Complications and Survival After Long Posterior Instrumentation of Cervical and Cervicothoracic Fractures Related to Ankylosing Spondylitis or Diffuse Idiopathic Skeletal Hyperostosis

Yohan Robinson, MD, Anna-Lena Robinson, MD, and Claes Olerud, MD, PhD

Study Design. Prospective cohort study.
Objective. This study investigates the results of long posterior instrumentation with regard to complications and survival.
Summary of Background Data. Fractures of the cervical spine and the cervicothoracic junction related to ankylosing spinal disease (ASD) endanger both sagittal profile and spinal cord. Both anterior and posterior stabilization methods are well established, and clear treatment guidelines are missing.
Methods. Forty-one consecutive patients with fractures of the cervicothoracic junction related to ASD were treated by posterior instrumentation. All patients were followed prospectively for 2 years using a standardized protocol.
Results. Five patients experienced postoperative infections, 3 patients experienced postoperative pneumonia, 2 patients required postoperative tracheostomy, and 1 patient had postoperative cerebrospinal fluid leakage due to accidental durotomy. No patient required reoperation due to implant failure or nonunion. Mean survival was 52 months (95% confidence interval: 42–62 mo). Survival was affected by patient age, sex, smoking, and spinal cord injury.
Conclusion. Patients with ASD experiencing a fracture of the cervicothoracic region are at high risk of developing complications. The posterior instrumentation of cervical spinal fractures related to ASD is recommended due to biomechanical superiority.

Key words: spinal fractures, spinal fusion, ankylosing spondylitis, diffuse idiopathic skeletal hyperostosis, postoperative complications, mortality.
Level of Evidence: 4
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Ankylosing spondylitis (AS) and diffuse idiopathic skeletal hyperostosis (DISH) lead to reduced mobility of spinal segments. Although 2 completely different entities, these ankylosing spinal diseases (ASDs) have 1 important biomechanical feature in common: the relating long lever arms result in potentially hazardous instability in case of vertebral fractures. Especially, fractures of the ankylosed cervical spine and the cervicothoracic junction (CTJ) are a serious condition, because both sagittal profile and spinal cord are endangered.1-3 Treatment may be complicated due to comorbidity, and the mortality of fractures related to AS or DISH is considerably high.4,5

During the last decades, antirheumatic treatment of AS with disease modifying drugs has improved the course of the disease dramatically,6 still, the incidence of cervical spinal fractures related to AS is increasing steadily.6

Multiple operative techniques have been developed to address the instability of these fractures. Traditionally, surgeons have chosen the anterior approach in cervical fracture treatment, because it can be performed in supine position avoiding dislocation during positioning. The anterior approach is furthermore less traumatic and less likely to lead to soft-tissue complications. Still, the anterior approach is complicated by the kyphotic deformity in AS, minimizing the possibilities of reaching the region of interest. In addition, it is hardly possible to neutralize the long lever arms of an ankylosed spine from anterior if the fracture is in the lower cervical spine.

Many surgeons nowadays prefer the posterior approach to stabilize AS-related fractures of the cervical spine and the CTJ.7 The main advantage is the possibility of long lever arm neutralization even in the CTJ. The complication rate of posterior instrumentation in ankylosing spine disease is high, but specific data with regard to long cervicothoracic fixation are...
not available, yet. The aim of this study is to investigate the common practice of long posterior instrumentation of cervicothoracic fractures with regard to complications and mortality.

**MATERIALS AND METHODS**

Between 2007 and 2011, 41 patients (35 males, 6 females, age 71 ± 12 yr [48–95 yr]) with fractures of the CTJ related to AS (n = 31) or DISH (n = 10) were treated by posterior instrumentation at a first level trauma center. All patients were followed prospectively for 2 years using the standardized protocol of the Swedish Spine Registry SWESPINE. Recorded were patient-reported baseline data, complications, and pain, as well as surgeon-reported data on surgical method. Data on perioperative bleeding and surgical time were added after a journal review. Mortality data were obtained from the Swedish Mortality Registry, where also survival data of all patients with subaxial cervical fractures (International Classification of Diseases, Tenth Revision: S12.2) treated between January 1, 1996 and December 31, 2011 registered in the Swedish National Hospital Discharge Registry were extracted.

The Statistical Package for Social Sciences (SPSS 22.0, IBM Corp., Armonk, NY) was used to perform the statistical analysis. Mean values are presented with standard deviation, followed by intervals in brackets. The U test by Mann, Whitney, and White was applied to compare means. Mean survival was estimated with the Kaplan-Meier method. The influence of possible covariates on survival was tested with Cox regression analysis, presented as hazard ratio (HR).

This study was approved by the regional ethical review board of Uppsala (dnr 2010/131, dnr 2010/297/1).

**RESULTS**

Of the 41 patients included in this study 31 had fractures of the CTJ related to AS, 10 patients had DISH. All fractures were involving anterior and posterior, bony, and ligamentous structures of the spine at the level of injury, and were classified as type I according to Metz-Stavenhagen et al. All fractures were hyperextension injuries and classified type B4 M2 according to the new AOSpine classification. One patient had a C2–C3 injury, 1 patient a C3–C4 injury, 5 patients a C4–C5 injury, 5 patients a C5–C6 injury, 20 patients a C6–C7 injury, and 9 patients a C7–T1 injury.

Neurological deficit was found in 11 patients (Frankel A: n = 6, Frankel C: n = 4, Frankel D: n = 1). The remaining patients had no neurological deficit (Frankel E: n = 30). The occurrence of an epidural hematoma was not assessed in all patients because magnetic resonance imaging was only performed in neurologically deteriorated patients.

All patients were treated by posterior stabilization from the upper cervical to the upper thoracic spine using titanium screw-rod systems (Figure 1A–E). Screws were inserted in 3 vertebrae cranially and 3 vertebrae caudally of the injury. If extending to C1, Goel-Harms screws were placed, in C2 short, nontransarticular Magerl screws were placed, subaxially in most cases lateral mass screws were placed, whereas in the thoracic spine transpedicular screws were placed 3 levels below the injury. Thirty-seven of the instrumentations were cranially extending to C2, 2
to C1, and 2 to occiput. Caudally, in 44%, the instrumentation ended at Th3, in 27% at Th2. Decompression by laminectomy was performed in the 11 cases with neurological deficit. Mean surgical time starting from the placement of the skull clamp to the release of the skull clamp was 255 ± 90 minutes (80–488 min), bleeding 2128 ± 3005 mL (300–17,000 mL). Postoperatively, no external immobilization was applied.

Pre-existing comorbidity was common. Six patients were smokers. Arterial hypertension was found in 24 patients, and a cardiac comorbidity was present in 16 patients. Diabetes mellitus was evident in 9, of which 5 were insulin dependent. Seven patients had some kind of psychiatric disorder. Four patients had pulmonary and 4 had renal comorbidity. Three patients had a history of malignancy.

Five patients experienced postoperative infections, of which 4 could be treated with antibiotics alone, but one required revision surgery. Three patients had postoperative pneumonia, 2 patients required postoperative tracheostomy due to pulmonary insufficiency, and 1 patient had postoperative cerebrospinal fluid leakage due to accidental durotomy.

At 1-year follow-up, the patients reported a neck pain of 24 ± 25 (0–85) and an arm pain of 21 ± 24 (0–84) on the 100 points visual analogue scale. Neck pain improved after 2 years to 14 ± 18 (2–54) visual analogue scale, arm pain to 20 ± 27 (0–67). No patient required reoperation due to implant failure or nonunion during the first 2 years.

The mean survival was 52 ± 5 months (95% confidence interval: 42–62 mo). The Kaplan-Meier survival plot is presented in Figure 2. Cox regression analysis revealed that survival was significantly affected by patient age (HR = 1.22; P < 0.001), female sex (HR = 0.05; P = 0.024), smoking (HR = 23.23; P = 0.038), and Frankel A spinal cord injury (SCI) (HR = 8.31; P = 0.020). Survival was not affected by the type of ankylosing disease, comorbidities, level of cranial or caudal fixation, surgical time, or amount of surgical bleeding.

DISCUSSION

Complications of Posterior Spinal Fracture Stabilization in Ankylosing Spine Disease

Patients with ankylosing spine disease experiencing a fracture of the cervical spine or the CTJ are high-risk patients. (1) The fracture itself is due to the long lever arms of the cranial and caudal fragment highly unstable and requires rigid stabilization measures, involving neutralization of lever arms.3 (2) The transverse injury of all bony (and ligamentous) structures endangers the spinal cord, and neurology may deteriorate, if complete SCI is not already present after the injury.3,8 (3) Because of the patients’ usually kyphotic deformity, which is often prevalent after lifelong AS, external stabilization is troublesome, because collars will not fit and halo-type fixations will loosen if the head is positioned more anteriorly. (4) The osteoporosis related to AS may cause delayed union due to implant loosening, and subsequently instrumentation failure.12 (5) The reduced soft-tissue coverage over the CTJ in ankylosed patients predisposes for wound healing deficiency, especially in the presence of kyphosis. (6) Considerable surgical blood loss related to AS must be expected, which may require massive transfusion.14 (7) Most patients with fractures related to ankylosing spine disease are at a higher age, and often have numerous comorbidities.2 Comlications are therefore expected, and in most cases rather related to the underlying disease than to the surgical treatment.

The retrospective study by Caron et al8 reported complications in 84% of the 112 patients treated for fractures in ankylosing spine disease, most of them similar to those found in our study group (Table 1). They had pulmonary complications in 35% of all treated patients, but could not relate this complication to the type of treatment. Whang et al3 found
pulmonary complications in 13% of the 30 treated cases. Interestingly, all 3 reported fatal pulmonary complications occurred in patients with halo vest treatment, again demonstrating the drawback of external cervical immobilization. In the series of de Peretti et al, 63% of the 48 conservatively treated patients with AS or DISH had bed-rest-related complications because of decubital ulcers or pneumonia. In this context our pulmonary complications rate appears relatively low, which is possibly caused by the beneficial effect of early rehabilitation without the necessity of bed rest, orthosis, or halo vest immobilization.

Spinal fractures related to AS or DISH are associated with considerable bleeding. It is generally known that patients with AS have a greater amount of blood loss. Reasons for this may be (1) complicated positioning causing vena cava compression, (2) increased vascularity if skeletal structures due to the inflammatory neovascularization, and (3) unawareness and delay of massive transfusion protocol activation. Olerud et al had a mean surgical blood loss of 3500 mL (0–16,500 mL), Cornefjord et al 2119 mL (450–6800 mL), Lu et al 2100 mL, and our study 2128 mL (300–17,000 mL). Multiple cases with massive blood loss are described. Olerud et al had 2 cases of bleeding more than 10 L, Tetlakaff et al 1 case with 17-L blood loss, and our study had 1 case of 17-L massive blood loss. Early communication of the expected blood loss, and timely activation of the massive transfusion protocol is recommended to avoid deterioration of this potentially fatal condition.

With regard to loss of fixation, the available publications present a dramatic reduction of implant or fixation failures during the last decades (Table 1). Although Olerud et al reported 1996 a loss of fixation in 23% of the treated cases, Einsiedel et al found 2006 only 14% failures, which was further improved 2009 in the study by Whang et al to 7% and in 2010 by Caron et al to 1%. The case series of thoracicolumbar fractures related to AS by Hitchon et al of 2002 sticks out in this regard, because it was free of implant failures. They chose in contrast to most published studies of that time continuously a multilevel posterior approach for surgical fracture stabilization. This approach has been popularized during the last decade and recently published studies have few cases of implant failure. In the presented study the multilevel posterior-only approach was chosen, which had successful results with regard to healing and implant stability. Obviously, the long posterior lever arm neutralization was biomechanically reasonable.

One major drawback of posterior stabilization in the CTJ is the critical soft-tissue covering of the mostly severely kyphotic area. Postoperative wound infections of up to 16% were reported, requiring in some cases revision surgery and/or long-term antibiotic treatment. Most studies were including even thoracic and lumbar fractures. DVT indicates deep vein thrombosis.

<table>
<thead>
<tr>
<th>Publication</th>
<th>Year</th>
<th>N (all)</th>
<th>N (surgical)</th>
<th>Wound infection (%)</th>
<th>Pulmonary complications (%)</th>
<th>Tracheostomy (%)</th>
<th>Dural tear (%)</th>
<th>Epidural hematoma cord compression (%)</th>
<th>Loss of fixation (%)</th>
<th>Most studies were including even thoracic and lumbar fractures.</th>
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<td>1</td>
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<td>2010</td>
<td>112</td>
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<td>15</td>
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<tr>
<td>de Peretti et al</td>
<td>2004</td>
<td>31</td>
<td>31</td>
<td>10</td>
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invasiveness was associated with postoperative wound infections.23 Furthermore, prominent spinous processes may be a constant threat to CTJ skin covering, and may need revision surgery with resection of these bony structures.1

**Spinal Cord Injury and Mortality of Cervical Fractures Related to AS or DISH**

In our series, 27% had SCI associated with the injury. Caron et al7 had 58%, and Whang et al22 even had 67% with SCI. The high occurrence of SCI documents well the neurological hazard a fracture related to ankylosing spine disease represents. Caron et al7 found during follow-up improved neurological function in 34% of the patients, still 5% had worsened neurology. Rowed21 even reported 19% with worsened neurology after initial treatment. Common reasons for neurological deterioration are dislocation of the fracture with spinal cord compression and epidural hematoma. Already during the prehospital resuscitation and transport, neurologically fatal fracture dislocations may occur, if a patient with a kyphotic deformity is forced onto a spine board. Tracheal intubation often requires fiberoptic assistance to avoid dislocation of a cervical fracture. Furthermore, fracture displacement may occur in the operating theater during patient positioning in general anesthesia, where neither muscle tone nor reported pain protect the spinal cord from injury.24,31 Dislocation with neurological worsening may even occur during conservative fracture treatment.26 Extensive bleeding from ruptured epidural veins and the cancellous bone of the vertebral fracture may cause severe epidural hematoma.22 Fracture mobility hinders proper clotting, and an initially harmless fracture hematoma can develop into a large epidural mass causing spinal cord compression. This is especially true if the patient has clotting malfunction.

It is well known that SCI is associated with greater mortality.22 With regard to the considerable number of patients with SCI the relatively high mortality of spinal fractures related to AS is not surprising. The published 1-year mortality after fractures related to ankylosing spine disease ranges between 24% and 33% (Table 2). A patient in Sweden with a surgically treated subaxial cervical fracture without AS has a mean survival of 218 months (95% confidence interval: 213–225 mo; unpublished data from 2059 cases in the Swedish National Hospital Discharge Registry), but the patients in our study only had a mean survival of 52 months (95% confidence interval: 42–62 mo) (Figure 2). Other factors than ankylosing spine disease may have an influence on patient survival and are not adjusted for in this comparison. For example, the patients have higher age and more comorbidity in our study than in the general C-spine trauma population. In our study population, survival was significantly affected by complete SCI (Frankel A) associated to the fracture. The series in patient with DISH-related cervical fractures by Meyer et al29 found a strong association of neurological injury and mortality, as well, although, despite the relatively high number of included patients, Caron et al7 could not relate SCI to increased mortality. Still, most studies agree, that about 25% of patients with spinal fractures related to AS or DISH do not survive longer than 1 year after the injury.

**TABLE 2. Published 1-yr Mortality After Spinal Fracture Treatment in Ankylosing Spine Diseases**

<table>
<thead>
<tr>
<th>Publication</th>
<th>1-yr Mortality (%)</th>
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<td>This study</td>
<td>24</td>
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<tr>
<td>Schoenfeld et al9</td>
<td>33</td>
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<td>Caron et al7</td>
<td>32</td>
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<td>Meyer29</td>
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<td>Olerud et al6</td>
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**Classifications of Spinal Fractures Related to Ankylosing Spondylitis**

Because of the biomechanics of the ankylosed spine the classical 3- or 2-column models are not applicable for spinal fractures related to AS or DISH.3 The new AOSpine fracture classification system therefore introduced the modifier M2 for ankylosing spine disease to mark the severity of the injury.31 During the last decades multiple classifications have been developed concerning spinal fractures related to AS.

In the Germanic region, most commonly the classification by Metz-Stavenhagen et al31 is used, which describes 2 subtypes: type I, the complete disruption of anterior and posterior bony and ligamentous structures, and type II, the sintering fracture, often after a minor injury, unnoticed by the patient, and they may be confused with an Andersson lesion, the inflammatory discitis related to AS.3

The classification of de Peretti et al32 describes 4 fracture types according to their radiographical dislocation: type I with anterior opening, type II with horizontal dislocation, type III nondisplaced, and type IV being similar to spinal fractures unrelated to AS. The de Peretti type III fracture is related to delayed diagnosis, but no impact of fracture type on outcome was described.17 Furthermore, radiographical displacement classifications are highly sensitive to positioning of the patient during examination.

The classification introduced by Caron et al7 involves the radiographical course of the fracture line: type I a transdiscal disruption, type II a vertebral body injury, and types III and IV being a combination of disc and vertebral body injuries. No impact of fracture type on patient treatment or outcome has been described until now. Because a transverse injury of the ankylosed spine can biomechanically be compared with a transverse diaphyseal long bone fracture, the classification of “minor” radiographical differences remains academic and without any clinical impact.

**Anterior Versus Posterior Versus Circumferential Stabilization**

Supporters of the anterior stabilization technique for fractures related to AS point out that the anterior access is less traumatic, minimizes the risks of displacement during positioning, and has less postoperative infections.3
And indeed, in nonankylosed spinal columns most surgeons would choose an anterior approach to stabilize the noncompressed fracture, with the benefits of a lesser traumatic access, greater fusion rate, and kyphosis reduction. But, in the nonankylosed spine the anterior-only approach is not recommended for transverse, rotationally unstable fractures. Because spinal fractures related to AS are virtually almost transverse injuries there is no reason to deal differently with them. Beyond that, many patients with AS are highly kyphotic, and the anterior access is anatomically impossible. Furthermore, a long lever arm neutralization of cervicothoracic fractures from anterior requires long anterior plates extending into the upper thoracic region, which then would require access extension into the upper mediastinum. In addition, most surgeons do not routinely perform bicortical anterior screws, which would be necessary to get as much screw purchase as possible in the osteoporotic bone of the ankylosed spine. The inferiority of the anterior-only approach is reflected by the results of several studies, where implant failure solely occurred in the anterior-only treated patients.

Some authors regularly perform combined posterior-anterior or anterior-posterior stabilization of patients with spinal fractures related to AS or DISH. This approach adds anterior stability to the posterior stabilization. Unfortunately, the possible complications of the second access are added as well.

In our experience if a long posterior instrumentation is performed, the anterior access will become obsolete, because stabilized fractures related to AS have a tendency to heal, even if anterior defects are present. In case of spinal cord compression by an irreducible anterior fragment, the anterior approach has to be added of course, but this is rarely found in the hyperextension injuries of patients with AS or DISH.

**CONCLUSION**

Fractures of the CTJ related to AS or DISH are highly unstable injuries, and most patients experience multiple complications during treatment. The suggested type of treatment using long posterior constructs allows early rehabilitation without bracing at a considerably low complication rate. In most patients, the posterior instrumentation construct is biomechanically most feasible and rarely requires additional anterior support. Long posterior constructs in case of cervical fractures related to AS or DISH should be considered as primary choice, whereas other stabilization measures may have their place, but seem to be inferior with regard to biomechanical stability.

**References**


**Key Points**

- Complications are common in the treatment of fractures in the CTJ related to ASD.
- Survival after CTJ fractures related to ASD is negatively affected by tobacco smoking and SCI.
- Long posterior instrumentation of CTJ fractures related to ASD can be recommended to avoid biomechanical failure.


