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CASE REPORT

Esophageal Perforation in a Cervical Fracture Patient With Progressed Ankylosing Spondylitis

Case Report and Review of the Literature

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Study Design. Case report and review of relevant literature.
Objective. To report a rare complication of esophageal perforation in cervical fracture patient with progressive ankylosing spondylitis (AS) and review of relevant literature.
Summary of Background Data. Esophageal perforation in cervical fracture is extremely rare. This complication has never been reported in AS patients. Key points in the early diagnosis and management of this severe complication along with the cervical fracture were presented in detail in this case report.
Methods. A 61-year-old male with AS, who sustained C6 fracture and dislocation after a vehicle accident. Initial treatment strategy was combined anterior-posterior fixation and fusion. Although esophageal perforation in the dorsal wall of esophagus was detected in anterior surgery. After thorough debridement, the esophageal lesion was closed by resorbable interrupted sutures. Surgical incision was washed by saline and hydrogen peroxide without primary closure. Cervical fracture was treated by long level posterior fixation alone. After surgery, the anterior surgical incision was irrigated with normal saline twice a day and closed 3 weeks later.
Results. Esophageal perforation was healed completely and neurologic results were improved significantly after surgery. Union of the fracture and segmental stability of cervical spine was confirmed radiologically at 1-year follow up.
Conclusion. Esophageal perforation in cervical fracture patient with progressed AS is an extremely rare and potentially life-threatening complication. Surgical debridement and drainage with intensive care are essential for good outcome.
Key words: ankylosing spondylitis, cervical fracture, complication, esophageal perforation.

Level of Evidence: 5
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CASE REPORT

A 61-year-old man suffering from AS for about 25 years was admitted to our emergency room 12 hours after a traffic accident. His chief complaints were neck pain, left upper limb pain, and numbness. Muscle strength of the upper extremities was 3/5 (left) and 4/5 (right). Hypertonia and hyperreflexia were observed in the left upper extremities. The Japanese Orthopedic Association (JOA) score for evaluating the spinal cord function was 11 points. Preoperative radiographic examination revealed typical ''bamboo spine'' with C6 fracture and dislocation. CT scan demonstrated transverse fracture through C6 vertebral body with laminar and spinous process fracture. MR imaging showed prevertebral edema, rupture of anterior longitudinal ligament, and compression of spinal cord (Figure 1A–F).

Treatment was commenced with skull traction immediately after admission. Three days later, the patient developed continuous fever (about 39°C) and throat pain that intensified when swallowing. Laboratory tests revealed a leucocyte count of 20.8×10^9/L, with 93.6% neutrophils. After treatment with moxifloxacin, the body temperature quickly returned to normal.

Radiography revealed no reduction of the fracture after traction. The initial treatment strategy was combined anterior-posterior fixation and fusion. During the anterior approach, yellowish viscous pus was observed in the prevertebral space. An esophageal perforation measuring about 0.5 cm in length, caused by the sharp fracture fragment, was detected on the dorsal wall of the esophagus. A nasogastric tube was inserted, and after thorough debridement, the esophageal lesion was closed by resorbable interrupted sutures. The surgical incision was washed using saline and hydrogen peroxide without primary closure. Subsequently, posterior surgery was performed and fracture reduction was accomplished by fixation from C4 to T2 (Figure 2A, B).

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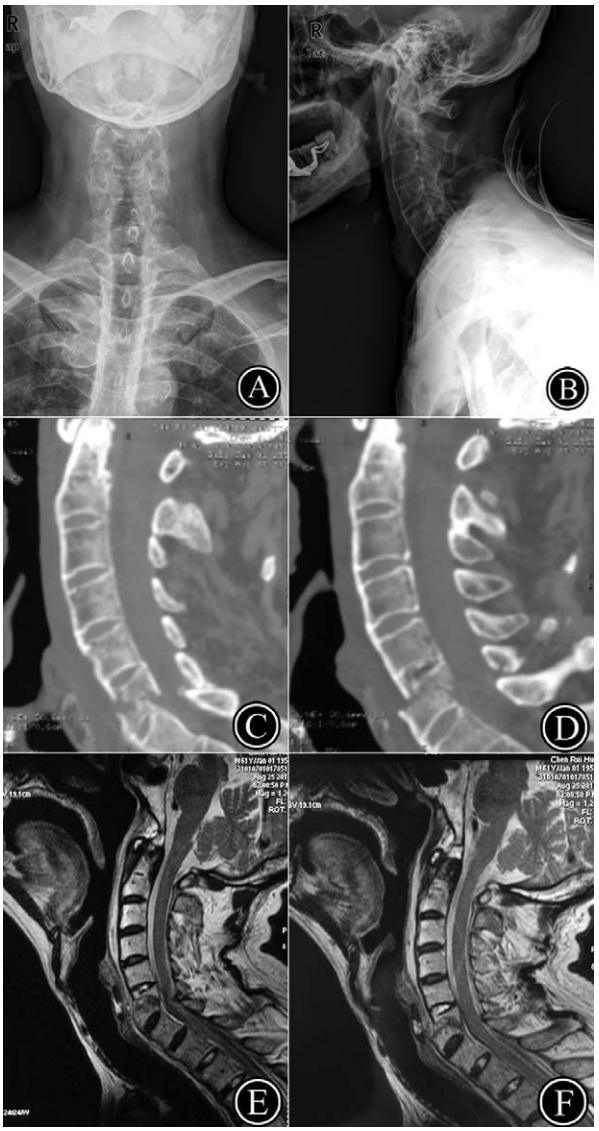

Figure 1. Preoperative imaging assessment. **A** and **B**, Preoperative x-ray examination suggested a typical ''bamboo spine'' with C6 fracture and dislocation. **C** and **D**, CT sagittal reconstruction confirmed the transverse fracture through C6 vertebral body with laminar and spinous process fracture. **E** and **F**, MRI demonstrated prevertebral edema, rupture of anterior longitudinal ligament, and compression of spinal cord.

The patient was treated with total parenteral nutrition (TPN) and broad-spectrum antibiotics after surgery. The surgical incision was irrigated with normal saline twice a day. The incision was closed 3 weeks later, after which it healed completely. Flexible endoscopy was performed 4 weeks after surgery, with no evidence of abnormality. The patient was immobilized with a hard cervical collar for 2 months. His neurological symptoms improved significantly after surgery, and his JOA score increased from 11 points preoperatively to 15 points at the final follow-up. Postoperative radiographs revealed satisfactory reduction and fixation of the fracture. Union of the fracture and segmental stability of the cervical spine was radiologically confirmed at the one-year follow-up (Figure 3A–D).

DISCUSSION

Esophageal perforation is an extremely rare complication in patients with cervical fracture. We performed a review of the literature and the results were summarized as follows. Only 10 cases with detailed clinical information reported in 9 case reports from 1960 to 2013 are available (Table 1).[3–11] The mean age of the patients in the 10 cases was 51.2 ± 20.7 years (range, 19–79 years). Vehicle accidents and falls were the most common causes for injury. The cervical injury level was mostly in C4 to C6. However, none of these reported cases were complicated with AS.

In this report, we presented a case of esophageal perforation in the cervical fracture of a patient with progressed AS, and described the diagnosis and management of this unique and severe complication in detail. Owing to the atypical signs and symptoms accompanied by the injury, prompt diagnosis of this complication can be difficult. Clinical signs that could lead to the diagnosis include increasing anterior cervical pain and tenderness, swelling and edema of the cervical area, dyspnea, and dysphagia.[7] Patients with prevertebral edema or emphysema in CT or MR imaging should be highly suspected. Barium esophagogram and esophageal endoscopy can help confirm the diagnosis.

To treat this complication, most published studies emphasized the importance of surgical debridement and drainage with intensive care. Continuous irrigation is an alternative

Figure 2. A, The surgical incision without primary closure was irrigated with normal saline twice a day after surgery. **B**, The surgical incision was closed 3 weeks later and healed.

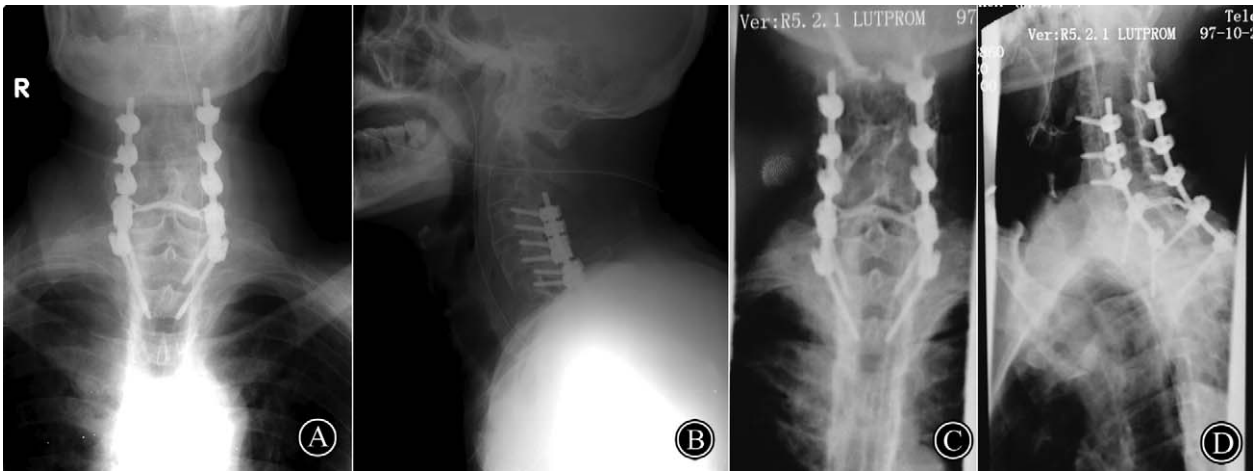

Figure 3. A and **B**, Postoperative anteroposterior and lateral radiographs revealed satisfactory reduction and fixation of the fracture. **C** and **D**, Union of the fracture and segmental stability of cervical spine was confirmed radiologically at 1-year follow up.

TABLE 1. Summary of Cases of Esophageal Injury in Cervical Fracture Reported in Literature

Author	Year	Journal	Age/Sex	Cause of Injury	Injury Level of Cervical Spine	Description of the Esophageal Injury	Management of the Esophageal Injury	Results
Dabija *et al*[3]	2014	Rev Med Chir Soc Med Nat Iasi	65/F	Fall	C6–7	A small bone fragment inserted in the esophagus on his left side, with a small defect at the adventitia level	Drainage, esophageal suture and gastrostomy	Recovery with a permanent gastrostoma
Delos *et al*[4]	2013	Int J Surg Case Rep.	51/F	Fall	C4–C6	Contained esophageal perforation at the C3–C4 level	Broad-spectum empiric antibiotics, total parenteral nutrition, oral hygiene	Complete recovery
Makoyo PZ[5]	1979	J Natl Med Assoc	22/M	Vehicle accident	C4–5	Esophageal perforation in the posterior wall of esophagus	Debridement, irrigation and gastrostomy	Complete recovery
Morrison A[6]	1960	J Bone Joint Surg Br	73/M	Fall	C4–7	A two centimeters long tear in the back of the oesophagus to the left of the mid-line	Drainage and chemotherapy	Dead
Nerot *et al*[7]	2002	J Spinal Disord Tech	55/M	Vehicle accident	C2,C5–6	A tear of the esophagus on the right side with copious outflow of pus	Debridement and drainage	Complete recovery
Reddin *et al*[8]	1987	J Trauma	79/F	Fall	C4–6	1.5-cm vertical laceration of the posterior midline of the cervical esophagus	Repaired in two layers with interrupted 3–0 Vicryl sutures, irrigation	Dead
Stringer*et al*[9]	1980	J Neurosurg	53/F	Fall	C5–6	Anterior and posterior tears of the esophagus 1 to 2 cm above the sternal notch	Separate suture closure and drainage	Complete recovery
Tomaszek *et al*[10]	1984	Neurosurgery	19/F	Dive	C5–6	A small cervical esophageal perforation and mediastinitis	Gastrostomy, jejunostomy and drainage	Complete recovery

TABLE 1 (*Continued*)

Author	Year	Journal	Age/Sex	Cause of Injury	Injury Level of Cervical Spine	Description of the Esophageal Injury	Management of the Esophageal Injury	Results
			32/F	Vehicle accident	C6–7	A left paratracheal abscess at the level of the cricoid cartilage extended posteriorly toward to esophagus and cervical spine	Drainage and chemotherapy	Complete recovery
Agha FP[11]	1982	Br J Radiol	63/F	Vehicle accident	C7	A wide postero-lateral perforation of the upper oesophagus	No details	Dead

management method for wounds with discharging pus. In early diagnosis, repair of the esophageal laceration is possible. Treatment with antibiotics after surgery is essential for the prevention of incision and mediastinal infection. As shown in Table 1, three of the patients in the 10 reviewed cases died, suggesting the high mortality rate of this complication. Diagnosis of esophageal perforation was not made before surgery in the current case and the treatment strategy was modified accordingly during operation. The outcome was satisfactory, which was attributed to the reasonable treatment and postoperative intensive care.

➢ Key Points

- ❑ Esophageal perforation complicating in cervical fracture patient with progressed ankylosing spondylitis is extremely rare and potentially life threatening.
- ❑ To avoid delayed diagnosis, great importance should be attached to clinical signs that could lead to esophageal perforation in cervical fracture patients.
- ❑ Surgical debridement and drainage with intensive care are essential for good outcome of this severe complication.

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