Infection of an esophageal cyst following endoscopic fine-needle aspiration

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Abstract
In this report, we describe an unusual presentation of an esophageal cyst. Esophageal cysts are generally benign and are frequently asymptomatic until progressive enlargement leads to symptoms of obstruction. Incidental discovery usually warrants excision. In the described case, a patient presented with signs of enlargement and concerns for infection after an attempted endoscopic biopsy of the lesion. After admission and initial management with antibiotics she was taken to the operating room for resection via a thoracotomy. We review the literature and underscore the conventional practice of operative management of esophageal cysts without the use of invasive diagnostic evaluation.

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1. Introduction
Esophageal cysts are the second most common benign esophageal lesion after leiomyoma, yet their occurrence is rare and represents a spectrum of pathology which includes true esophageal duplication cysts, bronchogenic cysts, and enteric cysts.1 Esophageal cysts are frequently asymptomatic until progressive enlargement leads to symptoms of obstruction. They may also present with complications, including ulceration, hemorrhage and infection.1,2 It is generally accepted that even incidental discovery is indication for surgical excision; however, here we present a patient who was referred to us after clinical deterioration following endoscopic biopsy of the cyst.

2. Presentation of case
We present the case of a 41-year-old female with a significant past medical history of Crohn’s disease necessitating a subtotal colectomy with end ileostomy. Of note, her Crohn’s was stable and she was not on any form of immunomodulatory therapy at the time of this presentation. During a workup for dysphagia she was found to have a mass in the posterior mediastinum. She was evaluated by endoscopic ultrasound which revealed a 4 cm × 1 cm mass which appeared homogenous with concern for malignant potential. An endoscopic ultrasound-guided fine needle aspiration of the mass was performed which yielded thick material. The final cytology demonstrated proteinaceous debris with abundant macrophages which was consistent with the working diagnosis of an esophageal duplication cyst.

Five days after her endoscopic procedure she complained of increasing chest pain and odynophagia. She presented to our institution’s emergency room where she underwent a routine workup for chest pain. She was admitted for observation, with an overall clinical picture concerning for presence of infection with low grade fever. Her laboratory tests at this point indicated a leukocytosis of 12,800 with 86% neutrophils. A chest CT and barium swallow revealed the cyst had enlarged (Figs. 1 and 2). On EUS the cyst measured 4.0 cm in maximal diameter, and five days later this had increased to 6 cm at the time of her symptoms. She was started on broad spectrum antibiotics. Despite this, she developed febrile episodes with temperature spikes of 102.2°F. Over the course of 24 h she had no resolution of her chest discomfort. Although her leukocytosis resolved on antibiotic therapy, the decision was made to take her to the operating room for exploration given her ongoing pain and fevers.

The distal esophagus was exposed through a right thoracotomy, and the cyst was readily identified beneath the inferior pulmonary ligament in an intramural location (Fig. 3A). During the mobilization, the cyst ruptured and purulent material was collected and cultured. A complete excision was possible (Fig. 3B), and following esophagoscopy to confirm the integrity of the esophageal mucosa, the chest was closed with chest tubes placed for drainage. Final cultures of the cyst material revealed only white cells without any organisms (Fig. 4).

Our patient made a rapid recovery with chest tubes and nasogastric tube being removed by postoperative day five. She was commenced on a diet and did well.
3. Discussion

Esophageal duplication cysts represent a rare congenital anomaly found in the posterior mediastinum. In the case of a true esophageal duplication they are intramural without communication with the esophageal lumen. They are generally benign but malignant degeneration has been reported. More commonly, acute presentation is due to ulceration, hemorrhage or infection, which can occur due to the presence of gastrointestinal epithelium within the cyst.

Esophageal cysts should be removed when found incidentally due to the likelihood of progression in size and the risk of complications. Modern imaging modalities including CT and MRI allow for excellent characterization of esophageal masses and can with some certainty predict the fluid-filled nature of these benign lesions. Barium swallow will reveal an extrinsic, smooth-bordered mass.

Endoscopy is generally performed in these cases to rule out the presence of mucosal abnormalities. Benign lesions will demonstrate normal smooth esophageal epithelium, with evidence of extrinsic compression. Endoscopic ultrasound (EUS) will further characterize the lesion and demonstrate whether or not the mass is contained within the esophageal musculature. EUS-guided fine needle aspiration (FNA) is increasingly performed in the diagnosis and staging of intrathoracic malignancy and has an excellent safety profile in the setting of solid masses with a complication rate of approximately 0.5%. However, this is not true in the case of cystic lesions where up to a 14% rate of complications has been described. In general, an esophageal mass which does not present with mucosal abnormality should not be biopsied or sampled with either EUS-FNA or biopsy forceps. Complications include infection, hemorrhage, and mediastinitis.

Fig. 1. A contrast chest CT demonstrates a homogenous lower mediastinal mass with lateral displacement of the esophagus.

Fig. 2. Barium swallow demonstrates extraluminal compression of the esophagus without evidence of communication between the cyst and the esophageal lumen.

Fig. 3. (A) The large intramural esophageal cyst is displayed here after taking down the inferior pulmonary ligament. (B) The cyst was completely excised.

Fig. 4. The pathology specimen demonstrates a well-defined cyst wall, with ulcerated tissue lining. Cartilaginous components were noted. The cyst fluid evaluation contained large amounts of polymorphonuclear white blood cells.
In our patient it is likely that the EUS biopsy led to bleeding within the cyst and likely superimposed infection given the presentation of increasing pain, dysphagia, fever and leukocytosis. Our operative cultures were not taken until a 48-h course of broad-spectrum antibiotic therapy had been followed, which likely explains the bland purulence found at operation. Although rare, infection of an esophageal cyst after EUS-FNA has been described previously.\(^5\) Infection in that case, described by Trojan and colleagues, was with Candida albicans. However, any organism colonizing the esophagus may be introduced into the previously sterile cyst fluid. The indication for EUS-FNA in Trojan’s case was concern that this lesion may be malignant given the patient’s history of a high-grade non-Hodgkin’s lymphoma. The authors comment that the practice of EUS-FNA should be avoided when an esophageal cyst is suspected without overwhelming concern for another pathology which might preclude operative management.

4. Conclusion

In summary, our case illustrates again that conventional practice of operative management of esophageal cysts without the use of invasive diagnostic tests should remain the standard of care. The differential diagnosis of posterior mediastinal cystic masses is sufficiently limited such that tissue diagnosis is not required. The hazards of performing EUS-FNA in the setting of a cystic lesion outweigh any potential benefit.

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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.

Conflict of interest statement

The authors have no conflicts of interest to disclose with regard to this case report.

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