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Case Report

A sudden hemorrhage in the esophageal duplication cyst: A rare cause of acute dysphagia in an adult

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ABSTRACT

A 42-year-old male pediatrics physician was admitted with a history of acute chest pain and sudden severe dysphagia to solids and liquids. He denied any history of abdominal pain, vomiting, dyspnea, nausea weight loss. He could not even swallow saliva. The patient denied any history of drug abuse. A computed tomographic scan of the chest (CT)showed a posterior mediastinal mass inseparable from the esophagus and descending aorta. Magnetic resonant imaging (MRI)scan revealed a cystic mass full of blood inseparable from the esophagus and adherent to the aorta. The mass was resected entirely through the left thoracotomy; post-operative recovery was uneventful; dysphagia resolved as the post-operative contrast swallow study showed a free flow of contrast to the stomach patient resumed his regular diet.

1. Introduction

The esophageal duplication cyst is an embryonic malformation first reported by Blassium in 1711. It is a rare form of congenital gastrointestinal duplication cyst found commonly in the esophagus ileum, jejunum, and colon. Pathogenesis is due to incomplete vacuolization during embryogenesis's fifth to eighth week and migration into the esophageal wall [1,2]. Esophageal duplication cysts are primarily diagnosed in early childhood, and in adults, there are primarily asymptomatic and are incidentally discovered during the radiological examination. They are more common in the distal esophagus. They are often asymptomatic until adulthood, and they can cause chest pain, dysphagia, esophageal hemorrhage, and recurrent respiratory infection [3,4]. Before surgical resection, a comprehensive evaluation of the mass and adjacent structures and other concurrent systemic anomalies is mandatory. This case is reported in line with scare criteria [5].

2. Case report

A 42 years old male was admitted through the Emergency Department with a history of sudden onset of dysphagia and chest discomfort. Nothing significant in the past medical history. He was unable to swallow saliva and was constantly spitting in a bag. Basic blood investigations showed Hemoglobin of 9mg/dl, normal white cell count, and normal renal and liver panels. Chest X-Ray and computed tomographic scan (CT)revealed a mediastinal mass adherent to the lower esophagus. A multiplanar and multi sequential Magnetic resonant imaging scan (MRI) with contrast showed 12.2×8cm lower chest posterior mediastinal cystic mass with no soft tissue enhancement, and there is severe mass effect on the lower esophagus and displacement of the heart anteriorly; most likely, there is bleeding in the esophageal duplication cyst Fig. 1 (A, B&C &D. Endoscopy revealed obstruction in the middle part of the esophagus. In a multidisciplinary meeting, we discussed surgical management with the radiologist, gastroenterologist, anesthetist, and intensivist.

The chest was entered through the 7th intercostal space via the left posterolateral thoracotomy. We found a large mass along the lower esophagus and descending aorta with visible feeding blood vessels. The branches from the aorta were ligated, and with meticulous dissection, the esophagus was identified, and EDC was resected without any damage to the esophagus. The cystic mass was full of blood and clots. A nasogastric tube was inserted, and with one chest drain, 30Fr chest was closed in layers as routine. A Gastrografin swallow study on post operative day 2 revealed free flow of contrast to stomach with no evidence of leakage and the patient started oral intake Fig. 2 (C&D) Dysphagia resolved, and the patient was discharged after five days in the hospital for further follow-up in outpatient. Histopathology report of the resected specimen revealed features consistent with foregut-enteric duplication

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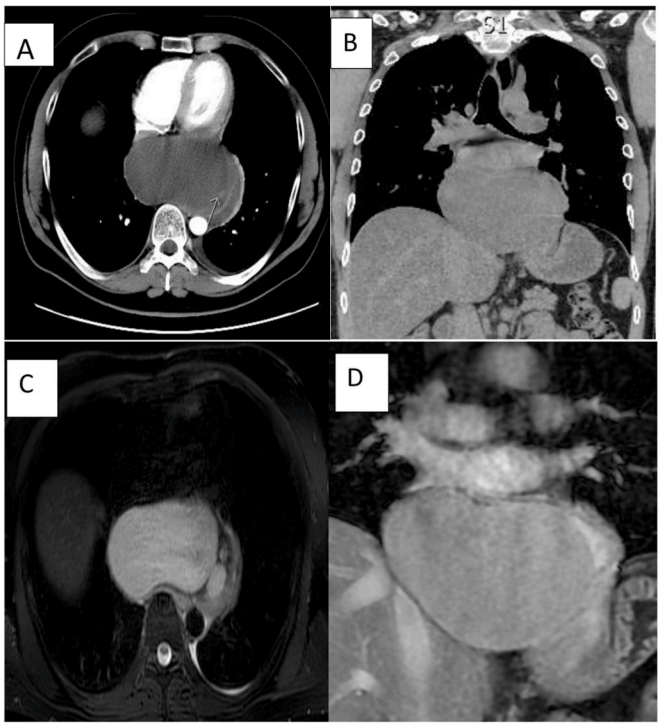

Fig. 1. (A) CT scan chest showing large cystic mass with severe compression on esophagus. (B) CT scan chest coronal view showing mediastinal mass with cardiac and esophageal displacement. (C&D) MRI scan of thorax showing posterior mediastinal hyperintense cystic lesion with severe mass effect on esophagus and displacement of heart anteriorly, no evidence of soft tissue enhancement. (Hemorrhagic duplication cyst).

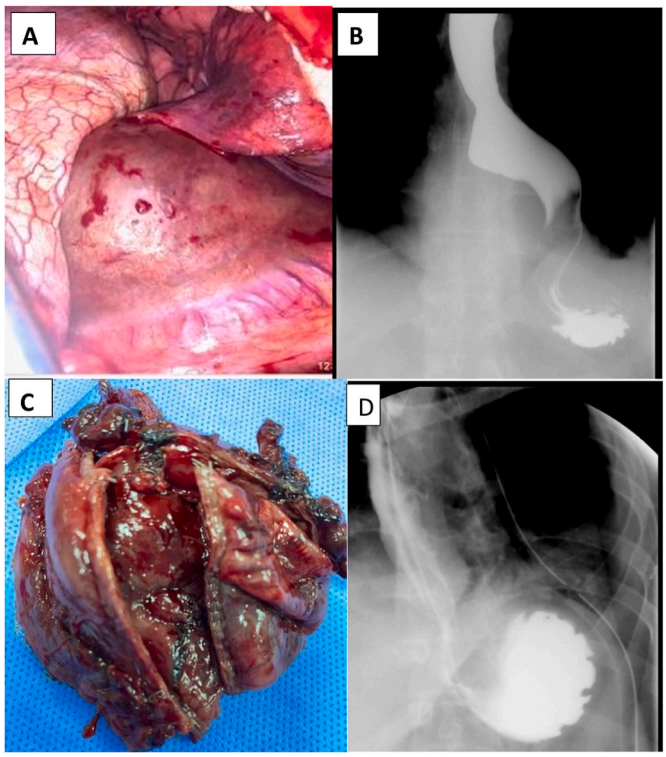

Fig. 2. (A) Operative view of hemorrhagic cystic mass after thoracotomy prior to surgical excision. (B) Pre operative oral contrast study showing dilated esophagus and obstruction at lower end. (C) Resected specimen of esophageal duplication cyst(D)Post -operative oral contrast study showing free flow of contrast without any leak.

cyst associated with aggregates of hemosiderin-laden macrophages. Fig. 2 (A&B).

3. Discussion

Esophageal duplication cyst (EDC) is an exceedingly rare congenital anomaly; the reported estimated incidence is 1:82000 (0.0122%) with male predominance; male to female ratio is 2:1. It accounts for 10% to 15% of all gastrointestinal tract duplication cysts and 0.5%–2.5% of esophageal tumors [6,7]. There are three types of EDC, cystic, tubular, or diverticulum shaped; cystic type is the most common [8]. The majority of these cysts, 60%, are located in the lower third of the esophagus,17% are in the middle, and 23% are in the upper third. These cysts in the upper part of the esophagus are usually diagnosed in infancy due to respiratory symptoms, dysphagia, and impairment of growth [9]. These are generally incidentally diagnosed during radiological examination for some other problem in adults. As these cysts continue to develop, they become symptomatic in adults. They can cause mass effects on the surrounding structures patient present with chest or epigastric pain, cough, stridor, retrosternal back pain, and dysphagia [10,11]. Infection is the most common complication; in addition to that, rupture, intracystic bleeding, peptic ulcer, tracheal obstruction, cardiac arrhythmias, and tamponade due to rupture into the pericardial space have been reported. Galinsky et al. reported an esophageal cyst with massive hemorrhage, which was managed with esophagectomy [12–14].

The majority of EDC (80%) are diagnosed in early childhood due to recurrent respiratory infections and slow growth. Whereas symptomatic EDC is rarely encountered in adults (<7%), most are discovered as an incidental finding on chest Xray [15].

The EDC is sometimes identified on routine endoscopy; an esophageal lumen is narrowed by external compression with normal mucosa; therefore, it is indistinguishable from benign esophageal tumors such as lipoma leiomyoma, gastrointestinal stromal tumors [16].

Ultrasonography may be helpful in a preliminary evaluation. But the best diagnostic imaging modalities for the mediastinal cystic lesion are CT scans and MRI. Magnetic resonance imaging (MRI) has a higher diagnostic value in evaluating the consistency and contents of mediastinal masses [17,18,19]. Transthoracic echocardiography is indicated if pericardium is involved or if there is a history of arrythmia [20]. Such patients pose numerous challenges for anesthesia providers due to mass effect and anatomical structure involved in the cyst Airway management, and central venous access may be difficult; in addition to those, patients with EDC are at risk for pneumothorax, pneumomediastinum, profound hemorrhage, vascular injuries gastric aspiration and cardiovascular collapse due to mass effect. A multidisciplinary team approach, including gastroenterologist, surgeon, anesthetist, and intensivist, is recommended [21,22]. The only definitive treatment for esophageal cyst is surgical excision. The best treatment is a conventional approach through a posterolateral thoracotomy and complete surgical resection [23,24]. Other surgical techniques, video-assisted thoracoscopic surgery (VATS) and robotic-assisted thoracoscopic surgy (RATS) have been reported. Although VATS AND RATS have the advantages of less post-operative pain, early recovery, and better cosmoses, their role is limited to pediatric and uncomplicated EDC resection [25,26]. For large EDC with complications, open surgical resection is the best approach as we manage our patients successfully [27]. A few sporadic case reports have advocated nonoperative management in asymptomatic EDC and follow-up with periodic ultrasound examination [28].

Surgery is recommended for EDC in every case, even though it may be asymptomatic due to the risk of malignant transformation. Salo et al. reported surgical complications, heartburn esophagitis, and (1%) mortality after surgical resection of esophageal cysts in pediatric and adult patients.[29,30]

4. In conclusion

We report a rare case of acute dysphagia in an adult due to sudden massive bleeding in the EDC. The source of bleeding was from the descending aorta, the feeding vessels were ligated, and complete surgical resection of EDC was accomplished without any damage to the esophagus. The conventional surgical approach is an excellent and safe option for treating complicated esophageal duplication cysts.

Ethical approval

IRB approval.

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Author contribution

Ikram ul Haq Chaudhry, Main author operating surgeon. Mohammed A Alsuhaimi Wrote introduction. Abdullah M Al Ghamdi, Wrote abstract. Maryam Almalali structured abstract. FatimaAlmalali searched references. Yousif A Alqahtani, structured abstract. Meenal A Al Abdulhai highlights, imaging and histopathology. Othman M Al Fraih, Wrote part of discussion.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request".

Registration of research studies

1. Name of the registry: Research registry
2. Unique Identifying number or registration
3. Hyperlink to your specific registration (must be publicly accessible and will be checked): http://www.researchregistry.com/browse-the-registry#home/

Guarantor

Ikram ul haq Chaudhry.

Declaration of competing interest

No conflict of interest and there was no funding or financial assistance in this case.

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