



Missed diagnosis of oesophageal perforation in ankylosing spondylitis cervical fracture: Two case reports and literature review

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Abstract

Oesophageal perforation after blunt injury cervical fracture in patients with ankylosing spondylitis (AS) is rarely reported. The early diagnosis of oesophageal perforation is extremely important. We present two cases of patients with AS who sustained cervical fracture dislocation and spinal cord injury. The ossified sharp fragments caused oesophageal perforation, and the delayed diagnoses had serious consequences. Oesophageal perforation should be suspected in patients with AS and cervical fracture if bone fragments are pressing against the oesophagus and a gas shadow is visible around the fracture site on computed tomography imaging.

Keywords

Oesophageal perforation, computerized tomography, cervical fractures, ankylosing spondylitis

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Introduction

The early diagnosis of oesophageal perforation after blunt injury is extremely important, but diagnosis is complicated by rarity and lack of specific symptoms.¹ We present two cases of patients with ankylosing spondylitis (AS) who sustained

cervical fracture dislocation and resulting cervical oesophageal perforation due to sharp, dislocated bone fragments. Concern over immediate, urgent symptoms caused the early signs of oesophageal perforation to be overlooked, with serious consequences.

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Case reports

Case I

A 43-year-old man presented at the Second Affiliated Hospital, School of Medicine, Zhejiang University, Hangzhou, China, on 28 September 2014, following a motor vehicle accident. Computed tomography (CT) imaging of the cervical spine revealed AS and fracture–dislocation at C6–C7 (Figure 1A). Magnetic resonance imaging (MRI) showed wide-ranging oedema and contusion of the spinal cord (Figure 1B).

On admission, physical examination revealed a midrange swelling of the neck

and tenderness of the left shoulder. X-radiography indicated a C6–C7 fracture–dislocation, with an appreciable perivertebral gas shadow (Figure 1C), but this was overlooked by medical staff. The patient's temperature increased to $>38.5^{\circ}\text{C}$, and sudden serious dyspnoea occurred 3 days after admission to the intensive care unit (ICU). The patient was intubated via oral endotracheal catheter and was successfully resuscitated. Repeat cervical spine CT imaging indicated perivertebral gas accumulation around the fracture site (Figure 1D). Oesophageal perforation and mediastinitis were strongly suspected.

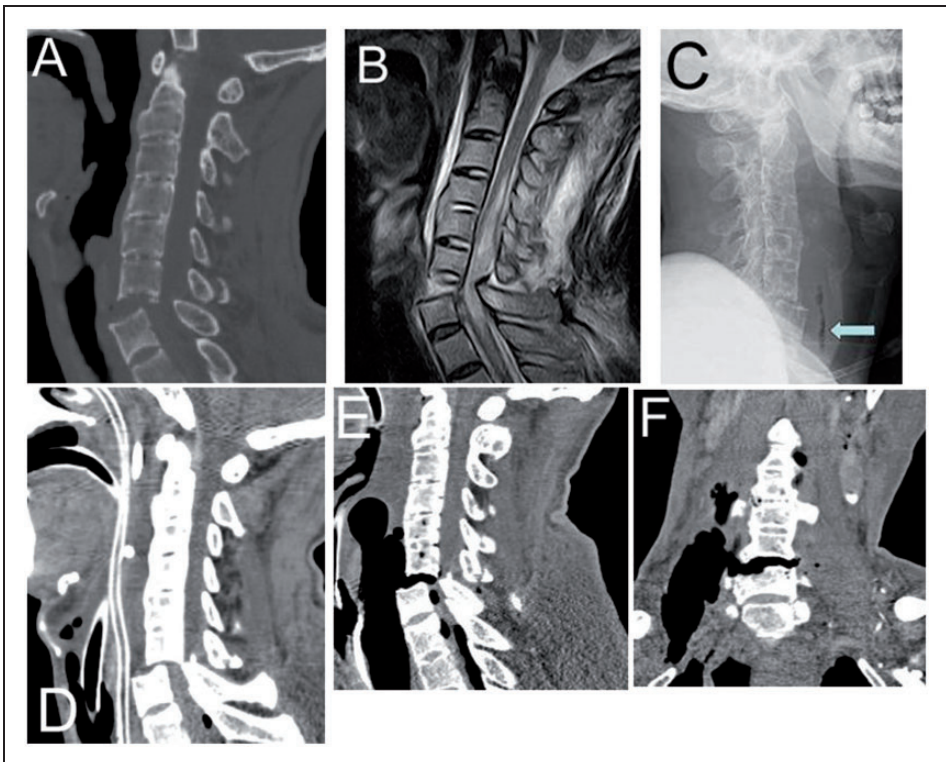


Figure 1. (A) Computed tomography (CT) image of the cervical spine of a 43-year-old man following a motor vehicle accident, revealing C6–C7 fracture–dislocation and ankylosing spondylitis (AS). (B) Magnetic resonance image indicating wide ranging oedema and contusion of the spinal cord. (C) Cervical X-radiography indicating C6–C7 fracture–dislocation, with an appreciable perivertebral gas shadow (arrow). (D) CT image indicating the accumulation of perivertebral gas, causing dyspnoea. (E) and (F) CT images showing massive perivertebral gas accumulation, leading to mediastinitis.

The patient was immediately treated with fluid infusion, intravenous broad-spectrum antibiotic therapy and supportive therapy, and was nil-by-mouth. His temperature gradually rose to $>39^{\circ}\text{C}$ and the neck swelling became increasingly apparent. CT imaging indicated a massive perivertebral gas accumulation at the cervical and thoracic spine (Figures 1E and 1F). White blood cell count was $25 \times 10^9/\text{l}$ with 96.2% neutrophils, and C-reactive protein (CRP) was 183 mg/l, indicating infection. After consultation with thoracic surgeons, tracheal incision and cervical drainage was carried out with extensive debridement, releasing large quantities of gas. Oesophagoscopy revealed a 17 cm-deep fistulous opening below the patient's incisor. The patient ultimately died due to complications resulting from the infection.

Case 2

A 77-year-old man presented at the Second Affiliated Hospital, School of Medicine, Zhejiang University, Hangzhou, China, on 12 December 2014 with a cervical fracture resulting from a fall. Before arrival, the local

hospital had placed a nasogastric tube to ensure gastrointestinal decompression. On admission, CT imaging revealed AS and fracture–dislocation at the C7 level. Bone fragments were pressing against the oesophagus, but there was no obvious gas shadow around the fracture site (Figure 2A). MRI indicated soft tissue swelling around the fracture (Figure 2B).

At 2 days after admission, the patient's temperature rose to $>38.8^{\circ}\text{C}$, his white blood cell count was $19.6 \times 10^9/\text{l}$ and his CRP was 82.01 mg/l. Chest CT imaging revealed compression of the soft tissue between the C7 fragments and the oesophagus, and a small gas shadow around the fracture site (Figure 2C). These findings strongly suggested oesophageal perforation. Fluid infusion, intravenous broad-spectrum antibiotic therapy and supportive therapy were given immediately. Oesophagoscopy revealed a 19 cm-deep opening below the incisor. The patient recovered following surgery and long-term anti-infective therapy.

Both patients provided written informed consent for publication. The School of Medicine, Hangzhou, China, does not

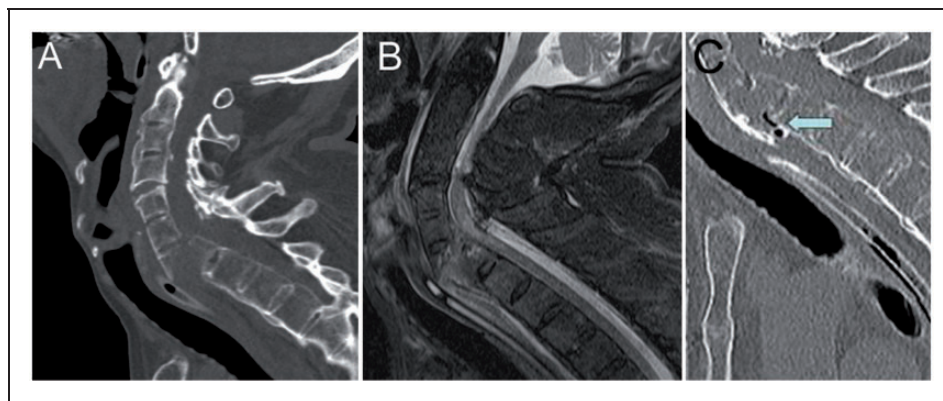


Figure 2. (A) Computed tomography (CT) image of a 77-year-old man following a fall, revealing fracture–dislocation at C7. Sharp bone fragments are pressed against the oesophagus. No gas shadow is visible. (B) Magnetic resonance image (MRI) revealing swelling of the soft tissue around C7, and near-disappearance of the space between the bone fragments and the oesophagus. (C) CT image revealing a small gas shadow around the fracture site (arrow).

require ethical approval for reporting individual cases.

Discussion

The early diagnosis of oesophageal perforation is very important in blunt injury, as primary closure within 24 h results in favourable outcome (92% survival). Delayed diagnosis and treatment is associated with significantly higher mortality.^{2,3} Few cases of oesophageal perforation in patients with cervical fracture have been reported^{4,5} and no cases in individuals with AS. AS increases the risk that ossified sharp bone fragments will perforate the oesophagus.⁶

Initial evaluation for oesophageal perforation can be performed by X-radiography, which has been shown to be suggestive in 86–88% of patients.^{7,8} Lateral cervical X-radiography can identify prevertebral gas earlier than chest radiography.⁹ However, X-radiography may fail to identify small-scale oesophageal leakage, especially in patients with cervical fracture (where the surgeon tends to focus on the dislocation and spinal injury). Evidence suggests that CT imaging enables better diagnosis and management than X-radiography,^{10,11} and should be performed before contrast-enhanced oesophagography.^{12,13} Extraluminal gas is the most common CT finding associated with oesophageal perforation, occurring in 92% of cases.^{14,15} In the early stage of oesophageal perforation there may be a negligible gas shadow around the fracture site, which can be easily overlooked. If CT imaging reveals pressing of bone fragments against the oesophagus, especially in patients with AS, close attention should be paid to the possibility of erosion and perforation of the oesophageal mucosa.

Contrast oesophagography is the standard diagnostic tool for oesophageal perforation¹⁶ and barium contrast oesophagography should be used if no perforation is detected.^{17,18} This technique

cannot be easily used in patients with cervical spine injury, however. Flexible oesophagoscopy has a sensitivity of 95.8–100% and a specificity of 92.4–100% in the evaluation of penetrating oesophageal injury and can provide a direct visualization of the perforation; it is thought to be a useful diagnostic tool with a good safety profile in the early evaluation of penetrating injuries.^{19–21} In our two cases, oesophageal perforation was confirmed via flexible oesophagoscopy, but no images were taken due to the emergent situation.

Ankylosing spondylitis is a chronic inflammatory rheumatic disease that can cause extensive remodelling of the spinal axis, leading to rigidity and fragility of the spinal column.²² Spinal fractures in patients with AS are usually sharper and more unstable than in individuals without AS.^{23–25} The ossified sharp fragments in AS may cause oesophageal injury during the process of fracture dislocation. In the cases reported here, mediastinitis and haematosepsis were a direct result of oesophageal perforation. Close attention should therefore be paid to the possibility of oesophageal injury in patients with AS.

In conclusion, very small perivertebral gas shadows on CT imaging should be regarded as a possible early diagnostic sign for oesophageal perforation in cervical fracture, especially for any patient with AS. Pressing of sharp bone fragments against the oesophagus may be a precursor to perforation. Delayed diagnosis and treatment can have serious consequences.

Declaration of conflicting interest

The authors declare that there are no conflicts of interest.

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