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A rare variant of esophageal atresia: Esophageal atresia with muscular continuity and upper pouch fistula

Özofagus atrezisinin nadir bir türü: Musküler devamlılık gösteren ve üst poş fistülü olan özofagus atrezisi

To the Editor,

Pure esophageal atresia (PEA) is characterized by a long segment between the esophageal pouches without tracheoesophageal fistula (TEF). Here, we present a very rare case of esophageal atresia (EA) in which the two blind pouches were connected by an atretic segment and proximal TEF was observed.

A girl baby was born with a birth weight of 1970 g. Radiologic diagnosis of PEA was made by detecting a blind pouch of the proximal esophagus and the gasless abdomen. A gastrostomy was made. At 4 weeks of age, the patient underwent thoracotomy. It was observed that both ends of the esophagus were connected by an atretic segment (Figure 1). This segment was resected, and both ends were anastomosed. Mediastinal exploration did not show TEF. Histopathological examination of the specimen showed that the atretic segment consisted of smooth muscle tissue without lumen (Figure 1). The patient was discharged on the 14th day. She was hospitalized twice due to right upper lobe pneumonia within the next 2 months. Contrast esophagogram showed a missed proximal TEF. The diagnosis was confirmed by bronchoscopy. Therefore, she underwent rethoracotomy. The upper TEF was identified at the level of thoracic inlet and was repaired.

This type EA is very rare and only a few cases have been reported in the literature. Membranous obstruction with normal outer appearance of the esophagus and an atretic esophageal midportion in which a cyst located in this segment have been described (1,2). Sanal et al. (3) described a new subtype in which the proximal and the distal segments of the esophagus are connected by an atretic segment as observed in our case. The diagnosis of upper TEF is more difficult and requires a high index of suspicion based on clinical symptoms. It is associated with choke while attempting to feed, unexplained cyanotic spells or bouts of pneumonia. It may be missed on routine esophageal contrast studies. Even though a confident radiologic diagnosis, bronchoscopy is often required to confirm the diagnosis. Preoperative bronchoscopy could not be performed in our case because of technical difficulties. The classic location of an upper TEF within the upper pouch is near its distal end, but it was at the most proximal location in our case. All of these might be the reason for the missed TEF.

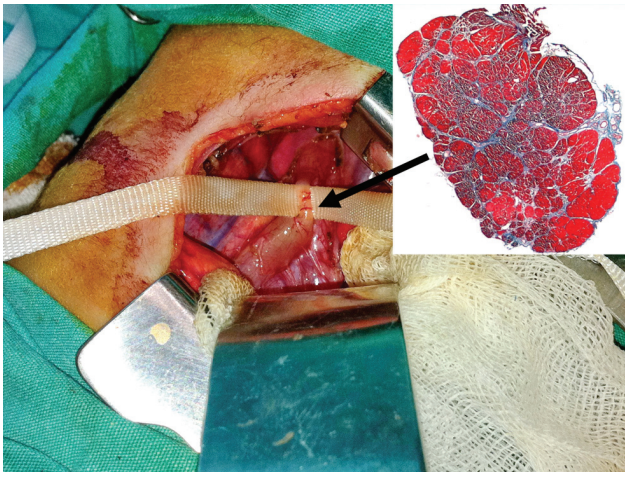

Figure 1. Operative photograph showing that the both ends of the esophagus were connected by an atretic segment and microscopic appearance of this segment showing bundles of smooth muscle cells without any lumen (Masson's trichrome staining, x20).

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EA with muscular continuity is a rare entity, and may be associated with upper pouch TEF. Therefore, great care must be taken to exclude a proximal TEF in these cases.

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Esophageal ulcers: A possible adverse effect of isotretinoin

İsotretinoin tedavisine bağlı özofageal ülser

To the Editor,

Drug-induced esophageal injury is a common cause of esophageal diseases. Many drugs have been reported to cause esophagitis and esophageal ulcers; among these are antibiotics such as doxycycline and tetracycline, nonsteroid anti-inflammatory drugs, aspirin, and potassium chloride (1).

Isotretioin is a synthetic analogue of vitamin A and is widely used in the management of acne vulgaris. However, several adverse effects of this drug have been reported, including mucositis and chelitis (2). Among these, a possible association of inflammatory bowel disease with isotretioin deserves further attention (3). Although the exact mechanism is not clear, the possible role of isotretinoin in the inhibition of epithelial cell growth, induction of apoptosis, lymphocyte migration, and immunomodulation have been proposed. We report a patient with multiple esophageal ulcers in which the only possible cause was the oral ingestion of isotretioin.

A 29-year-old woman who had no previous gastrointestinal complaints (including no reflux symptoms) and no serious medical or surgical history presented to our gastroenterology department with severe odynophagia. She had started to use isotretioin for acne vulgaris one month before and had not used any other medication recently. Her odynophagia began suddenly two days before presentation and was similar in intensity while swallowing solids and liquids. Her physical examination, routine laboratory tests, chest x-ray and upper abdominal ultrasonography were all within normal limits. An emergency endoscopy was performed and showed discrete esophageal ulcers (3-8 mm in size) starting 30 cm from the incisors and disappearing gradually towards the 38th cm (Figu-

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