

CASE REPORT

Encephalitis as a complication of a spinal-esophageal fistula due to discitis

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Abstract

In patients with back or scapular pain, if there is evidence of neurological symptoms, fever, chills, malaise, or other signs of infection, the work-up should consider the possibility of a spinal-esophageal fistula.

KEY WORDS

discitis, meningitis, spinal-esophageal fistula

1 | INTRODUCTION

A spinal-esophageal fistula (SEF) resulting from discitis is very uncommon. SEF can cause fatal complications or mediastinal infection, and antibiotics and surgical treatment may be necessary.

Discitis is an inflammatory disease caused by an infection of the intervertebral disk. It usually manifests as back pain, limited mobility, and fever. It occurs mainly as a postoperative complication or due to spread of infection from other sites.^{1,2} A SEF is a rare complication of surgery, chemotherapy, or radiation therapy.^{3,4} No SEF due to an intervertebral disk has been reported.

We report a 69-year-old woman with a history of Pott's disease and a T3-T4 compression fracture who presented to the emergency department (ED) with scapular pain and was diagnosed with an SEF due to discitis.

2 | CASE REPORT

A 69-year-old woman visited our ED with right scapular pain that had started about 16 hours earlier. At 5 years of age, she had Pott's disease. She had been diagnosed with hypertension 20 years earlier and was taking oral antihypertensive

medication. In 2016, she was diagnosed with asthma and has been using an inhaler once a day. The patient had been treated conservatively without surgery for 2 years at another hospital for a T3-T4 compression fracture caused by an accident 18 years earlier and returned to normal activities. She did not drink alcohol or smoke.

In the ED, the patient was alert. Her blood pressure was 184/96 mm Hg, pulse rate was 62/min, respiratory rate was 20/min, and body temperature was 37.7°C.

The patient complained of pain in the right scapula and at T1-T2, but no radiating pain was noted. There was no cough, coryza, sputum, or shortness of breath. Physical examination revealed no tenderness or skin lesions in the vicinity of the right scapula or T1-T2. Chest auscultation was normal, as was the neurological examination.

Laboratory tests showed an elevated leukocyte count (14 950/mm³; normal range: 4000-10 000/mm³), with 93.3% neutrophils (normal range: 42.2%-75.2%). The C-reactive protein was 1.77 mg/dL (normal range: 0-0.3 mg/dL). Cardiac enzymes were in the normal range. Arterial blood gas analysis showed pH 7.463 (normal range: pH 7.35-7.45), pCO₂ 38.4 mm Hg (3-45 mm Hg), pO₂ 57 mm Hg (83-108 mm Hg), HCO₃ 27.5 mmol/L (22-26 mmol/L), and O₂ saturation on room air 91% (95%-100%).

Chest and right scapula X-rays were normal. Because of persistent pain in the area of the right scapula and T1-T2, chest computed tomography (CT) was performed to rule out

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microfracture of the spine or lung lesion. This showed bilateral pleural effusions, right lower lung pneumonia, and old fractures in the thoracic spine. A third-generation cephalosporin was administered.

Eleven hours later, the left knee flexion had decreased to Grade IV. Spinal magnetic resonance imaging (MRI) was performed because an infectious lesion in the spine was suspected based on the persistent elevated temperature. This revealed C5-C6 disk protrusion, ossification of the posterior longitudinal ligament with the adjacent spinal canal pushed to the left, and a signal change at T1. After an orthopedics consultation, the patient was admitted to a general ward for observation.

On the second day of admission, chest CT showed esophagitis (Figures 1 and 2), an esophageal fistula, and a soft tissue lesion in the prevertebral space. An esophagogram showed an esophageal fistula with discitis and an esophageal diverticulum (Figure 3). A thoracic surgeon recommended an esophageal stent and observation rather than surgical treatment. On day 5, the patient underwent gastroscopy following hematemesis. Peripapillary vascular exposure with a gastric ulcer was seen, and this was clipped with a hemostatic clip. The SEF was confirmed at gastroscopy; a large exudate was observed with only the outer membrane of the esophagus remaining on the side of the aorta. The thoracic surgeon recommended esophageal bypass surgery, but she refused surgery and was treated with a third-generation cephalosporin.

On day 6, her consciousness decreased, and orotracheal intubation was performed in the intensive care unit. Spontaneous blinking was observed, but her pupils were fixed at 5 mm/3 mm, and she did not respond to painful stimuli. The motor grade of all extremities was reduced to Grade I, and myoclonus in both arms lasting about 30 seconds was seen. Brain CT performed to rule out a cerebral hemorrhage was normal. The patient's temperature remained above 38°C and intermittent myoclonus of the left arm was also observed. The cerebrospinal fluid

was examined under suspicion of encephalitis. The opening pressure was increased at 240 mm Hg with 60 leukocytes/ μ L and 95% neutrophils. Bacteria in the esophagus had caused encephalitis after invading through the spaces between the intervertebral disks. Third-generation cephalosporin and nitroimidazole were administered. Generalized brain dysfunction was observed on electroencephalography. Subsequently, her consciousness has not improved and she is still on a ventilator, although respiratory rehabilitation is in progress.



FIGURE 2 Sagittal chest computed tomography (CT) shows an inflammatory or infectious mass lesion associated with discitis (T2-T3)



FIGURE 1 Axial chest computed tomography (CT) suggests esophagitis and an esophageal fistula draining into the inflammatory mass



FIGURE 3 Esophagography shows discitis with a fistula tract in the esophagus and secondary diverticulitis

3 | DISCUSSION

There are several types of esophageal fistula, of which tracheoesophageal fistula (TEF) is the most common. Typically, TEF is a congenital anomaly seen in children, with a frequency of 1 per 2500 live-births. The diagnosis is usually made in the early postnatal period, and the treatment is surgical.⁵ Adult TEF can be divided into non-malignant and malignant etiologies; the most common non-malignant etiology is cuff-related tracheal injury following intubation.⁶ TEF due to malignancy has been reported in 5%-15% of esophageal malignancies and less than 1% of bronchogenic carcinoma patients.⁷

Another form of esophageal fistula is atrio-esophageal fistula (AEF), which occurs in 0.015%-0.04% of patients undergoing radiofrequency ablation for arrhythmias, such as atrial fibrillation (AF).⁸ Esophageal injury is induced by atrial ablation because the posterior left atrial wall and esophagus are adjacent to each other. Although it is extremely rare, AEF is fatal and accounts for 16% of the mortality after AF ablation.^{9,10}

In addition, aorto-esophageal fistulas caused by a thoracic aortic aneurysm or esophageal malignancy,^{11,12} and carotid-esophageal fistulas resulting from a foreign body and Behçet's disease have been reported.^{13,14}

An SEF, which occurred in this case, is a common complication associated with anterior cervical surgery.¹⁵⁻¹⁸ Other causes include cervical spine trauma, radiotherapy for glottic carcinoma, and esophagectomy with gastric pull-up of esophageal cancer.^{3,4,19} Most cases of SEF are iatrogenic, following surgery or trauma. Spontaneous fistula due to infection of an intervertebral disk has not been reported. An SEF can lead to a central nervous system (CNS) infection, such as meningitis, as seen in this case, which is an important determinant of patient prognosis.⁴

The clinical manifestations of SEF vary according to its cause and location and include neck or back pain, neurologic defects, and surgical-site swelling. Our patient visited the ED complaining of right scapular pain; the lesion progressed to the spinal cord and affected the sensation in the lower limbs.²⁰ The prognosis and mortality of SEF have not been reported. In similar cases, esophageal perforation had a mortality rate of 10%-40%.²¹

Discitis, the antecedent of SEF in this case, is an inflammatory disease caused by infection of the intervertebral disk that manifests mainly as back pain and limited mobility.² Infection can result from direct propagation by hematogenous spread, trauma, or surgery. Transmission of infection may lead to complications such as a subdural abscess, dural abscess, or meningitis. The diagnosis is made based on blood tests, CT, MRI, and culture at biopsy. Antibiotic therapy is the main treatment modality, but surgical treatment is

needed if the infection requires drainage or there are neurological symptoms, recurrent infections, or failure of medical treatment.^{1,2}

4 | CONCLUSION

Our patient had scapular pain and fever when she arrived in the ED. Chest CT and spinal MRI were obtained on suspicion of pneumonia or other traumatic lesions. After admission, discitis and SEF were diagnosed after a formal reading of the images by radiologists.

There are various causes of back and scapular pain other than musculoskeletal origin. Discitis and SEF are rare. SEF can cause fatal CNS complications or mediastinal infection, and antibiotics and surgical treatment may be necessary. In patients with back or scapular pain, if there is evidence of neurological symptoms, fever, chills, malaise, or other signs of infection, the work-up should consider the possibility of SEF.

CONFLICT OF INTEREST

The authors declare that they have no competing interests.

AUTHOR CONTRIBUTIONS

J H Lee, E Kim, and Y H Choi: drafted the case presentations and edited the manuscript. All authors read and approved the final manuscript.

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