Case Report

Successful surgical management of traumatic cervical spine injury in a patient with ankylosing spondylitis: a case report

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Abstract: Objective: The purpose of this study is to present an ankylosing spondylitis (AS) patient with fracture-dislocation of C7/T1, who got reduction and fixation successfully by surgical treatment. *Methods*: A 36-year-old Chinese male with 22-years history of AS was admitted to our Emergency Department (ED) complaining about sensory and motor disturbance for 12 hours due to the fall at home. AS and fracture-dislocation of C7/T1 were diagnosed by personal history, neurological examination and X-ray, CT and MRI. *Results*: Posterior cervical instrumentation and fusion from C5 to T3 (lateral mass screws of C5, C6 and pedicle screws of C7, T2, T3). Meanwhile, satellite rod was placed in C7-T2 to disperse stress. The lamina and ossified ligamentum flavum of both C7 and T1 were removed for decompression; no extra-dural hematoma was observed. Given the patient's longstanding history of AS, the fracture-dislocation site was highly unstable, combined anterior and posterior fixation was performed. Then, the patient underwent open reduction, internal fixation of C7-T1 fracture and anterior cervical plating with iliac crest autogenous bone grafting. The patient did well postoperatively, his neurological state recovered to Frankel D, with muscle strength of 3 of 5 in the upper extremities and 3+ of 5 in the lower extremities. Although neurological improvement was minor, his motivation increased, and pain from his neck disappeared. *Conclusion:* Surgical treatment of AS patients with subaxial unstable fracture-dislocation can be performed successfully.

Keywords: Ankylosing spondylitis, fracture-dislocation, cervical spine injury, surgery, satellite rods

Introduction

Ankylosing spondylitis (AS) is a rare (0.008%) inflammatory arthritis predominantly involving the axial skeleton [1, 2], which predisposes patients with balance and coordination problem, and ultimately falls and spinal fractures [3]. In addition, AS is accompanied with osteoporosis, ankylosis and syndesmophyte formation further increasing the risk of spinal fracture or dislocation after minor trauma [4].

The incidence of spinal fracture among patients with AS is reported to range from 0.4% to 12% [5, 6]. Patients with AS have a risk of spinal cord injury that is 11.4 times greater than general population [7]. Neurological deficits from spinal cord injury are more frequent in spinal fracture patients with AS than patients without AS, occurring in up to 92% of patients with AS and spinal fractures [5], with 65% of these

patients never regaining full neurologic recovery [5].

Treatment of patients with AS and spinal fractures is controversial, 33%-46% of these patients receive conservative treatment so far [8, 9]. In the presence of instability, surgery is performed to achieve stabilization mainly. Patients with AS and spinal fractures may benefit from rigid operative stabilization to prevent secondary displacement and spinal cord injury, so much better outcome can be obtained in patients treated surgically [10]. Operative stabilization usually consists of anterior, posterior, or combined antero-posterior fixation, accompanied by decompression.

The goal of this case report was to ascertain surgical management for AS patient with sub-axial cervical spine fracture-dislocation, which is rarely reported [11, 12].

Case report

An appropriate written informed consent was obtained from the patient reported in this study.

A 36-year-old Chinese married non-smoking male with 22-years of AS presented to the Emergency Department (ED) after falling backward and hitting his head at home. He complained about sensory and motor disturbance for 12 hours due to the fall, and he provided that he didn't treated his AS systematically, lower back pain along with stiffening and deformity of the spine and hip progressed gradually. Previously, he has been suffering from bilateral femoral head necrosis for 18 years, but did not receive proper treatment. He had significant joint stiffness in his arms and legs that caused difficulty in walking without assistance in the past 8 years. Patient denies smoking, illicit drug and alcohol abuse. None of his family members had history of AS or other rheumatologic diseases. On admission, he was conscious, physical examination revealed a midrange swelling of the neck and tenderness of the left shoulder, with muscle strength of 2 of 5 in the upper extremities and 3 of 5 in the lower extremities, according to the Medical Research Council muscle grading scale. No Achilles and no patella reflexes could be obtained, Hoffman sign and Babinskin sign was positive bilaterally. In the upper extremities, the triceps reflex but not biceps reflex was present and symmetric bilaterally. Sensitivity was absent below the level of the nipples. Laboratory examination showed fasting plasma glucose (FPG): 8.23 mml/L (3.9-6.1), White blood cell (WBC): 5.2×10⁹/L (3.5-9.5), Human leukocyte antigen B27 (HLA B27) (-), D-dimer: 13.37 μg/ml (0-1.0). Computed tomography (CT) imaging of the cervical spine revealed characteristic "bamboo" spine of AS and fracture-anterior dislocation at C7-T1 level (Figure 1B-D). Magnetic resonance imaging (MRI) showed wide-ranging oedema and contusion of the spinal cord (Figure 1E. **1**F).

He was subsequently taken to the operating room for posterior cervical instrumentation and fusion from C5 to T3 (lateral mass screws of C5, C6 and pedicle screws of C7, T2, T3). Meanwhile, satellite rod was placed in C7-T2 to disperse stress. The lamina and ossified ligament flavum of both C7 and T1 were removed

for decompression; no extra-dural hematoma was observed. Given the patient's longstanding history of AS, the fracture-dislocation site was highly unstable, so combined anterior and posterior fixation was performed. Then, the patient underwent open reduction, internal fixation of C7-T1 fracture and anterior cervical plating with iliac crest autogenous bone grafting. The patient did well postoperatively, his neurological state recovered to Frankel D, with muscle strength of 3 of 5 in the upper extremities and 3+ of 5 in the lower extremities. Although neurological improvement was minor, his motivation increased, and pain from his neck disappeared. Postoperative radiographs revealed satisfactory reduction and combined anteriorposterior 360 degrees cervical-thoracic stabilization of the fracture dislocation.

Discussion

AS is a rare condition and spinal fracture is a relatively rare complication [13], but patients with AS at a high risk for spinal fracture. Spinal fractures in AS patients usually have 3-column injuries and prone to displacement, so the risk of spinal cord injury is thought to be higher in AS patients with spinal fracture than general spinal fracture population [7].

Due to changed biomechanical spinal properties, the forces needed to fracture an ankylosed spine are smaller than normal spine. Therefore, even a trivial trauma (i.e., falls from standing/sitting position) in the ankylosed spine can result in an unstable fracture involving three columns of vertebral [14]. Sometimes, fractures can occur without any recognizable trauma [15]. Hyperextension is the most frequently observed mechanism of injury [15]. The lower cervical spine is the most commonly injured area because of increased mobility, small vertebral bodies, oblique articular facets, and the mobility of the skull on it [16]. The fracture-dislocation of the patient in our case occurred C7-T1 that are characteristically transverse and was called "carrot stick fractures" vividly.

As fractures of the ankylosed spine usually involve all three vertebral columns and ossified ligaments, both the anterior and posterior elements of the vertebra are often involved during fracture, and surrounding tissues are also damaged (**Figure 1B**). This type of cervical fracture

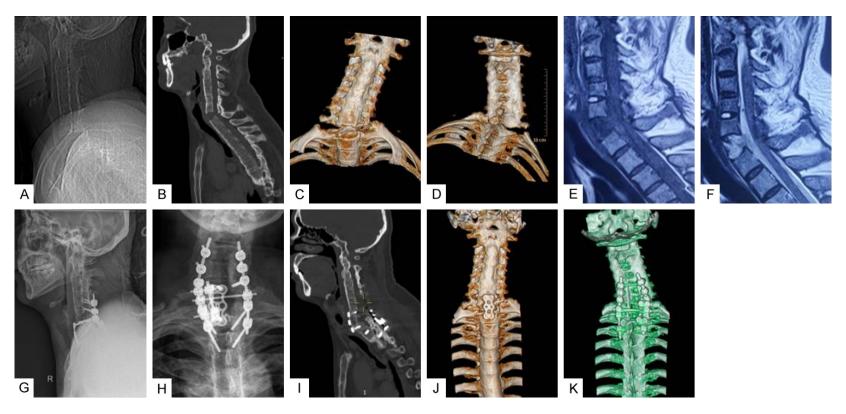


Figure 1. Fracture-dislocation of C7 to T1 with disruption of anterior and posterior complex in the X-ray (A), CT (B), 3D-CT (C, D) and MRI (E, F), which indicates tearing of the ALL and erosion of the vertebral body at the insertion of this ligament. The radiographic images in sagittal (G) and posteroanterior (H) views demonstrate the reduction and screws fixation; Sagittal CT (I) showed reduction of C7-T1 level fracture dislocation; 3D surface-rendered CT reconstruction (J, K) showed the postoperative stabilization in 360 degrees.

tends to be highly unstable and may be associated with neurological complications [17]. Many reports in the literature suggest the presence of neurological complications after vertebral fractures in patients with AS, including spinal cord and nerve root lesions, intramedullary edema, spinal epidural hematomas, and spinal stenosis, resulting in a variable degree of sensory and/or motor deficits [9]. Alaranta et al. [7] determined that the incidence of spinal cord injuries in patients with AS was 11.4 times that in the general population. Therefore, patients with traumatic fractures of the ankylosed spine are not only susceptible to initial neurological deficits, but also to secondary neurological deterioration due to highly unstable fracture configuration [9].

Because the incidence of AS and spinal fractures is very low, management of spinal fractures in AS patients is controversial [16], there is no high level evidence to guide medical and surgical management. Most subaxial spinal injuries involve the lower cervical spine or cervical-thoracic junction [10]. Although lower cervical spine is the most common site for fracture in AS population, management of subaxial spinal injuries in AS patients is still a challenge due to high rates of mortality and morbidity [18]. For asymptomatic patients or patients without neurologic symptoms but pain, it might be reasonable to pursue conservative management. For patients with neurologic symptomatic, surgery is essential, as one-third of these patients will experience disease progression if left without intervention [19]. Conservative management has long been the gold standard for the management of cervical spine fracture in AS patients [20]. It includes bed rest, traction and immobilization with a halo vest. However, conservative treatment has been associated with poor results such as bed rest usually lead to hypostatic pneumonia and decubitus, worsening of the regional deformity with loss of reduction, risk of nonunion [21], axial traction may result in serious medullary distractions [22] and risk of neurological aggravation [16]. Another argument against conservative management is the unstable nature of AS and spinal fractures, as most of the these fractures involve three columns [23]. Effectively, non-displaced or minimally displaced acute traumatic lesions may be treated by conservative methods [24], but the inherent instability of these

fractures, reflected by their high potential of acute displacement, may cause a catastrophic situation [25]. Therefore, surgical fixation with long segmental instrumentation and fusion is highly recommended [25].

Surgical stabilization usually includes anterior, posterior, or combined antero-posterior fixation, often accompanied by decompression with laminectomy, bone fusion (anterior, posterior, posterolateral), and various osteotomy for deformity correction [15, 26]. Definitive fracture fixation has many advantages over nonoperative approach, such as excellent deformity correction, immediate stability, direct and indirect decompression of the spinal canal, early mobilization, and prevention of secondary neurological complications [27]. Previously, some case series reported surgical treatment outcome for cervical fractures in AS patients. Early reports of operative management mostly relied on isolated posterior decompression and had very high mortality rates, the reason may due to isolated posterior decompression is not suitable technique for these unstable fractures [28]. With advanced technology, long segment posterior screws fixation with posterior bone graft have been described [29]. Although isolated anterior approach has been suggested, and Kouyoumdjian et al. [16] reported that anterior fixation alone could be sufficient if the hardware is long enough to avoid significant moment arms, but stability of isolated anterior fixation has been questioned. Failure with anterior or posterior stabilization alone suggested that combined anterior and posterior fixation would be the preference in the fixation of these highly unstable fractures, as it has resulted in no failure [11, 30]. Decompression may be performed if there is localized stenosis in the spinal canal, especially when the patient has neurologic deficit. Our case used the combined approach for fractures of the subaxial cervical spine include the posterior approach with lateral mass screws (C5, C6) and the pedicle screws (C7, T2, T3) and the anterior approach with the use of plates. In addition, we took full advantage of corrective osteotomies (the satellite rod of C7-T2) in patients with spondyloarthropathies in order to disperse stress and strengthen fixation [31].

However, complications specific to AS must be considered when opt for surgery. The disadvan-

tages of surgical intervention include postoperative complications, particularly respiratory, and increased mortality [32]. The posttraumatic epidural hematoma is most often seen in patients with AS [33]. Epidural hematoma often occurs due to bleeding from the epidural venous plexus and the fractured bone [34]. It's worth mentioning that delayed diagnosis of epidural hematoma may result in further deficit of neurological function. So imaging studies including MRI and CT should be reviewed carefully to rule out epidural hematoma [30]. The reduced bone density within the vertebral bodies typical of AS leads to a high rate of implant loosening of about 10-15% [35, 36]. Consequently, it is highly recommended to create multiple anchor points, by extending the instrumentation over at least three vertebral levels above and three below the fracture site [36]. To reach rigid internal fixation, we performed combined anterior-posterior 360 degrees cervicalthoracic stabilization.

In spite of more knowledge about early diagnosis and treatment of AS, little attention is being paid to the environmental hazards that pose a risk for patient outcome [37]. Efforts should be made to educate the patient and his or her caregivers about the risks of injury and to prevent similar severe injuries.

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Disclosure of conflict of interest

None.

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