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Case Report

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An uncommon presentation of double H type of tracheoesophageal fistula

Abstract

An eight-year-old child came with complaints of recurrent pneumonia since early childhood. At birth he was diagnosed to have Tracheo-esophageal fistula (TEF), for which he was operated. He was treated as asthma without much relief. Computed tomography of thorax showed Double H type of fistula with pneumonia and bronchiectasis.

Keywords: double H type TEF, asthma, recurrent pneumonia, bronchiectasis

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Abbreviations: TEF, tracheo-esophageal fistula; VACTERL, vertebralanalcardiovascular renal and limb

Introduction

Tracheoesophageal fistula (TEF) is an abnormal connection between the trachea and the oesophagus.[1] Most TEF abnormalities are diagnosed at birth since 98% of them are associated with esophageal atresia.[2] The incidence is approximately 1 in 4000 live births and in some cases, it may associate with VACTERL (Vertebral, Anal, Cardiovascular, Renal and limb) anomalies.[3] H type of tracheoesophageal fistula is rare and comprises only 4% of the cases of TEF.[4] In the H type, the fistula runs from the posterior wall of trachea downwards to the anterior wall of esophagus.[4] Double H type of fistula is even less common with only few cases reported.[5] There is no associated esophageal atresia. These cases are difficult to diagnose early because of nonspecific symptoms.[6] If untreated it leads to multiple complications such as recurrent pneumonia, lung abscess, ARDS, acute lung injury, poor nutrition, bronchiectasis from recurrent aspiration, respiratory failure and death.[1] Here we present a case of an eight-year-old who presented with recurrent lower respiratory tract infection mimicking asthma and was found to have double H type of TEF.

Case report

An 8-year-old boy came with history of recurrent cough, breathing difficulty since he was 8 months of age. Present episode was for 5 days with fever, purulent sputum and breathlessness with wheeze. The child came for evaluation of refractory asthma to pulmonary physician. His mother had polyhydramnios in pregnancy. She delivered him by full term normal vaginal route. Following birth, he had tachypnea and breathing difficulty and nasogastric tube could not be passed. He was diagnosed with Tracheo-oesophageal fistula and was operated. Surgical closure of upper fistula was done by pediatric surgeon. Post-operative period was uneventful.

He was then asymptomatic for 8 months. At 8th month of age he was first diagnosed with pneumonia, admitted and treated with antibiotics. After 1 year of previous episode he again had respiratory infection for which he was treated. Thereafter, since 5 years of age he had been having recurrent episodes of cough with purulent sputum, breathlessness with wheeze every 2 months for which he was diagnosed to have Asthma and treated with inhaled bronchodilators and inhaled steroids with not much relief. On examination, he was febrile with pulse oximetry saturation 90%. He had tachycardia and tachypnea. He was poorly built for his age. Respiratory examination showed bilateral diffuse polyphonic rhonchi and coarse crepitations. Other system examination was within normal limits.

Since the above presentation was not typical of Atopy and Asthma, Computed Tomography of thorax with contrast was asked. It showed presence of Tracheo-oesophageal fistula (TEF) with bilateral pneumonia and bronchiectasis. Double H type of fistula was seen with upper fistula forming a pouch and the lower fistulous connection persisting between trachea and esophagus. Diagnosis of recurrent respiratory infection secondary to TEF was made. This case is unique because recurrence of TEF post-surgical correction is uncommon. It also highlights the importance of a detailed birth history and its clinical correlation.

Discussion

TEF occurs due to abnormal development of Tracheo-esophageal ridges in fetus. There is failure of lateral septation of the two tracts, namely digestive and respiratory, resulting in formation of abnormal communication between the two[3]. It usually occurs between fourth to fifth week of development.[7] It was first described in 1929 by Negus. It was then an incidental postmortem finding in an adult who died of lung carcinoma.[3] Isolated H type of TEF is a rare disorder and is hence challenging to the diagnose.[1] H type of fistula is not associated with esophageal atresia.[3] If the defect is large, then these cases present early with respiratory symptoms due to aspiration and abdominal

distension.[3] Depending on the pressure difference between two structures, the air can enter into the oesophagus or food particles can enter into trachea, thus causing symptoms.[1] The common complaints are recurrent respiratory infections, aspiration and bloating of abdomen.[8] Our patient had recurrent respiratory tract infection with fever, purulent sputum and bronchiectasis. This complication leading to chronic lung disease in the form of bronchiectasis in later life is because of repeated lung infection secondary to aspiration.[9]

Survey by Killen and Grenlee showed that 43% cases were diagnosed in 1st month while 83 % cases were diagnosed in 1st year.[10] Early diagnosis is considered up to 4 years of age.[3] Our patient was diagnosed and operated at birth. He however had a recurrence and presented at a later age of 8 years which is rare.TEF antenatally presents with polyhydramnios due to inability of fetus to swallow amniotic fluid. The incidence of polyhydramnios is variable. Some studies have found an incidence of 61%, while few others have reported up to 91%.[7] This is rarely seen in H type pf TEF.[8] The mother of a child had polyhydramnios in pregnancy. At the time of our diagnosis he was found to have H type of TEF.

The various modalities of diagnosis include esophagogram,[11] Tc-99m sulfur colloid scintigraphy,[12] Computed tomography[13] and Magnetic resonance imaging.[14] The gold standard for diagnosis however, is combined bronchoscopy and esophagoscopy with passing of a fine catheter through the fistula.[15] In our case, Computed tomography of the thorax was able to pick up the fistula and its associated complications.

The treatment is by surgical correction. This can be done by cervical route or thoracic route depending on the location of the fistula.[5,16] Surgery involves ligation of the fistula with correction of the walls of the trachea and esophagus.[1] Alternatively, closure of the fistula has been attempted by endoscopic approach using tissue adhesives, electrocautery and Nd:YAG laser.[17] Few cases have been reported were Double H fistula has been reported and subsequently corrected by surgery. Mattei reported correction of one such fistula in one operation. The thoracic fistula was corrected by surgical repair and the cervical fistula was corrected by balloon catheter.[18]

Conclusion

This case highlights the importance of looking for second fistula while correcting H type of TEF.Everything that wheezes is not asthma. In cases of refractory wheeze, detailed evaluation should be done for cases that can mimic asthma and appropriately treated.

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Conflicts of interest

Author declare that there is no conflict of interest.

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