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Case Report
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Esophageal Tubular Duplication in an Adult Patient. A Case Report and Review of the Literature

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Abstract

Introduction: Duplications of the alimentary tract are infrequent congenital malformations that might occur at any level from the mouth to the anus. The esophagus is the second most common site of the alimentary tract duplications. Based on autopsy reports, it is estimated that gastrointestinal tract duplication affects 1 in every 4,500 to 8,200 individuals. These malformations are predominantly diagnosed during childhood. Pathologically, two types have been recognized, cystic and tubular. Most duplications are cystic, and tubular type is extremely rare. Herein, we present the case of an adult patient with tubular esophageal duplication.

Case Report: A 28 year-old male patient was evaluated for chronic epigastric pain and pirosis. During an upper endoscopy we observed a tubular esophageal duplication with endoscopic findings suggestive of Barrett's esophagus in both segments. This finding was corroborated with CT of the chest. The patient was started on proton-pump inhibitor treatment and surgical resection was recommended.

Conclusion: Tubular esophageal duplication is extremely rare; and patients might have late clinical presentation.

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Introduction

Duplications of the alimentary tract are rare congenital malformations that occur at any level the mouth to the anus [1]. The esophagus is the second most common site of alimentary tract duplications [1,2]. Studies based on autopsy reports estimated that gastrointestinal tract duplications affects 1 in every 4,500 to 8,200 individuals [3].

Esophageal duplications are predominantly diagnosed during childhood [4]. Two types have been described, cystic and tubular. Most cases are cystic, and tubular type is extremely rare [1]. Only a few cases of tubular esophageal duplication have been reported in the literatura [1,5-9].

Esophageal cystic duplications are rare developmental anomalies of the foregut, that constitute 0.5% to 2.5% of all esophageal diseases [9]. Esophageal duplications tend to be diagnosed more frequently in adult life rather tan childhood, because these patients are commonly asymptomatic unless other complications occur. When symptomatic, patients present with recurrent dysphagia [10], intermittent chest pain, and signs and symptoms of acute mediastinitis due to rupture of the duplication [8], The definitive treatment for esophageal duplication in surgical resection [5]. Herein, we report the endoscopic and radiological findings in an adult patient with late presentation of esophageal tubular duplication.

Case Presentation

A 28-year old male patient presented to our Medical Centre with history of chronic epigastric

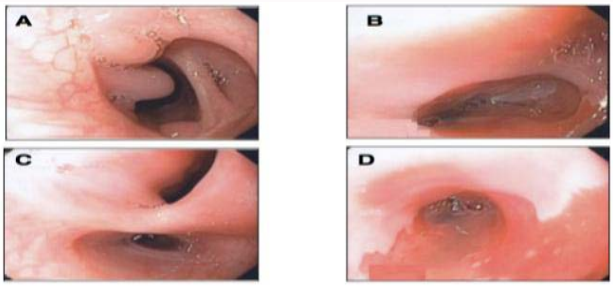

Figure 1: Endoscopy of the upper gastrointestinal tract. (A) Four polypoid lesions Zenker's diverticulum in the anterior segment of the esophagus. (B) 5 cm below the diverticulum is a training segmented elastic forming two esophageal tracts, both are connected to the stomach and allow the passage of the endoscope into the stomach; D) In the two distal esophagus are images of Barrett's esophagus; stomach and duodenum almost normal [15].

pain and pyrosis. Past medical history was only relevant for a remote appendectomy. An upper endoscopy revealed esophageal duplication, Zenker's diverticulum, two polypoid lesions on the mid esophagus, hiatal hernia, and endoscopic findings of Barrett's esophagus (Figure 1a-1d). A CT of the chest corroborated the esophageal duplication at posterior mediastinum; hiatal hernia; and mild fibrosis in the posterior segment of the right lung upper lobe (Figure 2a-2c).

Proton-pump inhibitors and prokinetic agents were prescribed. The response of medical treatment was appropriate and since five years ago a surgical resection was proposed to the patient order to decrease the risk of complications.

Discussion

Congenital duplications can occur anywhere in the gastrointestinal tract and one third of all duplications are foregut duplications involving the esophagus, stomach, or duodenum [11]. These duplications may be proximal or distal, and usually proximal or mediastinal duplications occur early in embryonic life [11]. Embryologically, esophageal duplications result from a defect in the tubulation (vacuolization) of the esophagus, which normally occurs in the sixth week of gestation [12], as this is the period when the foregut epithelium develops and elongates, the lumen forms and then undergoes dextro-rotations. Therefore, the majority of esophageal duplications occur distally and on the right side [13]. The etiology of gastrointestinal tract duplication is not fully understood; however, an initial developmental abnormality during the gastrulation stage is thought to be the most likely etiology [14]. The duplication of the esophagus could present in two types; a cystic form or a tubular form [15].

Esophageal duplication cysts, were first described in 1711 [9]. In 1937, Ladd suggested the use of the term "duplications of the alimentary tract" and applied the term to congenital lesions having three characteristics; the presence of a well-developed coat of smooth muscle [3,16], an epithelial lining representing some type of intestinal tract mucosa, and intimate anatomic association with some portion of the gastrointestinal tract [17]. Esophageal tubular duplication is extremely rare [5]. In an analysis of 281 duplications included in four case series, the esophagus was the second most common site of alimentary tract duplication, comprising 20% of all lesions, and all were noncomunicating and cystic type [1]. In another review, only two of 20 esophageal duplications were tubular type [6]. Only a few cases of esophageal tubular duplication have been reported [7,8],

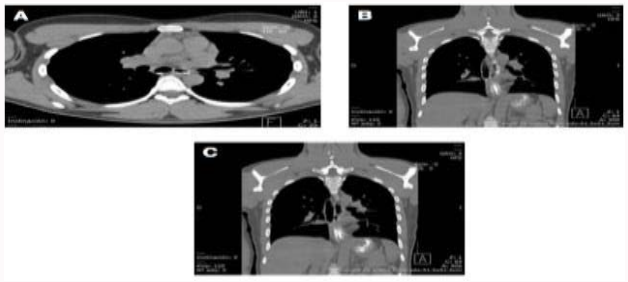

Figure 2: Computed tomography (CT) of the chest. (A) Axial plane of the CT scan at the subcarinal level showing to the tubular communicating esophageal duplication. (B&C) Coronal plane of the CT scan showing the esophageal duplications communicating with the stomach.

and most of them did not have both, endoscopic and radiographic findings [1,5-8].

Clinically, tubular esophageal duplications tend to be diagnosed more frequently in adulthood rather than childhood, as patients are asymptomatic unless complications occur. Because of the asymptomatic nature of this malformation, the incidence of these malformations might be underestimated [18]. Tubular esophageal duplications might become symptomatic when they had complications, such as hemorrhage, rupture, and displacement of adjacent organs, obstruction, respiratory distress, or infections [19,20].

Most of the case reports of esophageal tubular duplications communicate with their native lumen, often through multiple openings [7]. Duplications are commonly associated with other congenital malformations, such as spinal deformities, inguinal hernia, club foot, meningocoele, malrotation of the bowel, Meckel's diverticulum, umbilical and diaphragmatic hernia, bilobar right lung, hydrocephalus, congenital heart disease, prematurity and pigeon breast [1].

Treatment of esophageal duplication should be individualized according to the presenting symptoms and type of malformation. Various treatment procedures have been reported, ranging from surgical excision, enucleation, marsupialization, internal drainage [21], and more recently endoscopic management [22].

The definitive treatment for esophageal duplication is surgical excision [5]. Given the presence of Barrett's esophagus in both segments of esophageal duplication we have proposed to the patient to perform surgical resection of both esophageal segments. Currently, the patient continues on medical treatment based proton-pump inhibitor and prokinetic agents.

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