Late Presentation of Congenital Intrathoracic H-Type Tracheo-oesophageal Fistula: A Diagnostic Dilemma

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Citation

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Abstract

Congenital intrathoracic H-type isolated tracheo-oesophageal fistula is a rare malformation that presents with a characteristic triad of symptoms: choking and cyanosis on feeding, recurrent lower respiratory tract infection and abdominal distension. Children are invariably symptomatic from birth, although the symptoms may be intermittent and may vary in severity. A high index of suspicion is required because the symptoms are not specific. Establishing the diagnosis can be difficult and neither radiology nor bronchoscopy is infallible. Surgical division of the fistula is curative. We report two cases of intrathoracic H-type fistula which are very rare.

INTRODUCTION

Late presentations of tracheo-oesophageal fstulae (TOF) in childhood are uncommon. Congenital tracheo-oesophageal H-type fistulae without any associated atresia are very rare and usually diagnosed in the neonatal period. Persistence into childhood is very rare but if present will usually be present as either chronic or recurrent lung disease of unknown or unsuspected aetiology. Early diagnosis and adequate operative management avoid recurrent hospitalization for cough.

CASE REPORT

A 5-year-old male was admitted to our department with history of recurrent episodes of cough precipitated by feed since birth. This was associated with vomiting and abdominal distention. There was no history of cyanosis. On examination, the child was lethargic and malnourished. On chest examination, there were fine crepitations bilaterally in the lower chest. The abdomen was soft but distended, the rest was within normal limits. Chest x-ray examination showed a right lower-zone pneumonitis. Prone oesophagogram showed fistulous communication between trachea and oesophagus at the level of the fourth thoracic vertebra [figure-1]. Right thoracotomy was done through the 4 th intercostal space and the fistula was divided [figure-2]. The postoperative condition was uneventful and the patient was discharged on the 10 th postoperative day.

Figure 1

Figure 1: shows H-type fistulous communication on prone oesophagogram



Figure 2

Figure 2: shows H-type fistula between trachea and oesophagus



CASE 2

A girl had been investigated in two hospitals for recurrent attacks of chest infection and episodes of abdominal distension dating back to birth. Barium swallow examinations at 2 years were reported as normal. Finally, a prone oesophagogram was done that showed tracheooesophageal fistula at the level of the 4 th thoracic vertebra. She was managed by fistula ligation through right thoracotomy.

DISCUSSION

In 1873, H- or N-type isolated TOF was described by Lamb. Despite the elapse of a century since the original description, this condition continues to be often unrecognized [1]. Tracheo-oesophageal fistula without atresia has always been assumed to be a rare condition. The reported incidence varies from 1-8% [2] to 42% [3] among all types of tracheo-oesophageal fistulae; 70% of H-Type TOFs are at or above the 2nd thoracic vertebra and they can be as high as C7 and as low as T4 [4]. They are diagnosed clinically by the triad of

coughing and choking precipitated by feeds with or without cyanosis, gaseous abdominal distention and recurrent lower respiratory tract infection. The H-type fistula is associated with excessive tracheal secretions with bubbly respiration and improvement of symptoms with gastric tube feeding. The catheter test – gas bubbling with the tube in the esophagus – is unreliable.

In a large survey of reported cases by Killen and Greenlee in 1965 [$_5$] noted that the diagnosis was made within the first month of life in 43% and within the first year in 83%. In both our patients diagnosis was made above four years of age, the fistulae were intrathoracic and of a very rare type. The diagnosis can be made by prone oesophagogram (cine video-oesophagogram) which is reliable. Bronchoscopy and oesophagoscopy can be done in the preoperative period for fistula cannulation [$_6$] and also to confirm diagnosis. The fistula was successfully divided through a thoracic approach .The postoperative condition was uneventful without any complication in both cases.

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