoscopy were planned for a suspected H-type TEF. A nasogastric tube was inserted to the distal extend of the esophagus and isotonic water-soluble contrast dye was passed. We could not reveal a fistula tract and contrast dye was not observed in the trachea. Investigations could not be repeated because of infants being so small and severe desaturation problem during the procedure. At a postnatal age of 17 days, the case was discussed again with the pediatric radiologist and performance of MRI was suggested as being a noninvasive technique that lacks radiation and contrast requirement. MR imaging of the thorax was performed on a 1.5 T magnet using head and neck coil. The patient was transferred together with an intern doctor in transport incubator. He was not intubated. Sedation nor anesthesia was not performed. The patient was monitored by peripheral pulse unit of

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MRI. Whole MRI study was performed in 5 min including patient positing, monitoring, obtaining pilot images and the whole protocol. Sagital, coronal and axial T2 weighted images (TR: 2400, TE 90, slice thickness: 2/0.5 mm, matrix: 512 × 512, NSA: 2, time duration: 40 sec) were obtained. On axial images TEF was demonstrated as a linear hypointense structure between the trachea and esophagus at distance 1 cm above the carina (Figure 1). Although bronchoscopy failed to identify a fistula preoperatively, diagnosis was established in the operation and the fistula was successfully ligated through a standard right lower cervical incision (Figure 2). Sagital image was very useful for operation location (Figure 3). The infant made an uneventful postoperative recovery.

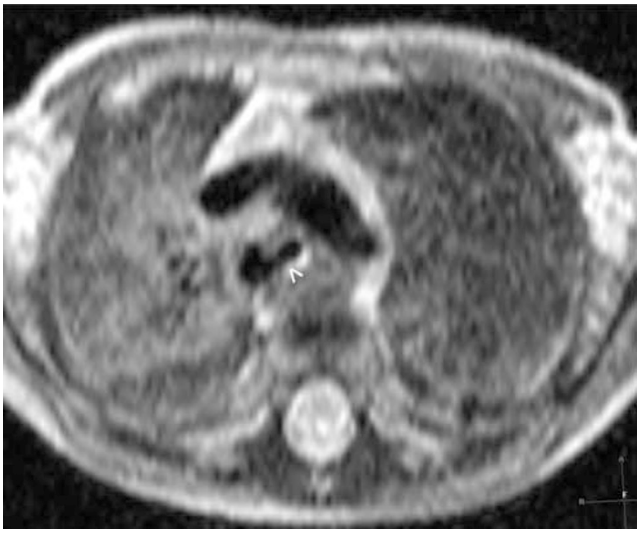


Figure 1 Axial T2 weighted magnetic resonance image demonstrates fistula between trachea and esophagus (arrowhead).

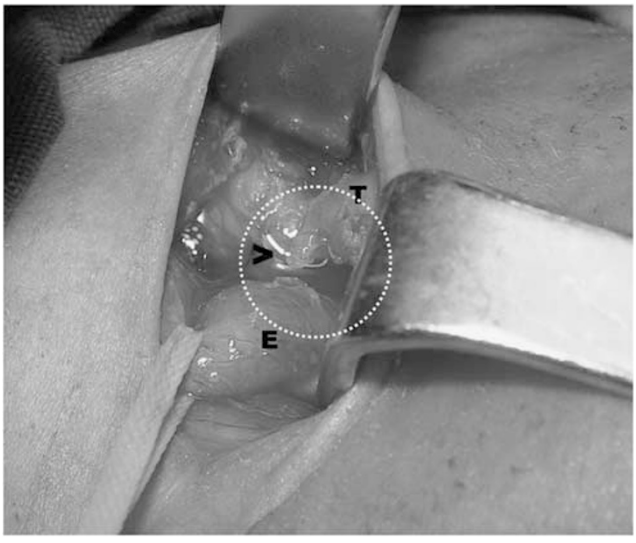


Figure 2 Dissection of the tracheoesophageal space shows the fistula. E, esophagus; T, trachea; F, fistula.

He was extubated and started feeding successfully. However, at a postnatal age of 47 days, he was lost due to septicemia.

Discussion

Many diagnostic methods have been used to detect a suspected H-type TEF. However, these various modalities do not remain entirely reliable and are inconsistent in the prompt diagnosis. We think that, if there is a presumptive diagnosis of an H-type TEF, besides all the standard diagnostic work-up tools a more aggressive, combined approach is advisable. This would increase the diagnostic potential and avoid unnecessary delays in the management.[3,7,8]

However, in the critically ill premature infant, invasive investigations could not be performed as being potentially hazardous, and not always easy to achieve. Ou *et al*.[10] reported that the high-resolution CT scan with air distention of esophagus to be a valuable diagnostic utility as an alternative noninvasive modality in this clinical situation. We think that, risk of aspiration of gastric contents still remains a major problem in this technique as the esophagus is distended with air. Instead, in our case we demonstrated that MRI was also a reliable noninvasive diagnostic modality in a sick premature infant with H-type TEF. MRI investigation seems to be safe lacking risk of radiation and aspiration of gastric contents. Besides contrast administration is not required to visualize the fistula and the time needed for the imaging is very short compared conventional esophagography.

The obliquity of the H-type TEF and close apposition of the trachea and esophagus mean that the fistula is occluded for much of the time. Pressure changes and the upward movement of the esophagus during swallowing may open the fistula, allowing air from the trachea to enter the esophagus or esophageal contents to enter the trachea.[3] MR images can demonstrate air tract between trachea and esophagus caused by H-type TEF thus rendering a beneficial aspect with no need to swallowing during the study, and without risk of aspiration as in the contrast work up.

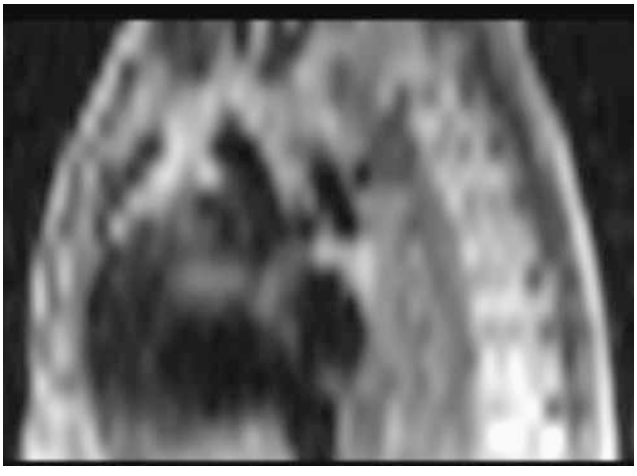


Figure 3 Sagital T2 weighted magnetic resonance image demonstrates fistula.

We believe that MR images could be used for diagnosis and localization of H-type TEF safely and accurately in a sick preterm infant. The demonstration of the fistula on MRI together with the clinical presentation is sufficient for the diagnosis if the initial investigation is negative or could not be done. Although these small infants usually require entubation, it is not a problem for evaluation of the fistula because it is seen as a hypointense structure due to air content. MRI does not seem to totally replace the initial standard investigations in H-type TEF cases it provides the surgeon adequate information to facilitate preoperative diagnosis and planning of surgery accordingly.

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