

## Case Report

# Esophageal Duplication Cyst in an Adult Masquerading as Submucosal Tumor

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### Abstract

Gastrointestinal duplications usually manifest in children and may involve the esophagus in 20% cases. Esophageal duplication cysts are a rare cause of dysphagia in adults. We report the case of a 35-year-old male who presented to us with progressive dysphagia of 6 months duration. Contrast enhanced computed tomography showed a soft-tissue lesion in right lateral wall of distal thoracic esophagus. On endoscopic ultrasound, a heterogeneously echotextured lesion with anechoic component present at intramural location in the lower esophagus was noted. The patient underwent surgical excision of the lesion and histopathology confirmed the diagnosis of esophageal duplication cyst.

**Keywords:** cyst; endoscopic ultrasound; dysphagia; computed tomography

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## INTRODUCTION

Gastrointestinal duplication cysts are rare entities, especially so in adults.<sup>1</sup> These represent spherical or tubular lesions, which occur in relation to the normal intestinal tract. The jejunum and ileum are the commonest location followed by mediastinum, colon, stomach, duodenum and rectum.<sup>1,2</sup> True duplication cysts result from the duplication of the submucosa and the muscularis without the duplication of the epithelium. Esophageal duplications cyst are usually diagnosed in childhood, but adults are more likely to be symptomatic.<sup>1,2</sup> Presenting manifestations may include dysphagia, chest pain, hematemesis or incidental detection. We report a young adult with esophageal duplication cyst who presented with dysphagia to both solids and liquids and posed a difficult diagnostic dilemma with endoscopic and endoscopic ultrasound (EUS) appearances closely mimicking a submucosal tumor.

## CASE REPORT

A 35-year-old male presented to us with complaints of gradually progressive dysphagia of 6 months duration. On the presentation he was having difficulty in swallowing of liquids also and had significant weight loss. He denied substance abuse. His clinical examination as well as hematological and biochemical investigations were normal. Upper gastrointestinal endoscopy revealed a well-defined submucosal lesion in the lower esophagus causing significant narrowing of the esophageal lumen (Fig. 1). Gastroscopy could be negotiated across this area of narrowing. Contrast enhanced computed tomography of the chest showed a lesion with soft-tissue attenuation in right lateral wall of distal thoracic esophagus with a likely possibility of submucosal or intramural lesion likely leiomyoma (Fig. 2). The magnetic resonance imaging of the chest also revealed similar features. EUS revealed a 2.6 cm × 1.6 cm heterogeneous echotextured lesion with anechoic component in the lower esophagus (Fig. 3). Muscularis propria was seen going around the lesion suggesting an intramural location. After obtaining an informed consent and giving antibiotic prophylaxis, EUS guided fine needle aspiration (FNA) of the lesion done with a 22-gauge needle and whitish fluid with thick consistency was aspirated (Fig. 4). Cytological

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Figure 1. Endoscopy showing a large submucosal bulge in the lower esophagus.

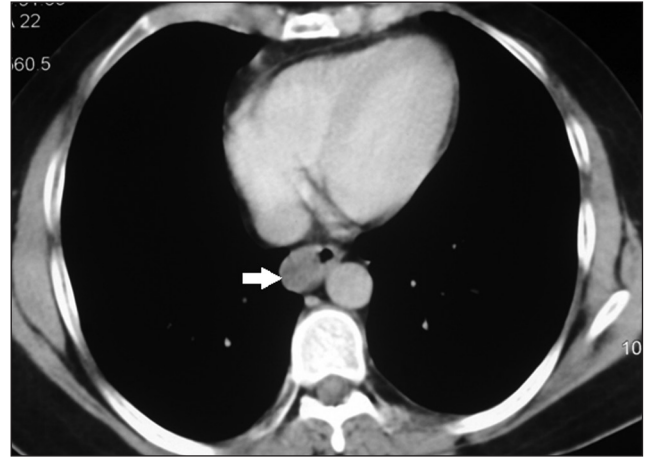


Figure 2. Contrast-enhanced computed tomography: Mass lesion (arrow) in the right lower esophagus.

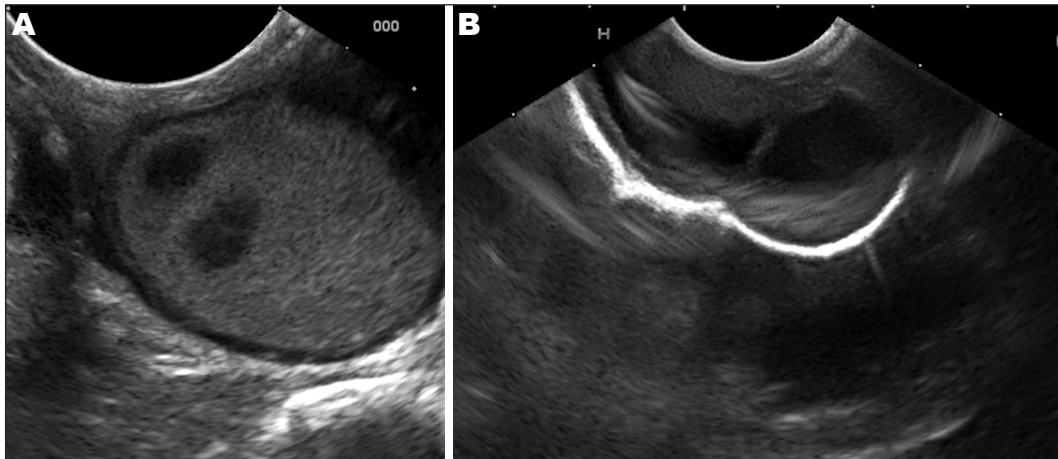


Figure 3. Endoscopic ultrasound: Heterogenous echotextured lesion with anechoic component in the lower esophagus.



Figure 4. Endoscopic ultrasound-guided fine needle aspiration.

examination of aspirate was inconclusive with it showing only necrotic cells and the culture of aspirate was also sterile. In view of severe symptoms of dysphagia and possibility of

an esophageal submucosal tumor, the patient was referred for surgery. A 2.5 cm × 1.5 cm cystic lesion was noticed in the lower esophagus that was excised with exteriorization of base. Histopathology of the resected specimen confirmed the diagnosis of esophageal duplication cyst. Patient is asymptomatic after 6 months of follow-up.

## DISCUSSION

Esophageal duplication cysts constitute around 20% of all the gastrointestinal duplication cysts.<sup>3</sup> Esophageal duplication cysts can be cystic or tubular, with cystic being the more common pattern. The location of the esophageal cysts may be variable: Lower esophagus being the commonest site while cervical and even intraabdominal esophageal duplications have also been reported.<sup>4,5</sup> Our patient had a cystic lesion in the lower esophagus. The esophageal duplication cysts may present with dysphagia, chest pain, respiratory difficulty, hematemesis, back pain or may be detected incidentally.<sup>6-9</sup> Most reports of symptomatic lesions are in children.<sup>6</sup> Some esophageal

duplication cysts may be associated with concurrent vertebral anomalies.<sup>10</sup> The differentials of esophageal duplication cyst includes other mediastinal cysts including hydatid cyst, muellerian cysts, bronchogenic cysts, pericardial cysts or cystic degeneration of mediastinal tumors.<sup>11</sup>

Traditionally the diagnosis is suspected on computed tomography, which usually reveals a homogenous of tissue lesion with regular margins.<sup>3</sup> EUS is also an important diagnostic modality for evaluation of mediastinal cysts and lymph nodes.<sup>12,13</sup> It provides a good tool to characterize these lesions and distinguish them from bronchogenic cysts by the absence of cartilage, proximity to the esophagus and double muscle layer around them.<sup>6,14</sup> EUS also provides an opportunity to do a FNA of these lesions for confirming the nature of contents. EUS will usually reveal an anechoic homogenous lesion with smooth margins in the submucosal wall. The muscularis propria of the lesion is continuous with the muscularis of the esophagus. It is important to consider the possibility of esophageal duplication even in patients with hypoechoic lesions or with heteroechoic lesions with an anechoic component as in our case because the presence of pus, blood or thick contents may modify the EUS features and result in confusion with other soft-tissue lesions like stromal tumors.<sup>15,16</sup> EUS guided FNA has been used to confirm the diagnosis in patients with mediastinal cysts but EUS FNA of duplication cysts may lead on to cyst infections despite using pre and post procedure prophylactic antibiotics.<sup>15</sup> Therefore, caution must be used while planning EUS FNA in patients with esophageal duplication cysts.

Esophageal duplications may get complicated and result in hemorrhage, perforation, infection and malignant transformation. They may undergo peptic ulcerations if lined by gastric tissue, which may lead to perforation or gastrointestinal bleeding.<sup>17</sup> Malignancy may also occur although the exact rate is uncertain.<sup>18,19</sup> For these reasons, the usual approach is to surgically excise these lesions although some workers suggest a wait and watch approach for asymptomatic lesions.<sup>20</sup> Some reports of endoscopic management of the duplication cyst are also available. Endoscopic fenestration of non-communicating cyst has been successfully done and a step by step incision of the septum between the esophageal lumen and the communicating duplication cysts has also been reported.<sup>21,22</sup> To conclude, esophageal duplication cysts must be considered as a rare cause of esophageal symptoms even in adults and the diagnosis may not always be clinched with conventional imaging. EUS may help in diagnosing the lesion if characteristic findings are seen.

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