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Case report

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Streptococcus pyogenes infection of a mediastinal cyst after endoscopic ultrasound-guided fine-needle aspiration

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HIGHLIGHTS

- EUS-FNA to mediastinal cystic lesions can cause life-threatening infection.
- Mucin-filled mediastinal cysts can show soft tissue density on CT/MRI.
- Mediastinal infection after FNA was latent initially and progressed rapidly later.
- Prophylactic antibiotics do not necessarily reduce the risk of mediastinal infection.
- FNA should be avoided if mediastinal cystic masses cannot be ruled out.

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ABSTRACT

Mediastinal masses are uncommon and difficult to diagnose. Endoscopic ultrasound with fine-needle aspiration (EUS-FNA) is a minimally invasive technique for diagnosis of mediastinal lesions with few complications. Our report described a mediastinal bronchogenic cyst with soft tissue density infected by *Streptococcus pyogenes* (*S. pyogenes*) after EUS-FNA, accompanied by respiratory cardiac arrest and superior vena cava syndrome. The patient underwent cardiopulmonary resuscitation to gain the chance for emergency surgery and recovered. Clinicians should be aware that mediastinal mass with soft tissue density shown on imaging may be mediastinal cyst containing high density mucin, FNA should be avoided if cystic masses cannot be ruled out.

1. Introduction

Mediastinal masses are relatively uncommon, usually incidentally discovered on radiographic examination in asymptomatic patients. Mediastinal masses can be benign and malignant (Duwe et al., 2005). Due to the proximity of many critical structures, it is difficult to obtain a sample of mediastinal mass tissue, which adds to the diagnostic and therapeutic challenges for physicians. EUS-FNA is a minimally invasive and safe technique which is helpful for the diagnosis and sampling of mediastinal masses, but patients with mediastinal cystic lesions have a higher risk of infection (Bartheld et al., 2012; Wildi et al., 2003). To our knowledge, there are no reported cases of mediastinal cyst infected by *S. pyogenes* after EUS-FNA which cause respiratory cardiac arrest. Our report describes a case of a mediastinal mass which developed to a life-threatening mediastinal abscess after EUS-FNA accompanied by cardiac and respiratory arrest, and discusses the role and implications of EUS in dealing with such cases.

2. Case presentation

A 33-year-old female presented with mediastinal mass was referred to us for EUS-FNA. Chest computed tomography (CT) revealed a mass of soft tissue density in the mediastinum adjacent to esophagus, about 4.2cm*3.5cm, the CT value of which was 50HU. During EUS imaging, a hypoechoic lesion of 40mm was seen outside the esophagus and close to thoracic aorta. The internal echo was uniform without obvious blood flow signal (Figure 1A). The lesion was diagnosed as ectopic thyroid possibly. A 22-gauge fine aspiration needle (COOK-ECHO-3-22) was successfully directed through the wall of the esophagus and into the lesion under direct ultrasound guidance. The needle core was slowly pulled out and aspiration was then performed by simultaneously moving the needle in a to-and-fro motion 10–20 times inside the target lesion while applying negative pressure using a 10 mL syringe. This operation was performed twice, and cytological smear and histopathology were sent at the same time. No specific pathological tissue was found due to

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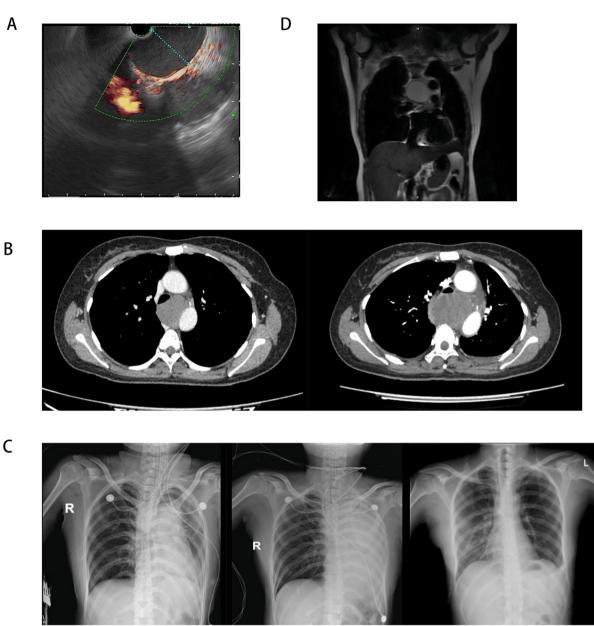


Figure 1. (A) Mediastinal hypoechoic mass presented on EUS. (B) CT demonstrated mediastinal mass growing with time: (L) before EUS-FNA, (R) 7th day after EUS-FNA. (C) CR after respiratory cardiac arrest: (Left) partial occlusion of left bronchus, (Middle) Complete left atelectasis in 2 h, (Right) remission of atelectasis 5 days after surgery. (D) MRI showed the enlarged mediastinal mass had the same signal with muscles.

too few puncture components, and cytological pathology showed well differentiated epithelium.

The patient had no discomfort within 5 days after FNA and no periprocedural antibiotics was used. On day 6, high fever and chest pain occurred. Physical examination revealed blood pressure of 109/77 mmHg, heart rate of 92 bpm, respiratory rate of 20 breaths/min, body temperature of 37.9 °C, and clear respiratory sounds in both lungs without rales. Laboratory findings showed a white cell count of 12,110/ μ L, hemoglobin level of 14.6 g/dL, C-reactive protein 20.85 mg/L, and arterial pressure of oxygen (PO2) 82 mmHg. Chest CT showed mediastinal mass went slightly larger. The patient was given meropenem as anti-infective therapy.

On day 7, the patient had dyspnea with low breath sound in left lung. CT indicated further enlargement of mediastinal mass, and left main bronchus was compressed (Figure 1B). Thoracic surgery consultation recommended enhanced magnetic resonance imaging (MRI) to identify cystic or solid lesions before surgery. During MRI, the patients had sudden convulsions and loss of consciousness (Figure 1D). Cardiopulmonary

resuscitation and early endotracheal intubation were given immediately, and the patient regained consciousness about 5 min later. Laboratory findings showed a white cell count of $31,400/\mu$ L, hemoglobin level of 14.4 g/dL, C-reactive protein 261.62 mg/L, and PO2 59 mmHg. Bedside chest radiograph showed complete atelectasis of left lung (Figure 1C). Emergency thoracotomy was performed from the right chest. There was obviously high tension in the mediastinum, and superior vena cava were compressed. The cyst wall was cut open above and below Azygos vein arch, and about 100 ml milky fluid was discharged (Figure 2). Bacterial culture of cyst fluid showed *S. pyogenes*, which was sensitive to penicillin and vancomycin. After incision, oxygen saturation and blood pressure of the patient quickly returned to normal. The endotracheal intubation was removed 1 day after surgery, and the patient was discharged 1 week later without neurological sequelae. The final diagnose was mediastinal bronchogenic cyst infection.

Informed consent was obtained from the patient for publication of this case report.

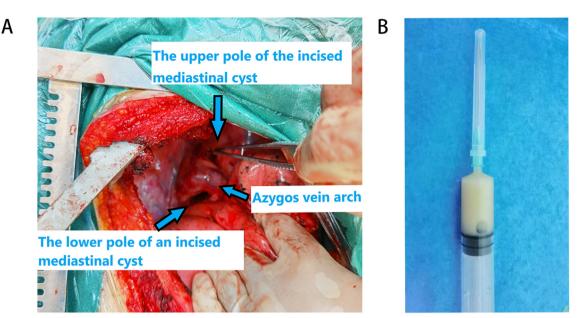


Figure 2. (A) Abscess incision. (B) Milky fluid extracted from mediastinal cyst.

3. Discussion

The mediastinal masses contain a variety of benign and malignant lesions, such as thymoma, bronchogenic cyst, neurogenic tumors, lymphoma, and esophageal tumors (Herth et al., 2010). Confirmation usually requires tissue sampling, either by taking a biopsy before treatment or by removing the entire mass (Patel et al., 2016).

EUS can be used for observation and sampling of pancreatic, gastrointestinal, mediastinal and retroperitoneal masses (Ardengh et al., 2011). EUS-FNA are generally safe and have a low complication rate (<1%) (Jenssen et al., 2012). Complications of EUS-FNA have been reported, including infection, hemorrhage, perforation of the esophageal wall, fistula formation, pneumothorax or mediastinal gas. EUS-FNA related complications, especially infectious complications, occurred more frequently in patients with cystic lesions (Bartheld et al., 2012). In a prospective cohort study of 457 patients, patients with solid lesions had a lower complication rate than patients with cystic lesions (0.5% vs. 14%) (Wiersema et al., 1997). Since the risk of infection in patients with mediastinal cystic lesions is higher, prophylactic antibiotics are usually given to such patients before FNA and continue until 3–5 days after the operation (Annema et al., 2003).

However, in clinical practice, it is sometimes difficult to distinguish between cystic and solid lesions. Mucin-filled cysts can appear hypoechoic rather than anechoic on EUS, and CT values are within the range of soft tissue density. CT and MRI may misdiagnose a cystic duplication lesion as a solid lesion in about 70% of cases (Fazel et al., 2005). Because of its intrinsic superior soft tissue resolution, MRI is an important tool in the evaluation of mediastinal masses that are incompletely characterized on CT (Madan et al., 2018). In our case, EUS showed a hypoechoic lesion rather than anechoic, which was diagnosed as a benign soft tissue lesion with high possibility of ectopic thyroid, so FNA was performed without prophylactic antibiotics. CT value of the lesion was 48-50 HU, within the range of soft tissue density. Even when the cyst was significantly enlarged by severe infection, the CT value just decreased slightly (50-47 HU). Contrast-enhanced ultrasound in combination with endoscopic ultrasound (CE-EUS) uses ultrasound contrast agents to delineate and identify lesions based on their pattern of vascular enhancement. Ultrasound contrast agents are microbubbles with 1-10 microns in size which are equal to or smaller than red blood cells (Jang et al., 2013). CEUS is most often used for imaging the liver, but it can also be used to image other organs (Piscaglia et al., 2012). In our hospital, CE-EUS is usually done for the evaluation of the liver and pancreatic lesions, and the patient did not receive contrast-enhanced ultrasound. The application of CE-EUS in more cases should be increased in the future.

S. pyogenes is a major human-specific bacterial pathogen that causes a wide array of manifestations ranging from mild localized infections to life-threatening invasive infections. Approximately one-third of patients with invasive pyogenic streptococcal infection progress to infectious toxic shock syndrome, characterized by shock and multiple organ failure, which can be life-threatening (Darenberg et al., 2007; Waddington et al., 2014). Streptococcus infection (Mahady et al., 2011) has been reported in several cases of mediastinal infection after FNA. In this case, mediastinal infection of S. pyogenes after FNA was insidious in early stage, but progressed rapidly on day 7. Within one day, the enlarged cyst compressed vital organs in the mediastinum, and caused life-threatening complications including respiratory cardiac arrest, superior vena cava syndrome, and atelectasis. The patient underwent cardiopulmonary resuscitation and emergency surgical abscess incision. If emergency surgery was not performed in time, there was a risk of cyst rupture. We administered an adequate dose of meropenem which is sensitive to streptococcus, but failed to prevent rapid progression of infection. It was reported prophylactic antibiotic didn't necessarily reduce the risk of mediastinal cystic infection (Diehl et al., 2010), which maybe because cyst wall limits the effectiveness of antibiotics.

In conclusion, EUS-FNA for mediastinal masses should be performed with great caution. Clinicians should be aware that mediastinal soft tissue density masses may be bronchogenic cysts containing high density mucin. If mediastinal lesions are clinically suspected to be cystic, surgical resection is the first choice. If FNA is unavoidable, sufficient prophylactic antibiotics must be used throughout the course. The time of fasting and duration of hospital observation after EUS-FNA should be extended to avoid the life-threatening mediastinal infection.

Declarations

Author contribution statement

All authors listed have significantly contributed to the investigation, development and writing of this article.

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Declaration of interest's statement

The authors declare no conflict of interest.

Additional information

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