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Case Report

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Video-Assisted Thoracoscopy or Laparoscopy for Excision of Esophageal Duplication Cyst

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1. Abstract

1.1. Object: Esophageal duplication is a rare congenital malformation of the digestive tract. We reviewed our diagnostic and therapeutic strategies for two patients.

1.2. Methods and Results: Two patients with esophageal duplication cysts, revealed by dysphagia and diagnosed by Esophagogastroduodenoscopy (EGD), Computed Tomography (CT), and esophageal endoscopic ultrasonography underwent video-assisted excisional surgery in the Digestive Surgery department of Viet Duc Hospital, Hanoi. One patient had transhiatal laparoscopic resection, the other, thoracoscopic excision. No complications or mortality were observed. Postoperative contrast esophageal radiology was considered normal: absence of esophageal fistula or stricture.

1.3. Conclusion: The preoperative diagnosis of esophageal duplication is usually difficult and often is only accurately diagnosed based on post-operative pathology. In our experience, both the laparoscopic and thoracoscopic approaches were safe and effective. This rare pathology requires major surgery, by teams with wide experience in laparoscopic and thoracoscopic surgery.

2. Introduction

Esophageal duplication is a rare congenital malformation of the esophagus, ranking second in enteropathic duplications after ileal duplication, and accounts for about 15% of esophageal congenital malformations. The cystic type is most common (95%); the tubular type is much rarer [1, 2].

Almost all cases of esophageal duplication are detected during gestation by fetal ultrasound or during the first years after birth; complications are rare in adults [3, 4]. Based on its location, position, and size, esophageal duplication can cause different symptoms. The most common are digestive symptoms: substernal pain, dysphagia, vomiting…or respiratory, cardiovascular symptoms such as dyspnea, arrhythmias... However, many patients with esophageal duplication are asymptomatic and their disease is diagnosed fortuitously. Barium esophagogram, esophageal CT, or esophageal endoscopic ultrasound can help to diagnose this disease before surgery, but histopathology is needed for definitive diagnosis [5, 6].

Although debated in asymptomatic adult patients, excision can be proposed because of potential incidents or complications: inflammation, bleeding ulcer, leak, or very rarely, cancer [7]. Surgical cystectomy is indicated for esophageal duplication cysts while esophagectomy is required for esophageal tubular duplication. However, some authors have reported successful results after endoscopic fenestration or resection [3, 5, 8].

Until now, almost all reports concerned traditional open surgery, and there are few centers applying endoscopic surgical techniques in treating this disease. In this report, we discuss the current diagnostic modalities as well as the advantages of minimal access (laparoscopic or thoracoscopic) surgery in the treatment of esophageal duplication.

3. Clinical Cases

3.1. Case 1

This 31year-old male patient, with no antecedent medical history, was hospitalized because of increasing dysphagia which started 4 months ago. An irregular stricture with a smooth border in the middle third of the esophagus was revealed by barium esophagogram. Esophageal endoscopy detected an extrinsic tumor narrowing the esophageal tube; the over-lying esophageal mucosa was smooth. Computed Tomography (CT) showed a well-defined 4 x 6 cm cystic tumor located in the middle third of the esophagus. Endoscopic ultrasound detected a 5-cm cystic tumor with clear borders, located in the esoph-

ageal wall, underneath the esophageal mucosa, 30 to 35 cm below the superior dental arch.

The patient underwent surgery on June 13th, 2018 with the preoperative diagnosis of middle third esophageal cyst.

3.1.1. Thoracoscopic Surgical Technique: After general anesthesia induction and selective endotracheal intubation, the patient was positioned in prone decubitus with 45° rotations to the right. Three 10 mm trocars were inserted (into the right thorax): one along the posterior axillary line in the 7th costal interspace, one on midaxillary line (5th intercostal space), and the third on the anterior axillary line (4th intercostal space).

A soft 5 cm cystic mass was found on the right posterior wall of the middle third of the esophagus, immediately inferior to the azygos vein arch. The mass was easily removed through a longitudinal incision on the esophageal muscular wall. The mucosa was intact. The muscular layer of the esophagus was closed with single layer interrupted 4.0 Vicryl sutures. The operation was uneventful with no intraoperative and postoperative incidents or complications over the 85 minutes span. The patient was discharged from the hospital 8 days after thoracoscopy. The pathology report confirmed that the resected 4 × 5 cm cyst was removed in totality, the specimen was intact, with smooth walls and the cyst was filled with cloudy yellow mucous fluid. Histopathological findings revealed that the cavity was lined by ciliated epithelium; the cyst wall consisted of two layers of smooth muscle without cartilage. Conclusion: esophageal duplication cyst (Figure 1 and 2).

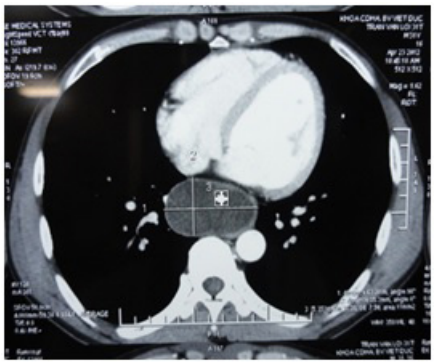

Figure 1: Image of esophageal duplication cyst on chest computed tomography

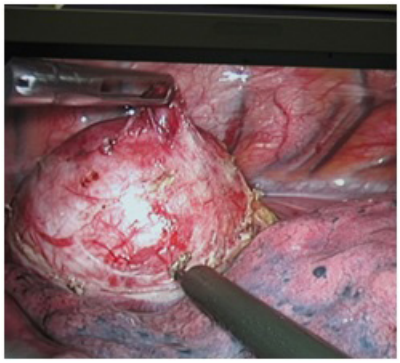

Figure 2: Dissection of esophageal cyst during thoracoscopy

3.2. Case 2

A 37-year-old male patient with no medical and surgical history, presented with a one-month history of dysphagia. He reported no weight loss and was in good body condition. Esophageal endoscopy showed an extrinsic protusion into the esophageal lumen of a 2.5 cm mass, under a smooth uninterrupted mucosa, located about 37cm from the upper dental arch. Thoraco-abdominal CT revealed a cystic 2 × 3 cm mass located in the lower third of the esophagus sized, apparently benign and non-invasive. Esophageal endoscopic ultrasound showed a well-defined 3cm hypoechoic mass under the esophageal mucosa and confirmed the distance from the upper dental arch.

The patient underwent surgery on August 22th, 2018 with the preoperative diagnosis of benign esophageal cyst of the lower esophagus.

4. Surgical Technique

The patient was placed supine, legs spread apart, head elevated 30°. Five trocars were used to enter the abdominal cavity.

After opening the omental bursa through the lesser omentum, the right and left crus of the diaphragm and the posterior wall of the abdominal esophagus were isolated and exposed. The phrenoesophageal ligament was divided, and ultrasonic dissection was pursued to release the esophagus from esophageal hiatus and mediastinum, caution excercised to conserve the anterior and posterior vagus nerves. A soft 3 cm mass was located on the left wall of the esophagus, close to the gastric cardia. A longitudinal incision of the esophageal muscle was made, providing access and allowing dissection of the cystic mass from the esophageal wall. The surgical esophageal muscular wall incision was closed with 4.0 Vicryl single layer interrupted stitches under intraoperative laparoscopic vision to ensure no perforation on the esophageal mucosa. Subsequently, an antireflux valve (Toupet fundoplication) was constructed. The operation lasted 105 minutes. No intraoperative and postoperative complications were noted. Postoperatively, the patient recovered well and was discharged 7 days after surgery. Postoperative pathological results confirmed a 3 cm cyst with a smooth-muscle wall whose lumen was lined by ciliated epithelium. Conclusion: Esophageal duplication cyst (Figure 3 and 4).

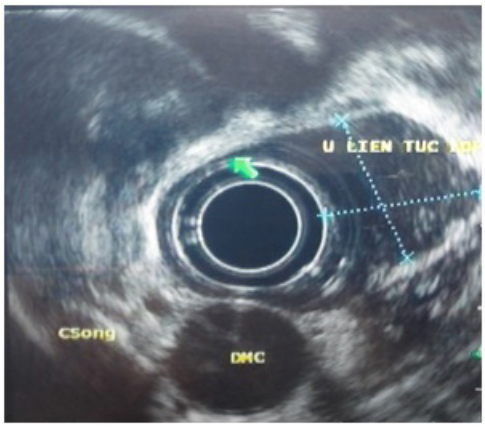

Figure 3: Image of esophageal duplication cyst on esophageal endoscopic ultrasound.

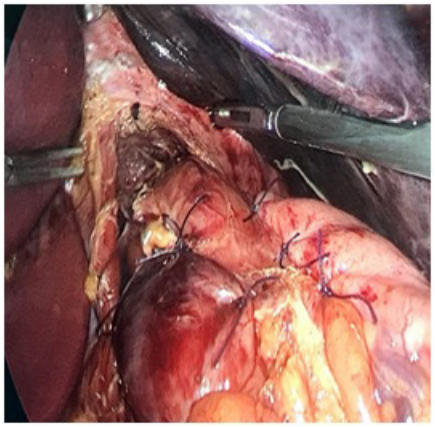

Figure 4: Toupet fundoplication during laparoscopy.

5. Follow-Up After Surgery

Both patients were seen 3 months, 6 months, and 1 year after surgery. The postoperative results were satisfactory: both patients were able to digest food normally without dysphagia or choking. Neither had stricture or symptoms of gastro-esohageal reflux.

6. Discussion

Esophageal duplication is a rare disease, especially in adults; consequently, there are no unified indications for treatment.

Most reports on treatment for esophageal duplication cyst involve cyst resection by open thoracotomy or thoracoscopy; esophagectomy is recommended for tubular-type esophageal duplication [4, 5]. Endoscopic management, opening the anomaly into the main esophagus through a flexible endoscopic has been reported with good initial results [2, 8].

Both patients in our study were symptomatic. Both had cystic type duplication and absence of communication with the main esophagus. No other congenital malformation was found. While some authors believe that surgical intervention should only be entertained when the disease is symptomatic or causes complications [3], we, as others, consider that surgery is necessary due to the risk of complications such as ulceration, perforation, inflammation... even the risk of malignant degeneration [4, 5], even when the disease is asymptomatic, especially now with the development of minimal access surgery. We systematically use laparoscopy (or thoracoscopy) for benign pathologies of the esophagus.

Approximately 60% of esophageal duplications occur in the lower third of the esophagus, 20% in the middle third, and 20% in the upper third. About 50% of cases of esophageal duplication are associated with another malformation, especially the spine and other parts of the gastrointestinal tract [2, 5]. Thoracoscopy is indicated for lesions in the upper and middle third of the esophagus, and laparoscopy is indicated for lesions in the abdominal esophagus or the lower portion of the thoracic esophagus near the cardia [9].

Most of cases of esophageal duplication are diagnosed in the early years after birth, cases detected in adults being very rare. Especially nowadays, thanks to the development of prenatal ultrasound, it is possible to diagnose esophageal duplication during pregnancy, thereby allowing earlier intervention and better results. After birth, about two-thirds of cases of esophageal duplication are diagnosed before 6 months, about three-quarters of cases are diagnosed before 2 years of age [4], and only a small percentage are found in adulthood. Depending on the location, size and type, the disease causes different symptoms [3, 6]:

These include respiratory symptoms when lesions are located in the upper third of the esophagus: persistent cough, difficulty breathing, wheezing, pneumonia... ; digestive symptoms such as dysphagia, choking, vomiting… when the lesion is large and compresses the esophagus, arrhythmia occurs when the lesion is located in lower third of the esophagus near the heart, and rarely upper gastrointestinal tract bleeding due to bleeding ulcers in the cyst, and even mediastinitis due to perforation of the cyst.

In our study, both patients were diagnosed with the esophageal duplication disease in adulthood with gradually increasing dysphagia. Because the tumor was located in the middle third and lower third of the esophagus and was relatively small in size, there were no respiratory and cardiovascular disorders.

Imaging has an important role in the diagnosis of esophageal duplication. The most widely used include chest X-ray, esophago-gastro-duodenal follow-throughs, chest computed tomography and esophageal endoscopic ultrasound [6].

On chest X-ray, the most common picture is a round or oval opacity with smooth margins in the posterior mediastinum, usually in the lower third of the esophagus. In addition, chest X-ray can detect malformations of the spine such as spina bifida, kyphoscoliosis... When there is a blurry mass in the posterior mediastinum accompanied by a congenital malformation of the spine on chest X-ray, esophageal duplication should be considered.

Esophago-gastro-duodenal follow-through: in cases of esophageal duplication with communication to the esophageal lumen, the image is one of a round cyst located adjacent to the main esophagus or a tube lying along the border of the esophagus with contrast uptake. Conversely, if there is no anastomosis between the main esophagus and the esophageal duplication, the image observed reveals a filling defect or amputation with a regular border, due to the cyst pressing on the lumen of the esophagus.

Esophageal endoscopic ultrasound allows to confirm the hollow nature of the cyst, the size and location of the cyst, and the relationship between the cyst and the adjacent organs. Endoscopic ultrasound with a high-frequency transducer can identify whether the cyst is covered by a muscular layer which is continuous with the esophageal muscle layer.

Chest computed tomography is a commonly used method to assess mediastinal tumors in general and esophageal cysts in particular. Not only does chest computed tomography with contrast enhancement allow assessment of the location, size and nature of the cyst but it also helps to evaluate the nature of the esophageal duplication cyst wall; contrast uptake of the cystic wall similar to the wall of the

esophagus is an important sign to help diagnose esophageal duplication cyst.

Both of our patients had symptoms leading to a full range of imaging methods to help diagnose the disease.

In 1937, Ladd suggested the use of the term "duplications of the alimentary tract" and applied the term to congenital lesions having three characteristics: (1) the presence of a well-developed coat of smooth muscle, (2) an epithelial lining representing some type of intestinal tract mucosa, and (3) intimate anatomic association with some portion of the gastrointestinal tract. The histopathological criteria for classifying a foregut duplication cyst as an esophageal cyst were developed by Arbona et al. [1] include attachment to or inclusion of the cyst to the esophageal wall, presence of two muscular layers, and squamous, columnar, cuboidal, pseudostratified, or ciliated epithelium lining.

The post-operative pathology results of our two cases met the esophageal duplication cyst criteria, helping to confirm the diagnosis of the disease.

Both of our patients had good results, without any complications, attesting to the safety of surgery. Follow-up after 3 months, 6 months and 12 months of surgery showed that the patient recovered well, complete regression of preoperative symptoms without any complications or sequelae.

Minimal access surgery avoids large incisions, providing less post-operative pain, faster recovery and reduced scarring. Surgical times for our two cases were 85 minutes and 105 minutes which was acceptable. However, this rare pathology requires major surgery and therefore should imply teams with wide experience in minimal access surgery.

7. Conclusions

Esophageal duplication is a rare disease; preoperative diagnosis is often based on clinical symptoms and paraclinical tests, especially imaging: endoscopy, chest CT, and endoscopic ultrasound of the esophagus… However, the definitive diagnosis is based on postoperative pathology.

Once esophageal duplication is diagnosed, we believe that surgery is indicated due to the risk of complications such as ulceration, perforation, inflammation… even the risk of malignant degeneration, even if the lesion has not caused any symptoms, especially when minimal access techniques are available.

This study showed that video assisted thoracoscopic or laparoscopic was safe and effective for the treatment of esophageal duplication cyst when performed in a center with wide experience in minimal access surgery.

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