SURGICAL MANAGEMENT OF SPINAL FRACTURES IN ANKYLOSING SPONDYLITIS: A CASE SERIES AND LITERATURE REVIEW

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ABSTRACT

Ankylosing spondylitis is a common disorder affecting 2 per 1000 individuals in the population. The hallmark finding is sacroiliitis with a variable degree of ascending spinal column involvement. Spinal fractures are a common complication and are characterized by instability, long lever arms across the fractured segment, and a high incidence of neurological injury.

We describe a case series of three patients with ankylosing spondylitis, who incurred typical spinal fractures seen in these patients, and presented to our unit over a three-year period. Our management highlights several of the important principles that should be employed. We further reviewed the literature on the subject and provide a review of current thinking on the subject.

Spinal fractures in patients with ankylosing spondylitis most commonly occur in the subaxial cervical spine and at the thoracolumbar junction. Conservative management is fraught with a high complication rates which includes pneumonia and secondary neurological injury and several studies recognize the advantages of early instrumented fixation and mobilization. As such these fractures are best managed surgically and there is a global trend towards this as the standard of care. A high incidence of neurological injury dictates early operative intervention to relieve compression and correct deformity and the prevention of secondary neurological injury is the fundamental aim of surgery.

Appreciating the nuances in the management of patients with ankylosing spondylitis who incur spinal fractures is essential to ensure an optimal neurological outcome. We provide a case series and literature review to provide spinal surgeons with an understanding of the specific considerations that should be employed, and thereby assist those who manage these patients.

Key words: Ankylosing spondylitis spinal fracture, Ankylosing spondylitis cervical fracture, Ankylosing spondylitis thoracolumbar fracture

INTRODUCTION

Ankylosing spondylitis comprises one of the seronegative spondyloarthropathies. The hallmark finding of the disease is sacroiliitis occurring together with a variable degree of spinal column involvement. The genetic marker of the disease is HLA-B27 positivity which occurs in 90-95% of sufferers. While strongly associated HLA-B27 positivity does not dictate clinical expression and in fact only 1-5% of individuals who are HLA-B27 positive develop the disease (1,2). Despite this the disease is relatively common affecting 2 per 1000 individuals with a typical age of onset in the 3rd to 5th decades and a male to female of 2:1 (3). The *modified New York criteria* allow diagnosis by clinical and radiological means and for the diagnosis of definite ankylosing spondylitis only one clinical and one radiological criterion is required. The clinical criteria used in the diagnosis are;

(i) Axial lumbar pain for >3 months that is relieved by exercise and not by rest
(ii) Limited lumbar motion in both the sagittal and coronal planes
(iii) Decreased chest expansion inappropriate for age.

The radiological criteria used in the diagnosis are:
(i) Unilateral sacroiliitis on X-ray which should be at least grade 3; or
(ii) Bilateral sacroiliitis on X-ray which should be at least grade 2 (4,5).

With regards to the spinal involvement the disease is characterized by ascending inflammation which erodes and then calcifies the ligamentous insertion points, facet joints and the annulus fibrosis (6). The reduced spinal mobility coupled with the reduced bone mineral density of the remodeled spine renders the spine in these patients susceptible to fracturing with otherwise trivial trauma (7,8). Besides the fracture itself, which is

commonly unstable, the neurological injury incurred is often devastating (9-11). The devastating neurological fall-out commonly incurred is explained by the site of spinal fracturing which almost always occurs in the sub axial and thoracolumbar spine (12,13). A study by Caron *et al.* (9) considered a radiology database of 122 subjects with ankylosing spondylitis who incurred spinal column fractures noted 62 (55%) to involve the cervical spine, 36 (32%) to involve the thoracic spine and 14 (13%) to involve the lumbar spine.

The severity of the neurological risk is quantified in a study that comprised a meta-analysis of 345 cases and noted a 18% three-month mortality in patients with ankylosing spondylitis admitted post incurring a spinal fracture (12). Epidemiological studies report that individuals with ankylosing spondylitis have a 4-times increased risk of incurring a spinal fracture than the general population. The life-time risk in these individuals is reported to be 5% to 15% (14). While spinal cord injury is common it is pulmonary complications that are the most frequent cause of mortality in these patients (9).

We describe a case series of three patients known with ankylosing spondylitis who were admitted to our unit over a three-year period with spinal fractures. Our aim is to provide treating spinal surgeons with a guide to surgical management of this specific patient subset.

CASE SERIES

Case 1

A 46-year old male patient presented to the Orthopaedic Department at our institution after being involved in a minor motor vehicle accident where he was bumped from behind while travelling at a low speed. His only complaint was that of isolated axial neck pain. The medical history obtained was that he was known with ankylosing spondylitis and he had no prior surgical history.

Neurological examination was normal and he had normal gait. Cervical X-ray imaging was performed and showed the typical "bamboo spine" of ankylosing spondylitis and a fracture through the body of C6 with a left C5/C6 unifacet dislocation. A CT cervical spine was performed to better delineate the fracture which confirmed the X-ray findings (Figure 1). Due to the absence of a myelopathy and no

symptoms of a radiculopathy an MRI of his cervical spine was not performed. A Philadelphia collar was applied, and he was admitted for operative reduction and stabilization.

Figure 1

Pre-operative sagittal CT cervical spine showing the fracture through the body of C6 with less than 25% of anterolisthesis (red arrow) and on the more lateral view a C5/C6 unifacet dislocation (green arrow) can be seen
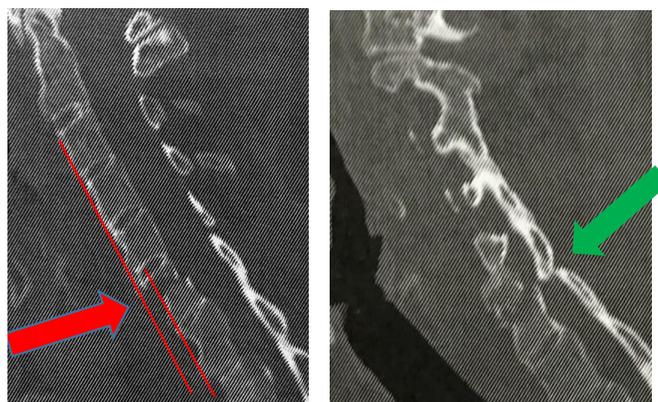
He was taken to theatre and due to the absence of significant cervical kyphosis standard prone positioning was utilized. A midline posterior neck incision was utilized. Once the laminae were exposed the left C5/C6 facet joint was burred to facilitate reduction of the unifacet dislocation. The bone was noted to be hard and brittle. To prevent injury to the spinal cord during the reduction laminectomies from C3-T1 were performed. In retrospect extending the laminectomies so far proximally and distally was an unnecessary precaution and a more limited decompression would have sufficed. Due to the brittle nature of the bone and the fact that the spine was ankylosed a long-segment construct was decided upon to ensure sound fixation and the decision was made to incorporate 3 segments above the fracture and 3 segments below the fracture. In the first part of the surgery C3 - C5 lateral mass screws were placed superior to the fracture where-after C7 lateral mass and T1 and T2 pedicle screws were placed inferior to the fracture. Placement of all screws was guided by fluoroscopy. The rods were placed and during reduction the fractured C6 vertebrae was reduced which was confirmed by fluoroscopy. The wound was closed in layers and the Philadelphia collar was re-applied which the patient wore for 6 weeks. At his 6-week follow-up out-patient appointment repeat X-ray's were performed which showed satisfactory fracture union (Figures 2-3).

Figure 2

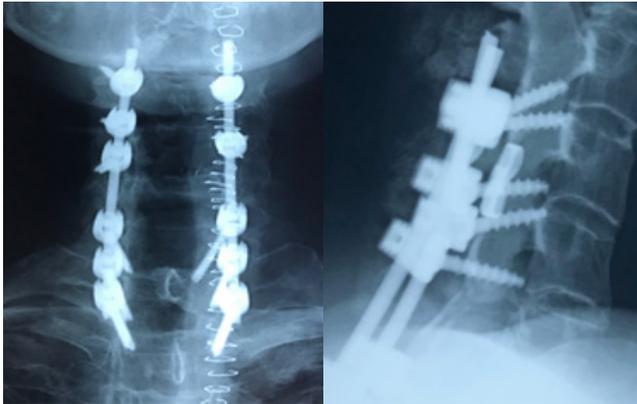*Post-operative 6-week antero-posterior and lateral cervical X-ray's showing fracture union*

Figure 3

Patient photograph at his 6-week follow-up appointment showing acceptable coronal and sagittal alignment

Case 2

A 30-year-old male patient presented to the Emergency Department at our institution after being hijacked while driving after which he had attempted to jump from the moving vehicle. The medical history obtained was that he was known with ankylosing spondylitis diagnosed by the modified New York criteria. He was fully conscious and only complained of an inability to move his arm or legs.

Neurological examination of his upper limbs on manual muscle testing revealed a quadriplegia with power 0/5 and a complete C5 sensory level (ASIA A). An urgent CT and MRI of his cervical spine was requested which sadly revealed a C5/C6 bifacet dislocation and on the MRI of his cervical spine the cervical cord was noted to be transected (Figures 4-5).

Figure 4

Pre-operative sagittal CT and MRI T2W cervical spine showing early changes of ankylosing spondylitis in this young patient and the C5/C6 bifacet dislocation is readily appreciated

The patient was admitted to the Intensive care unit and within 6 hours of the injury he was taken to the operating room for reduction of the dislocation and C5/C6 fusion using autologous bone graft. Due to the severity of his injury it was decided by the team that a single level anterior cervical discectomy and fusion was appropriate (Figure 6). As was expected his neurology did not improve and he was transferred to a local rehabilitation unit.

Figure 6

Post-operate lateral and anteroposterior cervical X-ray showing the reduced C5/C6 bifacet dislocation, the autologous bone graft, and anterior cervical plate in place with bi-cortical screw placement
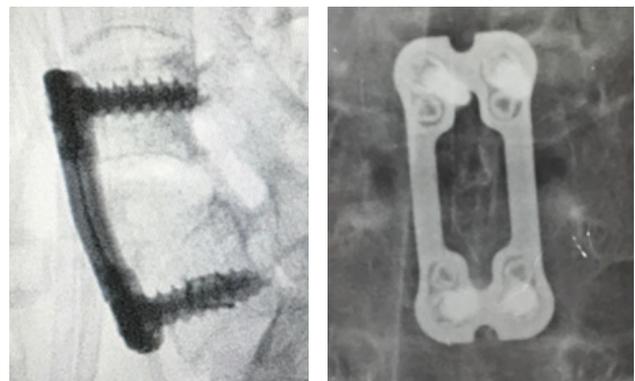
Case 3

An 81-year-old male patient was referred to our unit with the history that he had been kicked by his cow. He reported that he immediately experienced axial lumbar backpain of such severity, that although he was able to

move his legs, he was unable to walk. He reported no bladder or bowel symptoms. The medical history given was that he was known with ankylosing spondylitis.

Neurological examination utilizing manual muscle testing was normal. X-ray's of his lumbar spine revealed the typical "bamboo spine" of ankylosing spondylitis and an unstable dislocated L3/L4 fracture (AO Spine B-type injury) was noted with failure of the anterior column. A CT lumbar spine confirmed the X-ray diagnosis (Figure 7). Due to the absence of neurology an MRI of his lumbar spine was not performed.

Figure 7
Pre-operative sagittal lumbar CT scan showing the significantly displaced unstable L3/L4 fracture through the L4 vertebral body (AO Spine B-type injury)

The patient and his family were extensively counselled regarding a planned posterior decompression and L2-L5 pedicle screw and rod instrumented fusion however they declined surgical intervention. Utilizing non-operative management a lumbo-sacral brace was applied and he was kept in hospital for 6 weeks during which he received intensive non-weight bearing physiotherapy. During this period of conservative management his axial lumbar pain reduced, and he developed no neurological deficit. A follow-up CT of his lumbar spine post 6 weeks of conservative management showed the fracture displacement had reduced however a significant fracture gap was still

present (Figure 8). He was re-counselled regarding surgical intervention however he refused surgery. Once he could ambulate in his brace utilizing a walker, able to walk to the toilet and back, he was discharged for out-patient follow-up. He did not attend his follow-up appointment and despite several attempts was unable to be contacted.

Figure 8
Follow-up CT lumbar spine following 6 weeks of conservative management in a lumbosacral brace showing resolution of the fracture dislocation however a significant fracture gap is still present

DISCUSSION

Regarding the management of patients with ankylosing spondylitis who incur a spinal fracture it is reported in one Swedish study, that considered 1131 subjects from a National registry comprising 534 cervical fractures, 352 thoracic fractures and 245 lumbar fractures, that surgically managed patients had a statistically lower mortality rate than those managed conservatively (15). Several studies quantify an increased surgical complication rate and report pulmonary complications in up to 63% of patients, surgical site sepsis in up to 16% of patients and, adjacent level fractures to occur in up to 23% of patients (13,16,17).

Non-operative management: Patients with medical co-morbidities or simple wedge compression fractures (AO Spine A-type injury) are best managed conservatively.

Unfortunately in this population wedge compression fractures are rare and instability secondary to failure of the anterior or posterior tension band (AO Spine B-type injury), or instability secondary to failure of the anterior and posterior tension band (AO Spine C-type injury), are more common. Conservative treatment should be regarded as a poor second choice as bracing is often difficult in the presence of a fixed kyphotic deformity and prolonged bed rest significantly increases pulmonary complications (19).

Cervical fractures: While historical studies propose that unstable cervical fractures in patients with ankylosing spondylitis are best managed conservatively several studies report a significant global trend to operative intervention (18,20). One North American study reports, between 2003-2014, a 4-times increase in posterior spinal fusions and a 3.8-times increase in anterior cervical fusions performed for cervical fractures in these patients (20). Several other studies emphasize this point and recommend surgical stabilization for cervical fractures (21,22).

Cervical X-rays are often difficult to interpret due to kyphosis and interference from high shoulders. Due to the commonest site of fracturing being the subaxial cervical spine one study reported that 60% of cervical fracture dislocations were missed on screening initial radiography (23). CT imaging is often essential to increase the sensitivity in diagnosing fractures in these patients (24). Once an unstable cervical fracture is diagnosed reduction to pre-injury alignment, and not anatomical alignment, is the immediate concern. Traction should not exceed 5-10 pounds due to inherent paraspinal muscle incompetence and in the presence of kyphosis is hence commonly directed anteriorly and superiorly and maintained with pillows to support the back and head (25).

Intra-operative temporary fixation during positioning is best achieved with a halo brace, maintaining the degree of pre-injury kyphosis, which is then attached to a Mayfield clamp. It is recommended in several studies that unstable cervical fractures require combined anterior and posterior instrumentation to achieve adequate fixation. In many instances an anterior approach cannot be safely performed and a long segment posterior lateral mass or pedicle screw construct is advised to prevent implant failure (13, 18). Another study reported that anterior cervical fusion alone utilizing an anterior cervical plate with bicortical screw purchase is sufficient and reports the benefits of reduced operative time and post-operative complications (26). In the first case from our case series we performed the former approach advocated where a long segment posterior cervical instrumented fusion was performed. We considered this fixation stable and the patient's recovery was uneventful.

Thoracolumbar fractures: The thoracolumbar junction is the commonest site fractured in patients with ankylosing spondylitis who incur thoracolumbar fractures. Three types are distinguished namely;
(i)   Shearing type which typically presents acutely and often involves all three columns;
(ii) Compression type which are characterized by a late presentation;
(iii) Pseudoarthrosis which follows non-union union, commonly from multiple microfractures and secondary fibrosis (27).

All three types are usually unstable and require surgical treatment which is regarded as the optimal management (27,28). Several studies recognize the long level arms of the fused thoracic spine above, and fused lumbar spine below, that act across the fracture site. This translates in a very real risk of secondary neurological injury during patient transfers as well as if implant failure occurs before fracture healing has occurred (12,29). Inflammation of the anterior longitudinal ligament may cause aortic adhesions and injury including aortic lacerations and aortic dissection (14,31).

Definitive management has shifted from the historical conservative measures, such as bracing, to operative measures using long segment posterior instrumented fusions. Early indications for immediate definitive surgery include deteriorating neurology due to compression as well as an irreducible deformity (32). Mechanically monoaxial pedicle screws are preferred as they afford superior construct stability. Longer fusion constructs with pedicle screw placement at least 2 levels above and 2 levels below the fracture are recommended (9,18). Several studies report successful fixation with minimally invasive percutaneous pedicle screw and rod systems which avoid the paraspinal muscle dissection and subsequent scarring which further weakens these already inherently weak stabilizers (33,34). Post reduction, and post posterior instrumentation, fracture gaps are a special consideration in these patients and necessitate a combined posterior- anterior approach to avoid implant failure and pseudoarthrosis. Self-expanding cages or plating systems augmented by bone graft are recommended in these cases (18). In the third case from our case series we achieved an acceptable reduction of the L3/L4 fracture dislocation using bracing, which was confirmed on the 6-week follow-up lumbar CT scan, however a significant fracture gap was noted.

CONCLUSION

Patients with ankylosing spondylitis commonly incur spinal fractures post low-energy trauma and the commonest mechanism is falling. These fractures most commonly occur in the subaxial cervical spine and at the thoracolumbar junction and often incur devastating neurological consequences. Unstable injuries are best

managed by initial low weight traction to the pre-injury alignment and thereafter by long segment antero-posterior or long segment posterior instrumentation. Our case series provides illustrative examples of the typical types of spinal fractures incurred by these patients and our management highlights the surgical principles that should be employed.

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Declaration of interest

None of the authors have any financial nor personal relationships with other people, or organizations, that could inappropriately influence (bias) their work, all within 3 years of the beginning the work submitted.

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