Tracheo-bronchial remnants (chondroepithelial choristoma): An unusual cause of dysphagia

Sir,

Chondroepithelial choristoma also known as tracheo-bronchial remnants or cartilaginous esophageal rings is a rare cause of distal esophageal stenosis. Because of its rarity, the diagnosis is often overlooked in infants and young children presenting with feeding problems, especially when there are other major associated anomalies, and inappropriate treatment is often carried out before the correct diagnosis is established.

A 2-year-old female presented with vomiting immediately after food, the vomitus contained food particles and there was difficulty in swallowing solid food and inability to pass the Ryle's tube. A barium swallow showed a stricture at the lower end of the esophagus with proximal dilation. The esophagoscope could not be passed beyond the stricture, it showed inflamed esophagus with accumulation of food above the stricture. An esophageal resection of the stricture with anastomosis was done.

The etiology of this developmental anomaly is the defective separation of the embryonic respiratory tract from the primitive foregut resulting in sequestration of tracheobronchial precursor cells in the wall of the esophagus, which gets carried down during the growth process. The other postulate is localized metaplasia of mesenchymal cells in the wall of the esophagus producing the cartilage and epithelial tissues of the respiratory tract. Esophageal atresia occurs at the same embryonic stage in which the tracheal cartilage sequestrates in the esophageal wall. This explains the high incidence of the combination of esophageal atresia and stenosis due to tracheo-bronchial remnants. Anorectal anomalies are frequently associated with both these conditions.

The symptoms start in early infancy and include recurrent vomiting, regurgitation of fluids and progressive dysphagia usually beginning with weaning. These symptoms suggest partial esophageal obstruction. Aspiration into the lung also occurs and there are recurrent bouts of respiratory distress and respiratory infections. The X-ray or barium swallow shows a localized constriction of the distal esophagus with proximal dilation. Linear intramural clefts projecting horizontally from the area of stenosis correspond to the respiratory ducts. The stenotic segment fails to relax on swallowing.

The differential diagnosis includes stenosis due to caustic

Figure 1: Photomicrograph of esophageal biopsy showing stratified squamous lining epithelium, the submucosa shows cartilage & mucous glands of tracheobronchial origin (H/E, 40x).

Figure 2: Photomicrograph esophageal biopsy showing stratified squamous lining epithelium, the submucosa shows cartilage & mucous glands of tracheobronchial origin (H/E, 100x).

Figure 3: Photomicrograph esophageal biopsy showing stratified squamous lining epithelium, the submucosa shows cartilage & mucous glands of tracheobronchial origin (H/E, 400x).
agent ingestion, peptic esophagitis with or without hiatus hernia, achalasia cardia, esophageal webs and fibromuscular stenosis.[5] Our clinician’s differential diagnosis was reflux stricture, congenital esophageal stricture and achalasia cardia. A resection anastomosis was done.

Grossly, there is a fusiform constriction of the involved segment <2 cm in length. Cut section reveals incomplete cartilaginous rings. We received multiple bits of tissue 2 x 2 x 2 cm, with unremarkable mucosa, wall uniformly thickened with small whitish nodules.

Histologically, the esophageal wall shows large or small, cartilaginous tracheal rings, submucosal and mucosal glands of tracheal origin, ducts lined by respiratory epithelium, surrounded by lymphoid tissue, some communicating with the lumen, rarely is heterotrophic pancreatic tissue seen.[3,5,7] Sometimes all the structures may not be seen. In our case only cartilage and tracheal bronchial glands were seen [Figures 1-3].

The cause of obstruction may be mechanical, if the cartilaginous rings are large or interruption of progressive esophageal peristalsis, if they are small.[7] Arrangement of the respiratory elements could be in an orderly topographic manner, analogous to that occurring in the normal upper respiratory tract, as was in our case, where the name tracheo-bronchial remnant is more suitable.[6] If the lesion displays a loss of organized arrangement of its component elements with an intimate blending of glandular and cartilaginous structures, the term “chondroepithelial choristoma” is more appropriate.[8]

REFERENCES


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