RARE CAUSE OF RECURRENT PNEUMONIA

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Abstract: Recurrent pneumonia is a diagnostic challenge and it warrants many investigations to arrive at a diagnosis. Congenital tracheoesophageal fistula H type is a rare cause of recurrent pneumonia in infancy and childhood. We report two cases who presented with recurrent pneumonia and remained undiagnosed for a long time requiring repeated hospitalizations. Fiberoptic bronchoscopy and barium swallow study were needed to confirm the diagnosis. Surgical conditions should also be considered as a cause for recurrent pneumonia. Early diagnosis prevents long term morbidity.

Keyword: Tracheoesophageal fistula H type, fiberoptic bronchoscopy

Fig. 1. Case 1. Fiberoptic bronchoscopy showing the abnormal opening in the posterior wall of trachea

Fig. 2. Case 1. Barium swallow study showing the tracheoesophageal fistula H type
CASE REPORT

A 2-year-old girl, first born of nonconsanguineous marriage was brought with the history of frequent cough since birth. Episodes of cough often occurred immediately after feeds. The child had recurrent pneumonia requiring parenteral antibiotics almost once in 2 months. Child was asymptomatic at the time of referral. There was no history of polyhydramnios in the antenatal period. Baby was delivered by caesarean section and her birth weight was 2.5 kg. Postnatal period was uneventful. Developmental milestones were normal. There was no known contact with adult tuberculosis patient.

The child did not have any respiratory distress. There was no significant lymphadenopathy. Her weight was 10 kg which was within 0 to -2Z score (WHO growth standards) and weight/length ratio was within 0 to 1Z score (WHO growth standards). Systemic examination was within normal limits. Her haemoglobin was 7.1 g/dL and blood counts were within normal limits (Total count was 10300/cumm, differential count was neutrophils 55% and lymphocytes 45%, and platelet count was 380 × 10^3/cumm). Renal and liver function tests were normal. Mantoux was negative. Resting gastric juice was negative for acid fast bacilli. HIV was nonreactive. Chest radiograph showed multilobar pneumonia. Cardiac evaluation revealed an ostium secundum type of atrial septal defect (size:4.3mm, left to right shunt). Child did not respond well even after 3 weeks of antibiotic therapy and hence bronchoscopy was planned.

Fiberoptic bronchoscopy revealed a fistulous opening in the posterior wall of middle third of trachea. Tracheomalacia was also noticed. Air bubbles were seen coming out of the fistulous opening, which suggested a tracheoesophageal fistula H type. Barium swallow study showed the presence of tracheoesophageal fistula H type. The infant was operated via right cervical approach. The fistulous tract was identified, transfixed and ligated. Postoperative period was uneventful. There was no further attacks of pneumonia on follow up.

DISCUSSION:

Tracheoesophageal fistula H type (Vogt IV classification, Gross E classification) is very rare accounting for only 4 % of all cases of tracheoesophageal fistula(1,2). Both the cases presented with recurrent pneumonia since birth and there were frequent attacks of cough following feeds. The older child did not have respiratory symptom at the time of admission. The other infant had severe acute malnutrition and presented with respiratory distress and chest radiograph showed multilobar pneumonia. Symptoms did not improve even after 3 weeks of parenteral antibiotics. Hence an airway anomaly was thought of and fiberoptic bronchoscopy was done. In both the cases an opening was seen in the posterior wall of the trachea and the possibility of tracheoesophageal fistula was considered. Barium swallow study demonstrated the fistulous tract in both these cases and confirmed the diagnosis, but 50% of the cases of H type of fistula may be missed on routine oesophageal contrast studies(2,3).

Congenital tracheoesophageal fistula H type occasionally presents in the first two weeks of life with cough or cyanosis on feeding. More commonly they present much later with recurrent chest infection and with a history of coughing on feeds(4-7).

Tracheomalacia was present in both of our cases. Tracheomalacia has been noted in 75% of pathological specimens from patients suffering from tracheoesophageal fistula(2). The associated anomalies seen in other forms of esophageal atresia are least common in cases of tracheoesophageal fistula H type(2). Of the two cases, the infant only had a atrial septal defect. The H type fistula is usually high in trachea near cervical area and can be operated through cervical approach(8). Among the two cases that we have reported here only the infant had the fistula in the cervical region and was operated through cervical approach.

Diagnosis of tracheoesophageal H type fistula has been difficult and hence delayed according to various studies (9-11). Radiological and endoscopic procedures are complementary in both diagnosis and treatment of tracheoesophageal fistula H type(12). The delay in diagnosis is unfortunate and it is because babies with a H type fistula have a lower incidence of other anomalies than the other types of tracheoesophageal fistula.
H type fistula, although one of the least common type of fistula diagnosed after the newborn period, must be considered when there is recurrent pneumonia without a definite cause on routine investigations or when there is a history of bouts of cough or choking following ingestion of fluids(13,14). A high index of suspicion is necessary in considering the diagnosis.

REFERENCES: