

Unstable Cervical Fractures with Long Standing Ankylosing Spondylitis: Report of Six Cases

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Abstract

Purpose: Irrespective of the degree of trauma, patients with ankylosing spondylitis may suffer from cervical spinal fractures which are associated with high rates of morbidity and mortality. However, there has been no common consensus on how to manage such patients. It seems that the management depends largely on the physician's decision, some recommend nonsurgical treatment while others recommend surgical intervention. Our aim is to contribute to the literature regarding the decision-making in ankylosing spondylitis patients associated with cervical fractures by presenting six cases.

Methods: The clinical features and management of six patients were presented and discussed under the light of relevant literature. All six patients had history of ankylosing spondylitis for more than 15 years. Fractures were caused by minor trauma in five and major trauma in one patient. Three patients were managed conservatively, but one of them then underwent decompression surgery because of an unsuccessful reduction, while the other three had surgical stabilizations.

Results: Two patients died: one after surgery and one during nonsurgical management. Considering the cervical fractures of patients with ankylosing spondylitis, the decision-making must be carried out separately for each patient according to his or her clinical condition.

Conclusion: We advise surgery in patients with neurological deficit while nonsurgical management should be the first choice of treatment in cases with no neurological deficits. Furthermore, if nonsurgical treatment fails, irrespective of the neurological status, surgical intervention should also be considered.

Key words: ankylosing spondylitis, cervical fracture

Introduction

Ankylosing spondylitis (AS), first described in the 1890s, is a rheumatic condition with progressive ossification of the spinal ligaments and ankylosing of the facet joints, eventually leading to a totally stiff spine that acts as a single long bone. This disease is included in seronegative inflammatory spondyloarthropathies and is associated with osteoporosis as a result of chronic inflammation and disuse atrophy.⁽¹⁾ Patients suffering from AS are known to be more prone to spinal fractures and have two to three the times risk of morbidity and mortality than the normal population.^(2,3) Minor traumas may easily cause spine fractures in these patients. Sometimes history of trauma is never found. The cervical spinal column in patients with AS is also affected and the fractures most commonly occur in the lower region which can be missed in radiographs. Thus, the cervical fractures in patients with AS are unique with respect to mechanism(s) of injury, diagnosis, treatment and

prognosis. The correct management of cervical fractures in the presence of AS remains obscure.⁽³⁻⁶⁾ The aim of this paper is to contribute to the literature by presenting the clinical and radiological features, diagnosis and treatment of AS patients with cervical spine fractures.

Case Reports

Case 1: A 44-year-old man with AS history of 16 years was admitted to a hospital after a traffic accident. Severe nonradiating neck pain was the sole complaint of the patient. No neurological deficit was observed in routine examination (Frankel E). Beside typical bamboo spine appearance, C6-7 dislocation was detected on direct radiographs and he was referred to our center the next day. In his cervical magnetic resonance image (MRI), C6-7 disc fracture, anterior longitudinal ligament (ALL), posterior longitudinal ligament (PLL) and interspinous ligament rupture without

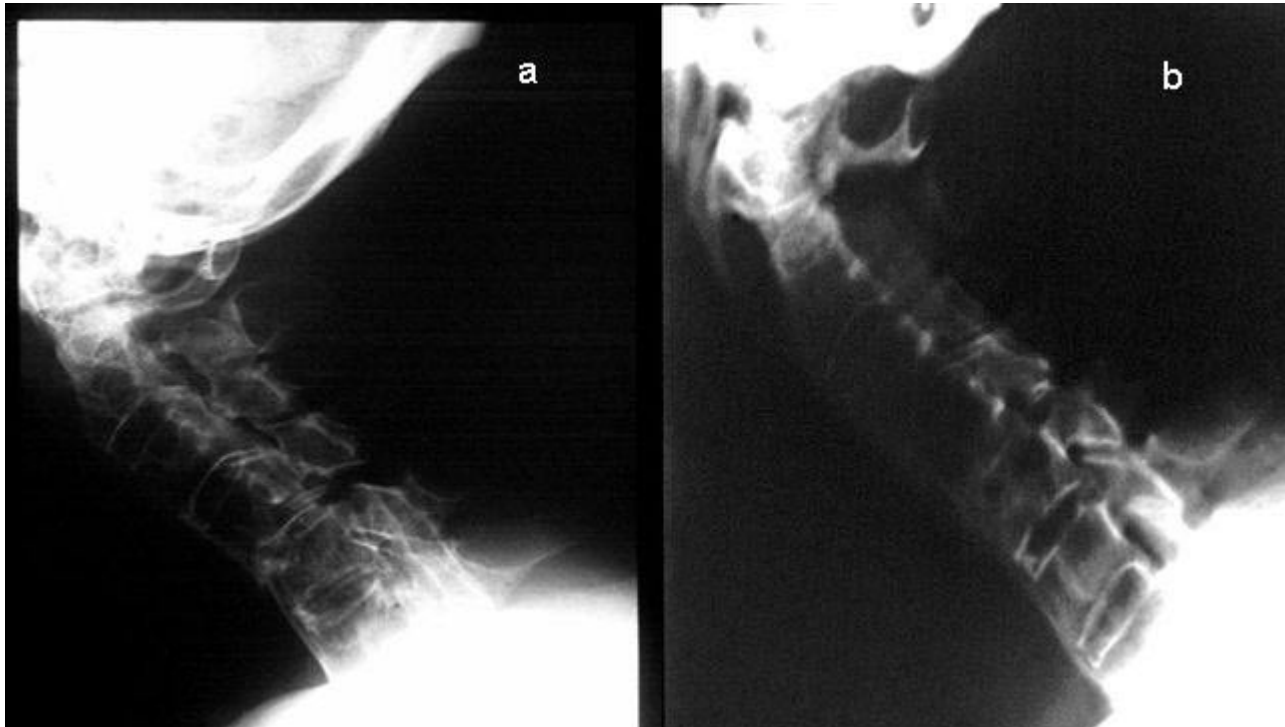


Figure 1. Lateral radiograph showing the dislocation at the level of C4-5 (a) after trauma. Following one year post-halo-vest treatment, normal alignment of the cervical spine was achieved (b).

any epidural hematoma was noted. We performed traction, gradually increasing up to 5 kg and achieved a complete reduction confirmed by serial cervical direct X-ray studies. Then, external immobilization was applied by halo-vest after the third day of trauma and the patient was mobilized. Twelve weeks after treatment, fusion was observed in cervical spine radiograph; halo-vest was removed, and a Philadelphia collar was recommended. In the first year of follow-up, he was totally pain free and adequate alignment was achieved.

Case 2: A 50-year-old male patient with AS history of 20 years hit a curbstone during a U-turn. He complained of severe nonradiating neck pain and his physical/neurological examination disclosed no abnormality (Frankel E). He was referred to our center with a diagnosis of C4-5 dislocation on the first post-traumatic day (**Figure 1a**). Because of a low degree kyphosis, traction was not applied; rather a halo-vest fixation in neutral position was applied immediately and the patient was mobilized. Twelve weeks after halo-vest treatment, fusion was achieved, the halo-vest was removed and a soft collar was recommended. In the first year follow-up, he was symptom-free and normal alignment was recorded radiologically (**Figure 1b**).

Case 3: A 50-year-old man with AS history of 20 years was admitted to our center after falling. He complained of severe neck pain and his neurological examination revealed a complete C4 lesion (Frankel A). C4-5 dislocation was observed in his radiological examination. Traction was applied

via Crutchfield skull calipers with increasing weight up to 8 kg with no reduction achieved, so he underwent surgical intervention. Bilateral facet removal and in situ fusion were performed. There was no epidural hematoma observed intraoperatively. The patient remained in skeletal reduction with 2 kg. On his second post-trauma day, he died of sudden cardiac arrest.

Case 4: A 52-year-old man with 25 years of AS was admitted three days after a fall. He complained of severe neck pain, and his neurological examination revealed minimal weakness at the right triceps muscle only (Frankel D). C5-6 dislocation was observed in his radiological examination. A C5-6 vertebrectomy, reconstruction with fibular allograft and anterior cervical plating between C4-7 was performed via anteromedial approach. The postoperative period was uneventful. He was discharged from the hospital without any additional neurological deficit.

Case 5: A 54-year-old man with 25 years of AS was admitted three months after falling. He reported that following the accident he was referred to a county hospital where he received a diagnosis of soft tissue damage and bed rest was advised. His neck pain persisted and he was advised physical therapy. During the second week of physical therapy he gradually developed a left leg weakness (Frankel D). An MRI of the cervical spine revealed a C6 corpus fracture and the patient was transferred to our clinic for further treatment. We performed a two-stage surgery. The first stage consisted

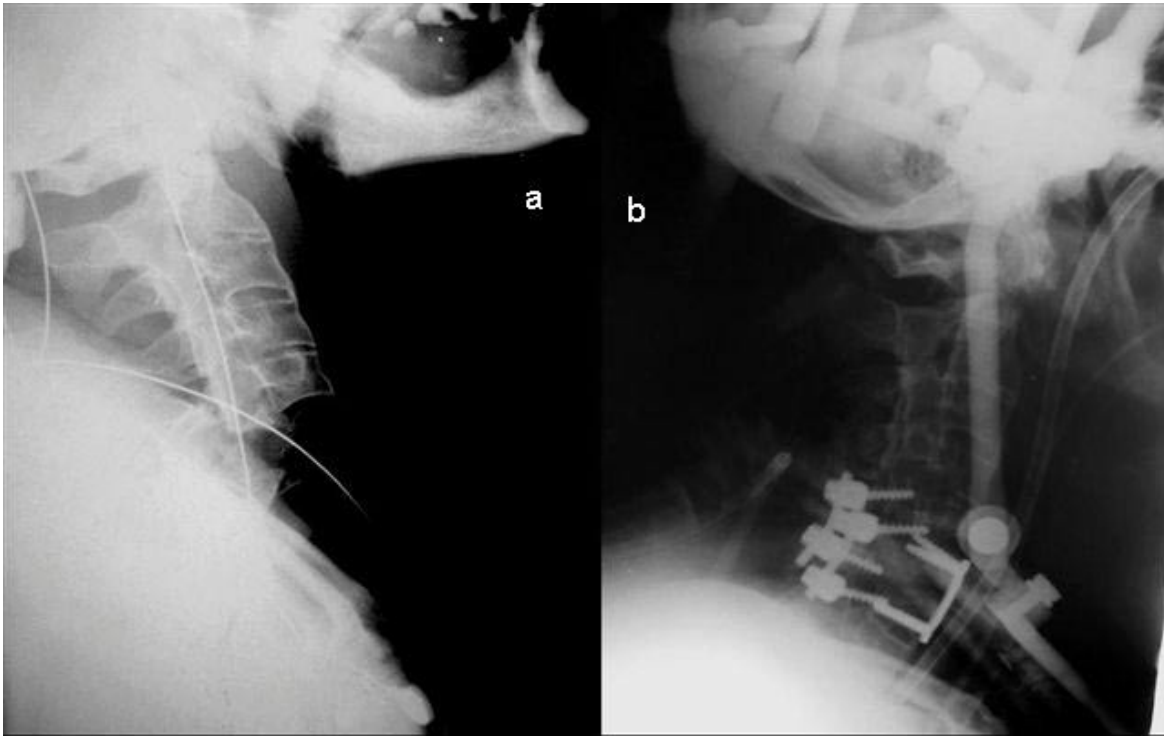


Figure 2. Lateral cervical radiograph demonstrating C5-6 dislocation (a). Note the degree of malalignment of the cervical spine. The patient had surgery in which both anterior and posterior fixation with instrumentation was performed in order to re-align the cervical spine (b). Unfortunately, the patient died due mainly to pulmonary infection and related septicemia.

of a C6 corpectomy reconstruction with an autologous iliac crest bone graft supported with an anterior plate fixation through anteromedial approach. The posterior stabilization was achieved using lateral mass fixation between C5-7. The patient was kept in a sponge collar and transferred to a rehabilitation facility on the fifth day postoperatively. The patient regained his preoperative activity level.

Case 6: A 60-year-old man with 30 years of AS was referred to us three days after a traffic accident. He originally was admitted to a county hospital and at that time was conscious and had paraparesis (Frankel C). His radiological examination revealed a C5-6 dislocation (**Figure 2a**). Traction was performed after a Philadelphia collar application. After a few hours he developed a complete tetraplegia and respiratory failure. He was intubated and artificially ventilated. On the third day, a halo-vest was applied and the patient was transferred to our ICU. On the second day after arrival, the patient was treated surgically by simultaneous anterior and posterior approach (**Figure 2b**). His neurological condition did not change and two weeks after surgery he died of pulmonary infection and related septicemia.

Discussion

The estimated incidence of AS in the general population is 1.4%. In patients with AS, the occurrence of traumatic cervical spine injury is three to four times greater than in the normal

population. The characteristic pathological lesions in AS are vertebral body osteoporosis, ankylosis of the apophyseal joints, intervertebral disc calcification and ligamentous ossification. The spinal column acts as if it is a single long bone with osteoporosis. The cervical spine loses its elasticity and becomes stiff and no absorption of head accelerations is maintained. Osteoporosis secondary to AS makes the cervical spine a single and relatively weak structure, acting as a stiff bar or lever, which bears the mass of the head on the upper end while supported by relatively strong thoracic vertebra (also strengthened by thoracic rib cage) on the lower end. Because of the flexion nature of originally ankylosed vertebrae, the forces causing extension can easily break this bar, and most frequently at sites that are close to the supporting or the weakest points (namely lower cervical regions). The affected vertebral column is transformed into a brittle, rigid column of tubular bone, which is very susceptible to fractures.(1, 3, 5, 7-8) The pathological process frequently involves the posterior elements and consequently the fractured spine may behave as a long bone fracture.(2-3) The cervical spine is a common fracture site which poses serious problems with a high degree of fatal outcomes. The mortality rate for cervical fractures was found to be 35% in patients with AS as compared to 20% for the normal population. The rate of severe neurological deficit is 57% for the former and 18% for the latter. (2, 4-5) Hyperextension

is the most common reason for fracture, mostly caused by minor trauma.(2-3, 5, 8) Up to 70% of cervical fractures occur through the calcified intervertebral disc.(5) These transdiscal fractures are more common in lower cervical vertebrae, which are often hidden by clavicles and scapula in the cases of kyphotic spine.(3-4) The abnormality on radiographic examinations can easily be missed because of the distortion of the anatomic architecture, osteopenia and difficulties in proper positioning of the patient.(5) Any unrecognized pathological change in the vertebral alignment of patients with AS may lead to devastating neurological consequences, so prompt and accurate radiological diagnosis of these fractures bears great importance.(3) All AS patients with neck pain must be managed as if they have a cervical fracture until proven otherwise. Radiological examination should include plain radiography, computerized tomography (CT) and MRI.(7) The X-ray films should include the full spine and should not be confined to the symptomatic region. CT is proven to be a rapid and safe method of diagnosis in these cases.(3) However, MRIs can evaluate the spinal cord and the canal at the level of injury and differentiate the causes of cord compression and identify cord contusion.(9)

The treatment of these cases is still controversial. Following adequate realignment, surgical stabilization plus external immobilization versus external immobilization alone are the treatment options.(1-2,5,7,10) When neurological deficit is present in association with the radiological evidence of spinal cord or root compression, decompressive laminectomy is preferred. Because there may be ossification of interlaminar ligaments and particularly when spinal stenosis is present, laminectomy requires a special technique and experience in order to avoid aggravating myelopathy. Steroid induced osteoporosis may soften the posterior bony elements and internal fixation by wiring is reported to be ineffective.(3-4) Considering the risks of surgery in these patients, in the absence of neurological deficit, persistent dislocation or bony fragments within the spinal canal, non-surgical external immobilization is advisable (3-4, 6). In majority of cases, because of the ossification tendency, it has been reported that a rapid healing occurs when external immobilization is adequately maintained.(3,10-11)

The complication rate is reported to be high in surgically treated patients (1) and the type of complications have been reported in detail in the literature.(1,5) They can be classified into three main groups: mortality increase, neurological impairment and loss of fixation.(1)

In patients without neurological deficit, external immobilization with the Halo apparatus mounted on a cast or a jacket is the preferred method because it provides rigid stabilization in all three planes and causes less discomfort for the patient. Halo-vests also allow early mobilization, quicken rehabilitation and prevent the physical and psychological problems that result from prolonged bed rest.(2,4,10) When

traction or external immobilization is planned, hyperextension should be considered as the main possible cause of secondary neurological deterioration.(1,5) Applications of traction should be performed when the cervical spine is in slight flexion or neutral position.(2,5,8) As observed in our cases, when the degree of kyphosis is low, both the traction and the fixation may be done in neutral position. The treatment of cervical spinal fractures is still controversial in cases with ankylosing spondylitis.

In cases with no neurological deficit, we advise nonsurgical treatment which includes a halo-vest application followed by closed reduction with skeletal traction in order to avoid the surgical complications and minimize the problems related to the prolonged bed rest. In cases with neurological deficit, an MRI should be performed in order to evaluate the presence of epidural hematoma, which is the main cause for the progression of the symptoms. Surgical interventions should be the first choice of treatment in patients with neurological deficits.

Conclusion

Cervical fractures in patients with AS are unique clinical entities with respect to their mechanism(s), diagnostic difficulties, management arguments and high mortality and morbidity rates. Every patient with a history of AS complaining of neck pain must be carefully evaluated even if there is no apparent medical history of trauma. Once the diagnosis is complete, deciding whether to operate or not must be guided by the patient's neurological status. If there is no neurological deficit, it is logical to try nonsurgical management with external reduction and stabilization; especially a halo-vest which obtains more reliable stabilization and early mobilization. But in the presence of neurological deficit, we recommend stabilizing the patient surgically. The physician must keep in mind that these fractures must be assumed to be unstable.

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Editor's Comments

There is accumulating evidence that the cervical spine in patients with ankylosing spondylitis is more prone to have fracture(s) than the normal population, even in the absence of trauma. Thus, the cervical fractures in patients with ankylosing spondylitis are unique with respect to mechanism(s) of injury, diagnosis, treatment and the prognosis. In the literature, there is different notions regarding the treatment of such patients, thereby the correct management of cervical fractures in the presence of ankylosing spondylitis still remains obscure.

The authors in this paper have discussed the clinical, radiological features, diagnosis and treatment of six patients with ankylosing spondylitis associated with cervical spine fracture. They treated the patients depending

mainly on the neurological status of the patients and I agree with the authors that the decision whether to operate must be guided according to the patient's neurological status. Conservative management including external reduction and stabilization especially with halo-vest should be performed in patients who have no neurological deficits. However, neurological deficits require surgical stabilization. The surgical treatment in a bone with poor quality requires a posterior and anterior fixation.

In general, the points discussed by the authors in the current study are consistent with previously published data and this paper is a useful contribution to the current literature.

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