CASE REPORT PEER REVIEWED | OPEN ACCESS

Deformity correction in the setting of acute cervical spine trauma in a patient with ankylosing spondylitis: A case report

Emmanuel N Osadebey, Karnesha Goins, Cierra N Harper, Damirez Fossett

ABSTRACT

Introduction: Ankylosing spondylitis (AS) is an autoimmune spondyloarthropathy marked by symptomatic alterations in skeletal anatomy and biomechanics. Ankylosis from the ossification of ligamentous structures and adjacent joints transforms the spine from flexible to rigid and brittle, easily susceptible to fracture. The pathophysiology of the condition is also notable for a progressive debilitating cervical kyphosis known as "chin-on-chest." Ultimately, the combination of a brittle, rigid, spine can permit trivial trauma to cause catastrophic injury, and in some instances, mortality. We discuss the multi-disciplinary approach, management concerns, and deformity correction in the setting of traumatic cervical spine fracture in a patient with ankylosing spondylitis.

Case Report: A 71-year-old man with ankylosing spondylitis presented to Howard University Hospital in a delayed fashion after a ground level fall at home. Neurological examination revealed loss of all motor and sensory function below the C4 level and an absence of rectal tone. Advanced imaging discovered a fracture-dislocation at the C4-5 level producing a severe hyper-lordotic angulation deformity. The profound fracture characteristics and displacement caused spinal cord compression posteriorly and tracheoesophageal stenosis anteriorly. The patient was treated operatively in a staged dual approach fashion correcting his pathologic deformity in consideration of long-term care needs. Post-operatively, upon completion of his final neurosurgical procedure, his sensory exam notable for return of sensation from C5-T1 and he was also able to appreciate and interact with the environment around him as his viewpoint was no longer rigidly caudally oriented. The patient was discharged from the hospital to a long-term care facility in stable condition.

Conclusion: Cervical fractures sustained after minor trauma in a patient with ankylosing spondylitis are not uncommon. However, surgical intervention with concomitant deformity correction in the traumatic setting is substantial undertaking with a paucity of literature on such surgical techniques. Surgical deformity correction in the acute trauma setting allowed for optimization of anticipated medical care initiatives and successfully provided newfound visual awareness of his environment, improving upon his ability to interact with the world.

Keywords: Ankylosing spondylitis, Cervical fracture, Deformity correction

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INTRODUCTION

Ankylosing spondylitis (AS) is an autoimmune spondyloarthropathy marked by symptomatic alterations in skeletal anatomy and biomechanics. The condition is known for its extensive vertebral column involvement, with the pathognomonic radiographic findings of a "bamboo spine" and bilateral sacroiliitis. Low bone mineral density is also very commonly observed. Werner et al. reported, in their series, that 62% of patients with AS had clinically relevant osteopenia or osteoporosis [1]. Ankylosis from the ossification of ligamentous structures, as well as zygapophyseal, and costotransverse joints, transforms a flexible spine into a rigid one which becomes easily susceptible to fracture [1]. Though minor low energy falls can result in fractures of the vertebral column, progressive deformity of the spine, devoid of trauma, is also quite common. In the cervical spine, wedging and micro-fractures of the vertebral bodies can cause a hyper-kyphotic deformity known as "chin-on-chest" syndrome [2]. This "chin-on-chest" deformity is both clinically and surgically problematic. Patients are relegated to a narrow scope of vision thus handicapping their ability to perform day-to-day tasks. Additionally, it can cause challenges with respiration and oral intake. Surgically, the chin-on-chest syndrome challenges the intubation process for general anesthesia, while simultaneously limiting the surgeon's ability to obtain adequate visualization of desired anatomy. Ultimately, the combination of a brittle, rigid, spine can permit trivial trauma to cause catastrophic injury, and in some instances, mortality. We discuss the multi-disciplinary approach, management considerations, and deformity correction surgical technique used in a patient with severe ankylosing spondylitis who had sustained acute cervical spine trauma.

CASE REPORT

A 71-year-old man presented to our Emergency Department after a ground level fall in which he struck the back of his head in the bathroom of his home. He was well-developed and well-nourished and did not show any science of acute distress (Temp: 98.5 °C, pulse: 92 beats per minute, BP: 160/77 mmHg, respiration rate: 18 breaths per minute, Oxygen saturation: 100% on room air). The patient remained conscious after the fall; however, he was incapacitated and unable to rise from the floor of his own volition. The patient was found by a neighbor approximately 12 hours after the incident. Emergency medical services were notified, and he was brought to Howard University Hospital. Upon inspection, the patient had a profound cervicothoracic kyphosis. Further examination revealed a loss of all motor and sensory function below the C4 level; as well, he had the absence of rectal tone. Imaging included computed tomography (CT) of the spine which identified a fracture-dislocation at the C4-5 level. The fracture was 100% posteriorly displaced producing a severe hyper-lordotic angulation deformity of the cervical spine, with consequent spinal cord compression posteriorly, and stenosis of the airway and esophagus anteriorly. Inspection of the cervical, thoracic, and lumbar spine imaging was notable for sweeping syndesmophytes of the vertebral bodies and diffuse osteopenia indicative of ankylosing spondylitis. A CT-angiogram of the neck was also obtained to assess the patency and integrity of the vertebral arteries. Compression of both vertebral arteries was noted as there was a paucity of contrast appreciated above the C3 vertebra. Overall, radiographic assessment of the patient showed that his cervical injury not only threatened the spinal cord but was also compromising his airway, upper gastrointestinal system, and cerebral perfusion. The patient was taken emergently to the operating theater where he underwent a decompressive laminectomy and posterior cervicothoracic instrumented fusion from C2-T1. The lab investigations were Sodium: 142 (135–145) mEq/L, Potassium: 4.2 (3.5–5.1), mEq/L, Chloride: 112 (95–111) mEq/L, Carbon dioxide: 20 (22–32) mEq/L, Blood urea nitrogen (BUN): 16 (7–25) mg/dL, Creatinine serum: 1.41 (0.6–1.2) mg/dL, Calcium: 9.3 (8.5–10.3) mg/dL, Total protein: 7.1 (6.2–8.3) g/dL, Anion gap: 14 (7–16) mEq/L, Creatine phosphokinase (CPK): 2650 (35–230) IU/L, Albumin:4.32 (3.2–5.5) g/dL, Lactic acid: 7.0 (0.5–2.2) mm/L, White blood cell count: 16.51 (3.2–10.6) ×10E9, Hemoglobin: 14 (14.6–17.8) g/dL, Hematocrit: 41.8 (40.8–51.9)%, Platelet: 246 (177–406) ×10E9. This would serve as the first stage of a planned two-stage operation, as he was assessed to be medically unfit to tolerate the physiological challenges of a combined anterior and posterior decompression and stabilization procedure.

Pre-operative concerns

As with any acute trauma, the basic management strategy of airway, breathing, and circulation (A-B-C) comes into play. Because of the patient's significant pre-existing kyphotic deformity, his baseline pulmonary function was expected to be sub-optimal. The severe angulation deformity caused by the fracture, as well as the steeple of the caudal fragment projecting forward into the anterior soft tissues of the neck (Figure 1), presented a significant worry, and was thought to be the most serious challenges regarding airway management.

Immediately upon presentation, consideration was made for early tracheotomy. However, this thought brought about some significant surgical concerns. Because of the proximity of the trach incision to the standard incision for an anterior approach to the cervical spine, the risk of infection loomed large in the conversation. Additionally, it was felt that the tracheotomy would need to be placed much further caudal than a standard tracheotomy because of, 1) the location of the fracture, 2) the area of impingement on the trachea and pharynx, and 3) the edematous soft tissue of the

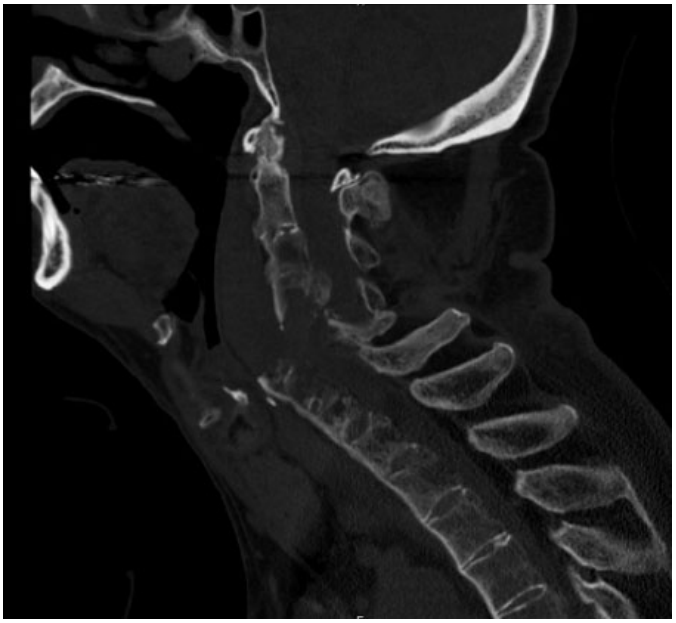

Figure 1: Pre-operative computed tomography of the cervical spine demonstrating the subaxial cervical fracture-dislocation with the caudal fragment steeple imbedded into the peri-pharyngeal soft tissues. Note the decreased bone mineral density of the spinal column and bridging ossification of the anterior longitudinal ligament classic of ankylosing spondylitis.

cervical spine. Tracheotomy was ultimately not performed because the patient had presented approximately 12 hours after being found down, yet during that time span he had been able to adequately maintain his airway, suggesting airway stability. Additionally, subsequent to his arrival in the ED, the patient remained fully communicative with no evidence of dyspnea on conversation and with normal levels of oxygen saturation while on room air. These facts suggested that however precarious the airway may have appeared on imaging, it was of no clinical significance. The decision was therefore made to electively intubate at the time of surgery. Endoscopic intubation was performed, and though it was difficult to identify the anteriorly displaced vocal cords, it was successful and without complications.

Given our patient's degree of vertebral column disruption and the anatomic location of the vertebral arteries within the transverse foramen, the partial or complete transection, dissection, and/or occlusion of one or both vertebral arteries was clearly of concern. Once again, the clinical exam was substantially helpful in allowing us to understand that the patient was achieving adequate blood flow to the mid-brain and brain stem either via collateral circulation from anterior to posterior, or direct posterior circulation from one or both of the vertebral arteries since he demonstrated no evidence of lower cranial nerve dysfunction or ocular dysmotility.

Imaging to further evaluate the pertinent skeletal and neuroanatomy was also a significant issue. Due to the patient's severe pre-morbid cervicothoracic kyphosis, his overall rigid body position was incompatible with positioning in a coil for cervical magnetic resonance imaging (MRI). This precluded our ability to obtain MRIs of the cervical spinal cord and MR angiography of the neck. Though restrictive of our ability to assess in a more detailed fashion the integrity of the spinal cord, presence of a hematoma, and degree of surrounding soft tissue edema, CT imaging and CT angiography were necessarily performed (Figure 2). CT-myelography to assist in evaluating the cervical neuroanatomy was deemed a non-viable option to employ as the patient had sustained an acute kidney injury secondary to rhabdomyolysis. The contrast load for myelography in addition to that for CT-angiography was felt to be too deleterious to the patient's already compromised renal function.

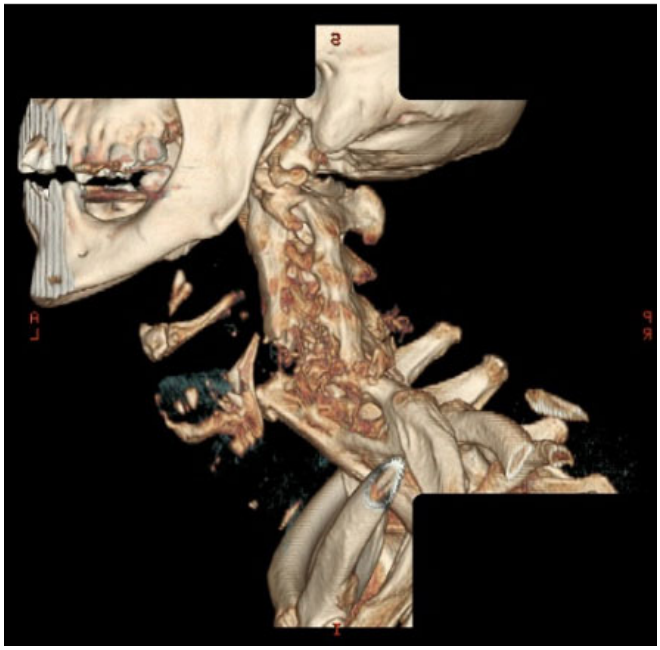

Figure 2: 3D reconstruction of pre-operative computed tomography.

Posterior operative technique

General anesthesia was induced and during endoscopic endotracheal intubation a significant narrowing of the airway caused by the underlying fracture was appreciated. Despite this narrowing, a patent airway was able to be successfully established. Baseline neuromonitoring data were recorded before and immediately after placing the patient prone in 3-point Mayfield fixation on a Jackson table. Comparative analysis demonstrated no change in electrical signals during positioning.

A standard midline approach to the cervical spine was performed, and assessment of bone quality revealed it to be soft and brittle. The major osseous injury at C4/C5 was easily visualized with comminution of the right lateral masses of the C4 and C5 vertebrae. Decompressive cervical laminectomies from C4 to C7 were performed revealing severe compression of the spinal cord at the

C4/C5 level by the C5 spinous process. Inspection of the dura showed a partial thickness tear without evidence of a cerebrospinal fluid leak. The partial thickness tear was repaired with prolene suture and augmented with DuraGen (Integra LifeSciences, Princeton, New Jersey/USA). Under fluoroscopic imaging, utilizing the Magerl technique, lateral mass screws were placed at C2 and C3 bilaterally. Pedicle screws were placed bilaterally from C7 to T2. Appropriately contoured rods spanning from C2-T2 were placed and locked into position thereby stabilizing the patient in situ. A crosslink was used to augment the construct and a mixture of morselized autograft and allograft (Osteofuse) was placed in the lateral gutters to promote bone fusion (Figure 3).

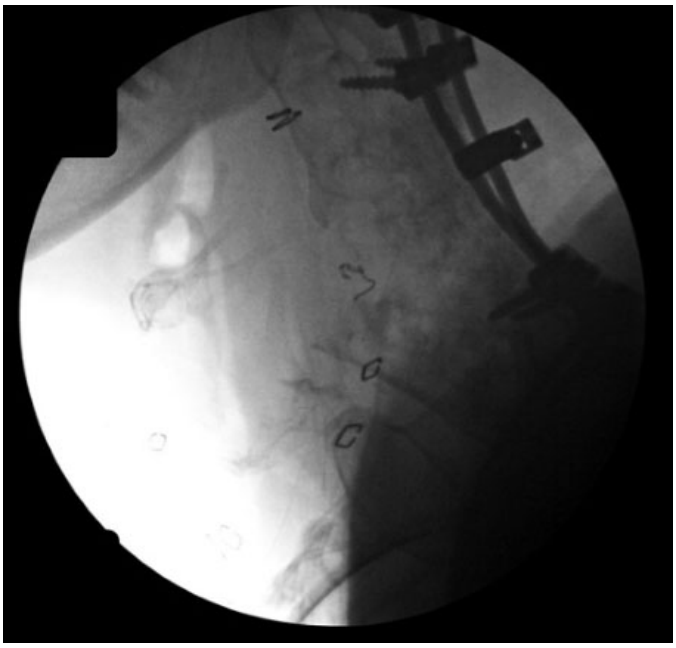

Figure 3: Intra-operative fluoroscopy of stage 1: Posterior instrumented stabilization.

Anterior operative technique

Between the posterior and anterior procedures, the patient underwent elective placement of a tracheostomy. After induction for the stage 2 procedure, the patient was anesthetized, placed on the ventilator, and baseline neuromonitoring signals recorded. A standard right-sided anterior longitudinal incision was fashioned and upon approaching the anterior cervical fascia, the surrounding soft tissues were noted to be considerably edematous. Tenting of the longus coli muscle by underlying bone just caudal to C4 vertebral body was appreciated with encroachment upon the esophagus. A Midas Rex high speed drill was utilized to shave down the tenting fracture fragment and under microsurgical vision, corpectomies were completed at the C4 and C5 levels. The inferior border of C3 and the superior border of C6 were then identified, the size of the bony defect measured, and an appropriately sized titanium vertebral body replacement cage packed with allograft was successfully placed. After placement of an anterior cervical plate spanning from C3 to C6, fluoroscopy was performed to confirm proper placement of all hardware.

Post-operative course

The immediate post-operative course was encouraging, with the posterior and anterior procedures staged 4-weeks apart in order to sufficiently address non-neurosurgical issues (Figures 4 and 5). The patient experienced the post-injury hemodynamic dysfunction commonly seen with high cervical spinal cord injuries, however with appropriate medical management adequate mean arterial pressures were maintained. In the early post-surgical period return of sensation to light touch from C5-T1 was appreciated and the patient's cognition was maintained as he was able to respond to questions by nodding his head and blinking his eyes appropriately. The patient was lost to follow up after being discharged from the hospital to a long-term care facility.

Figure 4: Computed tomography after stage 1: Posterior instrumented stabilization.

DISCUSSION

Cervical fractures sustained after minor trauma in a patient with ankylosing spondylitis are well documented in the literature. However, to our knowledge our case is the first to describe a complete spondyloptosis in which deformity correction was successfully achieved (Figures 5 and 6). Schneider et al. provide an account of deformity correction in the traumatic setting utilizing a novel surgical technique in a patient with a pre-existing cervical deformity from ankylosing spondylitis. However,

Figure 5: Computed tomography after stage 2: Anterior corpectomy, deformity correction, vertebral body replacement, and plate stabilization.

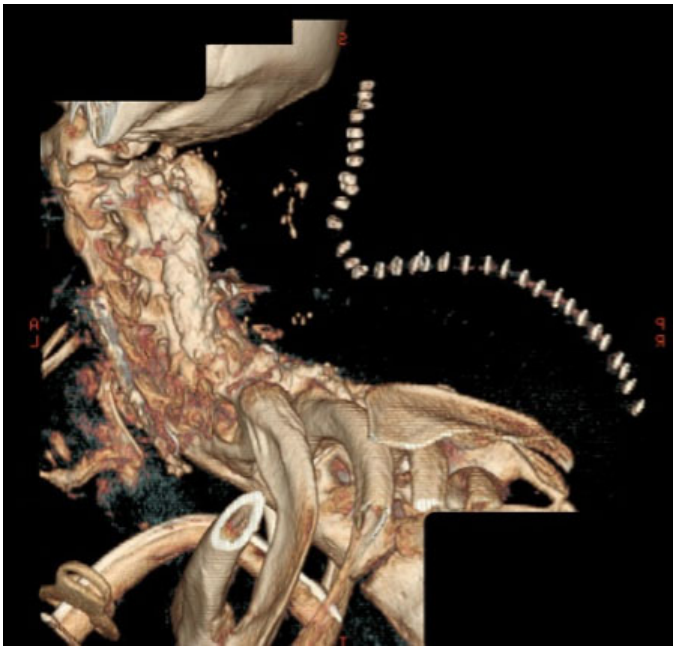

Figure 6: 3D reconstruction post-operative computed tomography after stage 2.

their patient's radiographs on presentation was devoid of abnormality with mild to moderate displacement at the C6/C7 level appreciated on subsequent advanced imaging while immobilized in a halo vest [3]. The remarkable fracture displacement seen on presentation in our patient resulted in respiratory and esophageal compromise. Acute management of pertinent multiple organ systems as well as the surgical fixation techniques utilized differentiate the clinical severity of the trauma from the few accounts of the matter in the literature. The sequela of the osseous injury produced acute multi-organ system compromise, additionally challenging both the acute and the long-term medical and surgical management of the patient. There is a paucity of literature providing strong recommendations for the management of trauma-induced angulation deformities of the cervical spine in the setting of ankylosing spondylitis and a pre-morbid chin-on-chest syndrome. The description provided by Schneider and associates as well as Urbański et al. who too reported on correction of a mild to moderately displaced cervical spine fracture electing to correct their patients deformity with a single approached and additional osteotomies are the only available accounts of acute deformity correction in the traumatic setting to our knowledge [3, 4]. The performing surgeon in our case elected to proceed with a posterior approach to the cervical spine as the initial procedure in this case because the primary surgical goals of decompression and stabilization would be appropriately and expeditiously achieved. Additionally, because our patient had sustained injuries which affected multiple organ systems, it was felt that if a decline in systemic function delayed the second stage of his operative management, posterior stabilization could more reliably stand alone as the only treatment option offered. There was minimal confidence that anterior stabilization alone would provide sufficient long-term support of the spine in the same scenario.

The incidence of spinal cord injuries associated with cervical fractures in the AS patient population is substantial. Westerveld et al. found that 57.1% of ankylosing spondylitis patients with traumatic spinal cord fractures had concomitant neurological disease with poor prognosis for improvement compared to 12.6% in the non-ankylosed control group [5]. Well aware of the high perioperative mortality and morbidity rates associated with upper cervical spinal cord injuries, an effort to optimize long-term care and function was of the utmost importance in directing this patient's management. His pre-injury chin-on-chest kyphotic deformity was medically and functionally significantly problematic. Reduction of his fracture-dislocation would re-create his severe kyphosis making securing a safe and patent airway by tracheostomy all but impossible to achieve. Additionally, it would also create the need for a permanent gastrostomy to mitigate swallowing difficulties, and provide a means for oral intake. Lastly, reduction and fixation of his fracture would once again severely restrict the patients' scope of vision and thus his ability to interact with the surrounding environment in any kind of meaningful way. By contrast, kyphotic deformity correction by in situ stabilization of the fracture-

dislocation provided airway patency for prolonged or even permanent ventilation via a tracheostomy tube, maximized speech, and swallowing capabilities thereby avoiding the need for gastrostomy tube placement, and optimized our patient's ability to see and interact with the surrounding world in a manner that had slowly eluded him for years. The fracture dislocation this patient sustained, though tragic and severely anatomically aberrant, placed our patients neck in a more appropriate functional position. Ultimately, the decision to stabilize the spine in this functionally beneficial position versus reduction and stabilization in an anatomically correct position was made by thinking about long-term management and quality of life issues in this severely debilitated patient. Optimizing the success of future medical care initiatives, and allowing him to integrate with his surrounding environment were vitally important goals that were met by our treatment paradigm.

CONCLUSION

We describe the management considerations of an elderly gentleman with a profound pre-injury chin-on-chest deformity secondary to ankylosing spondylitis, who sustained a C4/5 spondyloptosis secondary to a fracture-dislocation of the cervical spine. We have provided in detail our management strategy and the thought processes involved. The paradigm of A-B-C-D-E (airway, breathing, circulation, decompression, and establishing stability) is a straightforward pathway, but the more complicated the scenario, the more issues arise for consideration. Ultimately long-term medical management and quality of life are the driving forces behind decision making.

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Author Contributions

Emmanuel N Osadebey – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Karnesha Goins – Conception of the work, Design of the work, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Cierra N Harper – Acquisition of data, Drafting the work, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Damirez Fossett – Conception of the work, Design of the work, Analysis of data, Interpretation of data, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

Guarantor of Submission

The corresponding author is the guarantor of submission.

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Consent Statement

Written informed consent was obtained from the patient for publication of this article.

Conflict of Interest

Authors declare no conflict of interest.

Data Availability

All relevant data are within the paper and its Supporting Information files.