

Tubercular bronchoesophageal fistula

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Abstract

We present a commentary on bronchoesophageal fistulas (BOF) in children as per our experience in a sixteen-year-old girl with tubercular BOF. Acquired BOF in literature have been mostly described post traumatic or in association with malignancies in children. A child responded well to antitubercular therapy and supportive therapy unlike literature supporting surgical intervention.

Keywords: Bronchoesophageal fistula, Tracheoesophageal fistula, Mediastinal lymphadenopathy, Tuberculosis

Commentary

Tracheal or Bronchoesophageal fistula (BOF) is a rare complication of *Mycobacterium tuberculosis* (MTB) in children. Recently we reported a 16-year adolescent girl who presented with fever, cough, loss of appetite, weight loss for 3 months and breathlessness for 10 days [1]. Chest radiograph (hilar lymphadenopathy with bilateral pleural effusion) and positive GA gene Xpert (rifampicin sensitive) confirmed the diagnosis. Despite antitubercular therapy, dry cough persisted especially associated with food intake, more on liquids. Bronchoscopy and CECT chest confirmed bronchoesophageal fistula (right main bronchus, subcarinal). Child was managed with tube feeding and after 2 months of intensive phase showed clinical improvement with spontaneous closure of fistula on bronchoscopy. Child was allowed orally after 2 months and completed 9 months of treatment and is thriving well.

Tracheoesophageal Fistula: The Clinical suspicion

In children most acquired BOF are non-malignant and caused by either infectious processes (granulomatous inflammation of mediastinal lymph nodes) or trauma. Despite high prevalence of tuberculosis globally, BOF of tuberculous origin have rarely been described in children. Aspirations and respiratory failure remain most common presentations. Persistent coughing following the ingestion of solids or fluids, symptoms of acute pneumonia or a history of recurrent respiratory tract infections should alert the physician to the possibility of a trachea- or BOF.

Pathogenesis described are erosion of tuberculous peribronchial lymph nodes into the esophagus and bronchus or as a complication following surgical decompression of TB lymph nodes causing airway compression. It may be suspected in children with tubercular pneumonia with enlarged mediastinal lymph nodes and a significant amount of gastric air suspicion. Left sided fistular predominate (85%) [2-9].

Advances in Management

Medical management

Antitubercular drugs and nasogastric feeding had a good outcome. A recent study in South Africa concluded need of invasive ventilation due to extensive alveolar disease with severe hypoxia [10]. Ventilation in such children is challenging due to air leak from the airway to the oesophagus causing severe abdominal distention.

Surgical management

Stents: Self-expanding, nitinol-alloy- covered esophageal stent to obliterate the BOF opening,

which improved ventilation and resulted in the successful treatment have been reported in few reports [10]. Sengstaken-Blackmore tube has been used as a temporary intervention to reduce the air leak in few. This is only used short term due to the risk of necrosis of the esophagus.

All children were fed via a nasogastric or duodenal tube with percutaneous endoscopic gastrostomy in few during the acute phase of the disease. Anti-tubercular drugs initially administered via a nasogastric tube or intravenous drug regime (rifampicin, ofloxacin, and amikacin), due to severe abdominal distension. Standard oral four-drug treatment was reinitiated once the children were able to tolerate oral feeds.

Open closure: A recent report by Goussard et al. has shown various procedures in BOF closure in twenty patients as spontaneously (26%, n = 4), surgical closure via thoracotomy (46%, n = 7), fibrin glue closure (13%, n = 2) and esophageal stent closure 13% (n = 2) [10]. In the patients where the BOF closed spontaneously, the fistula appeared to be relatively small. In two cases where an esophageal stent was used for BOF closure, the closure was achieved after 42 and 67 days. However, in one patient there were esophageal stricture and granulation tissue formation, which required repeated dilatations over 6 months, after stent removal. In the second patient, the stent was removed after 3 weeks, due to compression of the trachea, after which extubation was successful and BOF closure occurred. In another five cases where fibrin glue was used, it was successful in three cases (60%). A single attempt was successful in one case, but in the remaining four cases, 3 to 4 attempts were required. The closure was confirmed 2 days after the intervention. In two cases, the glue failed and surgical ligation was required. The glue was installed into the fistula opening via the working channel of the bronchoscope after de-epithelialization of the fistula tract by pushing a cytology brush into the opening of the fistula. No complications related to the use of the fibrin glue were noted.

Open closure is challenging in both children and adults [11]. Goussard et al. reported that in children where surgical closure via thoracotomy was done, surgery was performed after 6 months of TB treatment except 1 case in which surgery was done after 2 months [10]. In all surgical cases the fistula could be identified and ligated. The fistula was left-sided in four of the six surgical cases. Postoperative complications included phrenic nerve neuropraxia (n = 1), pneumonia (n = 1) and empyema (n = 1).

Recent reports in adults with non-tubercular BOF have reported endoscopic methods such as clips, self-expanding metal stent (SEMS) and recently, the over scope clip (OTSC®) are increasing in popularity [11-14]. Another report from China in 26 patients reported in 26 non tubercular BOF where 23 patients were surgically treated (10 direct repair, and 13 either closure of esophageal defect or tracheal/bronchial defect). The concomitant procedures were permanent tracheostomy, tracheal resection and reconstruction, pulmonary resection, thoracoplasty esophagectomy, and esophagogastric anastomosis.

Outcome in Patients with BOF Closure

In study by Goussard et al., fifteen (75%) patients survived through definitive BOF management. The five patients who demised presented with severe respiratory failure and 40% (n = 8) required invasive ventilation. On univariate analysis, ventilation (P = 0.004),

CRP (P = 0.003), and albumin <18 g/dL (P = 0.001) were predictors of mortality. After 6 months, 67% (n = 10) of the 15 survivors were asymptomatic. But five had mild symptoms, with recurrent lower respiratory tract infections requiring hospitalizations (median duration 38 days: range 1-224 days). The chest X-ray of the 15 survivors at the end of the treatment normalized in 73% (n = 11), left-sided bronchiectasis 26% (n = 4) and right middle and lower lobes volume loss in 6%.

Another study reported paralysis of recurrent nerves, empyema, injury and ligation of subclavian artery, dehiscence of tracheal anastomosis, and contralateral pneumohydrothorax [11].

Conclusion

In children with combination of mediastinal lymphadenopathy and persistent cough following intake of food needs careful evaluation for trachea/bronchoesophageal fistula. Medical management in tracheoesophageal fistula is useful and few may need surgical closures. The outcome of broncho/tracheoesophageal fistulas is tuberculosis and is good if detected early and without complications.

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