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ESOPHAGEAL DUPLICATION CYST: A REPORT OF TWO CASES

Gastroenterology

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ABSTRACT

Esophageal duplication Cysts are rare congenital anomalies of the foregut occurring most commonly in lower esophagus. They are commonly detected in childhood, but adult manifestations can occur. Endoscopy is the most valuable tool in diagnosis and management. Thoracoscopy provides excellent exposure and, in most cases, is the preferred approach. Here we present a report of two cases of this rare anomaly

KEYWORDS

Esophageal duplication cyst, Endoscopy, Thoracoscopy

INTRODUCTION

Esophageal duplication cysts are congenital anomalies of the foregut resulting from aberrant development at 3-4 week gestation(1). It is a rare entity, especially so in adults with male predisposition (2).

They represent either simple epithelial-lined cysts, or true esophageal duplication, which is a duplication of the muscularis mucosa and externa without epithelial duplication. Most commonly occur along lower esophagus(3), causing dysphagia by compression.

The majority (about 90%) do not communicate with the esophageal lumen. They are treated either surgically or endoscopically.

In this report we describe case of two adult female patients in whom esophageal duplication presented as masses pressing on the esophageal lumen.

CASE REPORT:

CASE 1

- A 23 year old female ,presented with complaints of dysphagia and retrosternal pain since 2 years
- Upper GI endoscopy showed extrinsic impression at 25 cm from incisor with duodenum and stomach normal.
- Barium swallow revealed Well defined soft tissue density in mid thoracic esophagus on the right side causing luminal narrowing and deviation of lumen to left
- Well defined cystic lesion 3*4cm abutting right lateral wall of esophagus with no contrast enhancement seen on CECT Chest
- Patient underwent VATS cyst excision under endoscopic guidance. Post operative period was uneventful.
- Histopathology report of mass demonstrated Ciliated Columnar epithelium with PseudoStratification and Smooth muscle wall suggestive of Duplication cyst

Case 2

- A 40 year old hypertensive and diabetic female , with complaints of retrosternal pain for 2 months
- Endoscopy showed extraneous impression over right wall of esophagus from 30 to 35cm, stomach and duodenum normal
- CECT chest showed 7*4.8*3.1 cm well defined, non enhancing cystic structure at level of D7 on the right side with indentation on distal thoracic esophagus.
- Patient underwent VATS cyst excision under endoscopic guidance. Post operative period was uneventful.
- Histopathology revealed Ciliated Columnar epithelium with PseudoStratification and Smooth muscle wall suggesting Duplication cyst

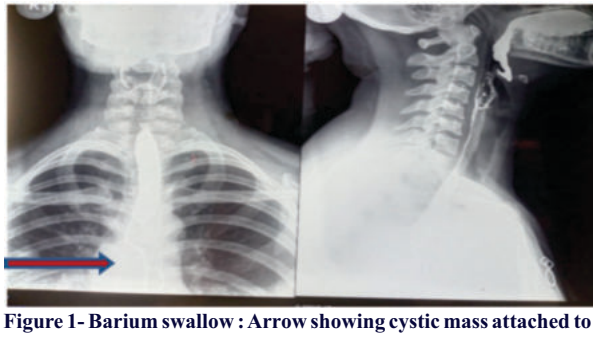

Figure 1- Barium swallow : Arrow showing cystic mass attached to esophagus

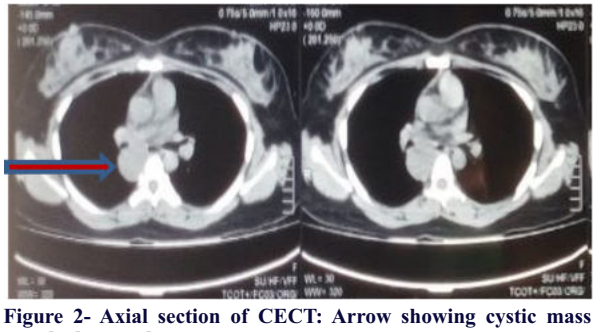

Figure 2- Axial section of CECT: Arrow showing cystic mass attached to esophagus

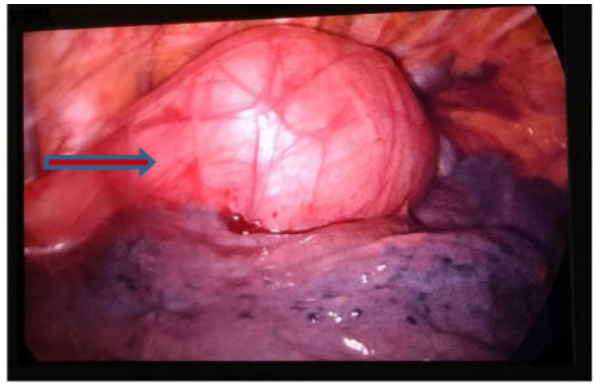

Figure 3- Arrow showing the duplication cyst attached extrinsically to the esophagus

DISCUSSION:

Esophageal duplication are rare congenital malformation with estimated prevalence of 0.0122% (1). According to Bentley and Smith the notochord fuses with the embryonic endoderm and later the endoderm separates from the notochord during the third to fourth week of gestation. If adhesions or a neuroenteric band persists during this separation time, traction diverticula and duplication cysts develop " The Split Notochord syndrome" (4)

Most of them are found in the mediastinum where they present either as separate masses along or in continuity with native esophagus. Intramural esophageal cysts as seen in our case and are considered very rare but more symptomatic in adults (5,6, 7). They have a double layer of surrounding esophageal epithelium. They are most commonly found in lower esophagus; however, cases of intraabdominal esophageal duplication cysts have also been reported (8). Duplication cysts may be cystic, tubular or present as a diverticulum mostly on the right side with cystic type being more common as in our case.

In adults esophageal duplication cyst can present with dysphagia, retrosternal pain, heartburn, reflux esophagitis, and infection(9) . Complications like Bleeding, hematemesis and malignant progression is considered extremely rare(10-12).

Preoperative diagnosis of Esophageal Duplication cysts usually needs a combination of various imaging techniques including Barium Swallow, CECT, MRI, EUS.

Traditionally the diagnosis is suspected on computed tomography, which usually reveals a homogenous cystic mass with regular margins(13) and its communication with the esophageal lumen (14). It also enables to distinguish esophageal duplication cyst from bronchial and vertebral anomalies.

Upper gastroenterological endoscopic examination helps to differentiate intraluminal mass or extrinsic compression and cyst communication with the lumen.

Endoscopic ultrasound (EUS) is a distinguished imaging modality to diagnose the esophageal duplication cyst. We can realize the origin of the mass, anatomical orientation with the surrounding organs (15,16). In addition, it is possible to perform fine needle aspiration biopsy (FNAB) , but is generally avoided so as not to violate surgical planes and introduce infection(17,18).

Magnetic resonance imaging MRI can be included for preoperative evaluation of soft tissue masses to allow for differentiation between benign or malignant tissue and other potential associated abnormalities such as GIST or leiomyomas (19) as these cysts have high signal intensity on T2-weighted images owing to high proportion of water.

Technetium 99m indicated in battery of tests when there is a suspicion of ectopic gastric mucosa.

Definitive diagnosis is according to Palmer's pathologic criteria (8): (I) attachment to the esophageal wall; (II) presence of gastrointestinal tract epithelium; non-keratinizing squamous or ciliated columnar epithelium; and (III) existence of two layers of smooth muscle.

Surgical resection which helps relieving symptoms and prevent future complications remain the treatment of choice for this condition regardless of symptoms. Video-assisted thoracoscopic surgery (VATS) (20,21) and more recently, robotic-assisted thoracoscopic surgery (RATS) are preferred over open surgery (22). For large cystic mass in the lower thoracic to abdominal esophagus combination of thoracoscopic and laparoscopic technique can be successfully used. Endoscopic submucosal tunnel dissection (ESTD) is a newest and challengeable treatment option (23) with short- and long-term favorable outcomes. Esophagogastroduodenoscopy(OGD) can be used intraoperatively for guiding the plane of dissection as was done in our case and also if injury occurs it helps in recognition,which can be managed with primary repair with or without soft tissue(intercoastal muscle, pericardial fat) buttressing.

CONCLUSIONS

Esophageal duplication cyst is rare congenital anomaly whose Definitive diagnosis is established following Palmers criteria. Surgery is the preferred treatment even in asymptomatic cases .

Oesophagogastroduodenoscopy(OGD) can be used intraoperatively to avoid injury and guiding the plane of dissection. Minimally invasive surgery in combination with Oesophagogastroduodenoscopy(OGD) can be a preferable treatment approach.

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