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A Rare Case of Intra-Abdominal Esophageal Duplication Cyst

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Abstract: ***Esophageal duplication cysts are congenital anomalies due to aberrations in the normal development of foregut. Its intra-abdominal variety is an extremely rare occurrence with even fewer reports in the literature. They present with atypical and a variety of symptoms, requiring a high index of suspicion and a multimodal approach for its diagnosis. Treatment is conventionally surgical excision, which with the advent of minimally invasive surgery is now largely laparoscopy based. This paper tends to present the total laparoscopic management of such a case using Indocyanine green (ICG) dye for safer excision. Follow up after 6 months shows the patient to be symptom-free. Further use of this technique and more reported literature will help determine the safety and efficacy of this procedure.***

Keywords: Esophagus, Duplication cyst, Laparoscopic excision, ICG, Minimally invasive surgery

1. Introduction

Gastrointestinal duplication cysts are rare entities which occur most commonly in ileum, jejunum, esophagus, and colon. Esophageal duplication cysts account for 10-15% of all congenital duplication cysts involving GI tract. Majority of these cysts are diagnosed in childhood but when present in adults they are more likely to be symptomatic.

Esophageal duplication cysts are congenital anomalies of the foregut resulting from aberration of the posterior division of the embryonic foregut at 3-4 week of gestation. They represent either simple epithelial-lined cysts, or true esophageal duplication, which is a duplication of the muscularis mucosa and externa without epithelial duplication.

These are considered rare, usually described in single case reports, but their true prevalence remains unknown. Up to 80% of cases are diagnosed during childhood. Most of them are found in the mediastinum and manifest themselves as separate masses along or in continuity with the native esophagus[1].

Symptoms are caused by compression or displacement of surrounding mediastinal structures and comprise dysphagia, respiratory distress, failure to thrive and retrosternal pain. Malignant progression is extremely rare.

Diagnosis is usually made by computed tomography scan or endoscopic ultrasonography.

Treatment of symptomatic esophageal cysts can be either surgical or endoscopic. Surgical resection of the cyst is usually carried out in childhood or in adults, in cases of lesions impossible to treat endoscopically[2]. Surgical treatment is currently moving from thoracotomy to less-invasive procedures, such as video-assisted thoracoscopic surgery, laparoscopic and to endoscopic treatments which, however, still remain challenging interventions.

2. Case Report

Here we present the case of a 24-year-old lady with no medical or surgical history, evaluated for nausea, regurgitation and recurrent colic pain in upper abdomen radiating towards back for past 10 months, but no vomiting or fever. Physical examination and laboratory testing did not reveal any abnormalities. Upper GIendoscopy was normal.

A computed tomography scan of the abdomen showed a cysticmass in the lesser sac of size 4x4x6 cm just below the celiac trunk (Figure 1 and 2).

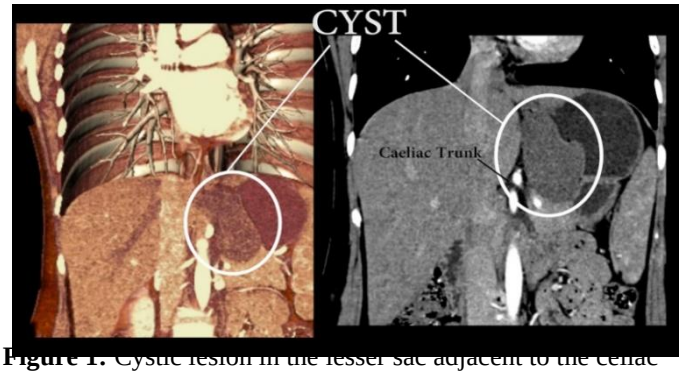

Figure 1: Cystic lesion in the lesser sac adjacent to the celiac trunk (coronal plane)

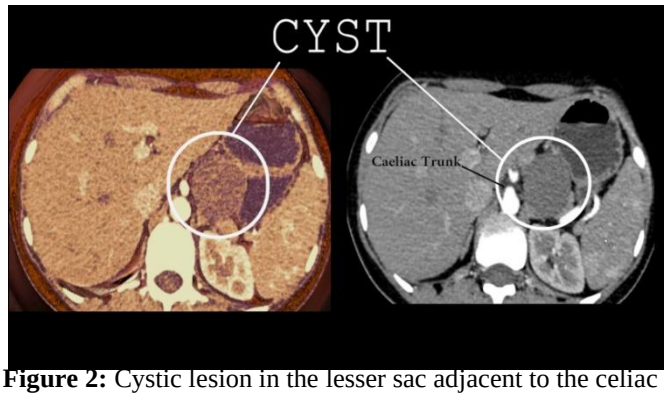

Figure 2: Cystic lesion in the lesser sac adjacent to the celiac trunk (axial view).

Gastro-intestinal duplication cyst was considered as one of the differentials and laparoscopic excision following diagnostic laparoscopy was planned.

After induction of general anaesthesia, patient was placed in supinewith leg split (French) position. Infra-umbilical camera port was placed followed by working ports under vision in a routine manner as for laparoscopic fundoplication. We approached the lesion through lesser sac and observed a 4x4 cm cystic mass in paraesophageal location. Meticulous dissection was carried out using ultrasonic energy device, monopolar and bipolar cautery. At this point ICG (Indocyanine green) dye was injected intravenously and using NIR (near infrared) and Spy mode of laparoscopic camera, further identification of the feeding vessel and delineation of its surrounding anatomy was done(Figure 3).

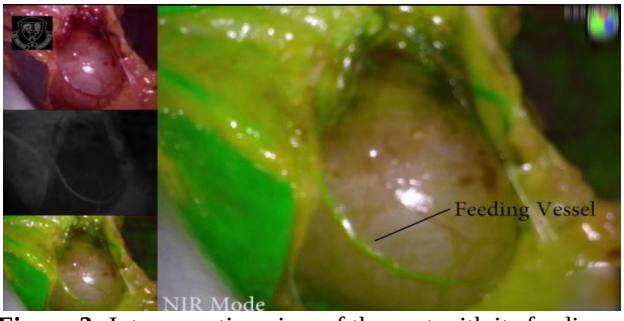

Figure 3: Intraoperative view of the cyst with its feeding vessel (NIR and Spy mode after ICG injection).

During dissection, the cyst ruptured inadvertently. Residual cystic fluid was aspirated and cyst wall dissected off its remaining attachments. Intra-cystic exploration revealed no communication with the lumen of esophagus. The cyst wall was placed in a sterile bag and removed under vision through a workingport.

Post-operative course was uneventful and the patient recovered well. Histopathology reports were consistent with esophageal cyst with squamous epithelial lining and layers of muscularis propria but no bronchial epithelium. At follow up, the patient did not display any of the earlier experienced abdominal symptoms.

3. Discussion and Conclusion

Gastrointestinal duplication cysts are very rare abnormalities. Autopsy studies have shown a prevalence of approximately 1:4500 and esophageal duplications occur even less frequently about 1:8200. In the reviewed literature, four cases appeared in minors and 11 cases in adults. The median age at diagnosis was 50 years [3,4]. Only a few cases have been reported and described in which the cyst is connected to the intra-abdominal part of the esophagus.

Only ten percent of the esophageal cysts communicate with the lumen of the esophagus. Studies show that most cysts are asymptomatic and found incidentally. In case of symptoms the most common complaint is epigastric pain.

Most cysts are surgically removed, of which nearly 50% is by means of laparoscopy. However, as per our research this is the first case reported in which ICGwas used for identification and delineation of cyst.

Besides the location of the cyst, a differentiation between the origin of the cyst can be made. Although it might not have clinical relevance in case of an asymptomatic cyst, the origin can be bronchogenic, esophageal, gastroenteric, neurenteric or pericardial. Gastroenteric and neurenteric cysts are lined with gastric mucosa and pericardial cysts are lined with flattened mesothelium.

To differentiate bronchogenic from esophageal duplication cysts, Palmer's pathologic criteria are useful. These criteria include: (1) attachment to the esophageal wall; (2) presence of gastrointestinal tract epithelium; and (3) presence of two layers of muscularis propria(4). Duplication cysts of bronchogenic origin do not have these two layers of smooth muscle, instead they contain cartilage, bronchogenic glands or both.

The majority (80%) of esophageal duplication cysts becomes symptomatic during childhood and will be removed for that reason. As a consequence, these types of cysts are rarely seen in adults[4,5]. If they are found during adulthood, they are mostly asymptomatic and most case reports are therefore based on an incidental finding during some kind of routine radiological investigation. However, cysts can become

symptomatic, like in our patient, causing abdominal pain, vomiting, dysphagia or reflux. The growing cyst may also cause some mechanical problems of the passage of solid food and fluids because of compression of the esophagus. Finally, these cysts can perforate, they can cause an upper gastrointestinal bleeding or can get infected.

Because of these complications it is recommended to excise the cyst, whether it is symptomatic or not. Minimally invasive excision of these cysts is feasible and recommended in an appropriate setting and experienced hands.Use of ICG is safe and should be considered for better visualisation of the cyst, its surrounding structures and vasculature.

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