

The importance of the occipitocervical area in patients with ankylosing spondylitis analysis of a cohort of 86 cervical fractures in surgically treated patients

ABSTRACT

Study Design: This was a retrospective analysis of prospectively collected data.

Objective: The effect of C0-C1-C2 cervical ankylosis in patients with ankylosing spondylitis (AS) is not documented. The objective of this study is to describe the radiological characteristics of the occipitocervical junction in patients with AS operated for a cervical fracture and to correlate them with their clinical evolution.

Materials and Methods: Analysis of patients with ankylosing spondylitis (AS) treated in a single institution of a cervical vertebral fracture between 2007 and 2014 who were prospectively followed through the SWESPINE registry. The integrity of the C0–C1–C2 joints was determined and classified into fused and nonfused joints. By determining the angle between C0–C1 and C1–C2 joints in the coronal view of the computed tomography scan (X-angle), the progressive degeneration of these joints was described. Intra- and inter-observer reliability of this test was determined. The instruments of health-related quality of life (QOL) and disability were EQ5D and Oswestry disability index (ODI), respectively.

Results: A total of 86 patients with AS treated surgically for cervical fracture had complete facet ankylosis between C3 and T1 due to their pathology. Mean age 69.2 years (standard deviation [SD]: 11.7). The most common level of fracture was in C5–C6. In 24 patients, the C0–C1 joint was fused, and in 15 patients, C1–C2 joint was fused. The intra- and inter-class reliabilities for X-angle measurement were very high (intraclass correlation coefficients = 0.94; 0.92). The mean X-angle was 125 (SD: 12) in nonfused patients and 136 (SD: 14) in fused patients ($P < 0.001$). There were no differences in QOL and disability at 2 years between the two groups: EQ5D-index of 0.54 and 0.55 ($P = 0.5$), ODI of 26.4 and 24, ($P = 0.35$) respectively.

Conclusions: X-angle is a reliable measure for joint integrity C0–C1–C2 in patients with AS. Total cervical ankylosis including the C0-C1-C2 segments is not related to poorer QOL and disability in these patients.

Keywords: Ankylosing spondylitis, cervical fracture, occipitocervical, X-angle


INTRODUCTION

Ankylosing spondylitis (AS) is an inflammatory rheumatic disease that can affect the cervical spine leading to a progressive ossification, rigidity, and kyphotic deformity, often altering the normal horizontal gaze. The reported prevalence of this pathology is 0,1%–1,4%.^[1] The cervical spine is the most affected area and can present spinal cord injury in about 27% of the patients.^[2,3] Usually, the mechanism of production is a low energy trauma in hyperextension of a stiffed kyphotic

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cervical spine, classified as type B4 M2 according to the new AOSpine classification system.^[4] Due to the thin fracture lines, high-resolution computed tomography (CT) imaging is preferable as one of the most reliable tools for identifying the fracture levels, usually transverse or short oblique passing through disc and bone.^[5] The most affected area is C5–C7.^[6] These fractures 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 considerable low complication rate. In most patients, the posterior instrumentation construct is biomechanically most feasible and rarely requires additional anterior support.^[2,7] Clinical outcomes are well-described in patients with AS in terms of complications derived from the fracture itself or from the treatment [Table 1], but data about quality of life (QOL) and disability are missing. The aim of this study is to determine if level of previous ankylosis affects the fracture pattern and the posterior clinical outcome. Our study is focused in the craniocervical junction of these patients. There is no information regarding how upper cervical ankylosis (C0-C1-C2) affects clinical outcomes in this cohort of patients.

MATERIALS AND METHODS

Analysis of patients with AS surgically treated in a single institution of a cervical vertebral fracture between 2007 and 2014 who were prospectively followed for 2-year postfracture using the standardized protocol of the Swedish Spine registry through the SWESPINE. Neurological status was classified with Frankel scale. The previous integrity of the C0-C1-C2 joints was divided into 3 types: type A was normal articulations, type B was degenerative, and type C was fused joints [Figure 1]. For the data analysis C0-C1-C2 joints were divided into fused and nonfused joints. By determining the angle between C0–C1 and C1–C2 joints in the coronal view of the CT scan, the progressive degeneration of these joints

was described, we called the “X angle” [Figure 2]. Intra- and inter-observer reliability of this test was determined. The instruments of QOL and disability were EQ5D and Oswestry disability index (ODI), respectively. Osteoarthritis changes in C0-C1-C2 and EQ5D and ODI were analyzed using the Pearson’s correlation coefficient and ANOVA test. Intra- and inter-class reliabilities were evaluated by interclass correlation coefficient (ICC).

RESULTS

Eighty-six patients with AS treated surgically for cervical fracture had complete facet ankylosis between C3 and T1 due to their pathology [Figure 3]. Cervical 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.*^[13] All fractures were hyperextension injuries and classified type B4 M2 according to the new AOSpine classification.^[4] Neurological status was classified by Frankel scale having 11 patients Frankel A, 3 Frankel B, 6 Frankel C, 3 Frankel D, and 64 Frankel E. The occurrence of epidural hematoma was not assessed in all patients because MRI was only performed in neurologically deteriorated patients. Patients were treated by posterior stabilization from upper cervical to upper thoracic spine using titanium screw-rod systems. Screws were inserted 3 vertebrae cranially and 3 vertebrae caudally of the injury. If extending to C1, Goel–Harm’s screw were placed, in C2 short, nontransarticular Magerl screws were placed, subaxially, in most cases lateral mass screws were used, whereas in the thoracic spine, pedicular screws were placed. In 8 patients (9%), a combined posterior-anterior approach was performed.

The mean age was 69.2 years (standard deviation [SD]: 11.7). The most common level of fracture was in C5-C6 [Figure 3]. Demographic data is shown in [Table 2].

In 24 patients, the C0–C1 joint was fused, and in 15 patients, C1–C2 joint was fused. The intra- and inter-class reliabilities

Table 1: Complications reported in the literature

	Complication rate (range)	Reported in citation
Neurologic impairment	16%-91%	1, 3, 8, 10
Epidural haematoma	8%-23%	6, 9
2-year mortality	3%-26%	2, 3, 7, 8
Implant failure	8%-73%	8, 11, 12
Wound infection	5%-7%	8, 12
Halo-related complications	7%	8
Respiratory failure	8%-14%	3, 8
Pneumonia	6%-14%	3, 8
Tracheostomy	7%-16%	8, 11
Delirium	21%	8
Decubital ulcers	7%	8
Other	9%-36%	3, 8, 11

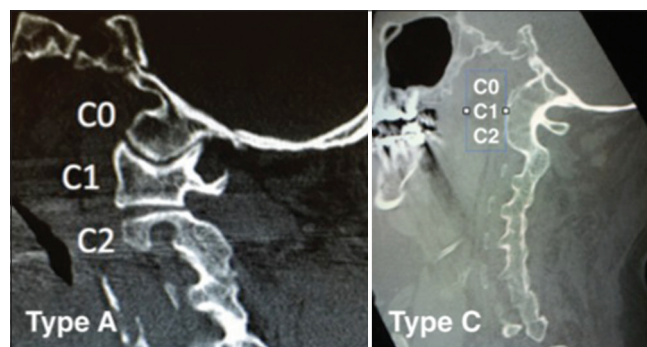


Figure 1: Types of occipito-atlanto-axial articulations in AS patients. From Type A, normal C0–C1–C2, to Type C, complete C0–C1–C2 ankylosis

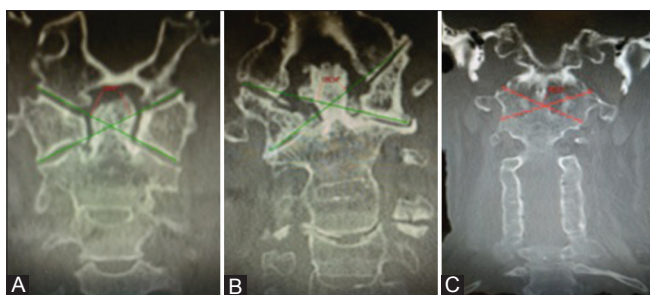


Figure 2: “X-angle”: Measured in the coronal section of the computed tomography-scan, 2 lines are drawn between the joints of C0–C1 and C1–C2 forming an X on the odontoid. Type A: Healthy articulation; Type B: Osteoarthritic changes; and Type C: Ankylosis. The mean value for healthy articulations (Type A) was 125° and that of ankylosed (Type C) 135° due to facet joints collapse

(ICC *r*) for X-angle measurement were very high: intrarater reliability: 0.94 (confidence interval [CI] 95%: 0.90–0.96) (*P* < 0.001), interrater reliability: 0.92 (CI 95%: 0.88–0.95) (*P* < 0.001).

The mean X-angle was 125° (SD: 12) in nonfused patients and 136° (SD: 14) in fused patients (*P* < 0.001) [Figure 2]. There were no differences in QOL and disability at 1 and 2 years between the two groups: EQ5D-index of 0.54 and 0.55 (*P* = 0.5), ODI of 26.4 and 24 (*P* = 0.35), respectively [Table 3].

DISCUSSION

Spinal fractures in AS patients usually result from low-energy trauma and hyperextension mechanism. Westerveld *et al.* reported that they are frequently admitted with a neurologic deficit 67.2% with secondary deterioration in 13.9%. In AS patients, complications and mortality rates were significantly higher than controls.^[8]

Complications caused by the fracture itself and the treatment are summarised in Table 1. Treatment options are from halo-jacket, anterior, posterior and double approaches, with long posterior contracts showing the treatment of choice due to the best biomechanical stability and less complications.^[2,5] Abnormalities of the cervical segment can be debilitating and induce adverse effects on the overall functioning and QOL of the patient. Furthermore, the cervical spine plays a pivotal role in influencing subjacent global spinal alignment and pelvic tilt as compensatory changes occur to maintain horizontal gaze.^[14] Because the cervical spine is the most mobile part of the spinal column, a wide range of normal alignment has been described.^[15,16] In asymptomatic normal volunteers, a large percentage (approximately 75%–80%) of cervical standing lordosis is localized to C1–C2 and relatively little lordosis exists in the lower cervical levels.^[14] In our

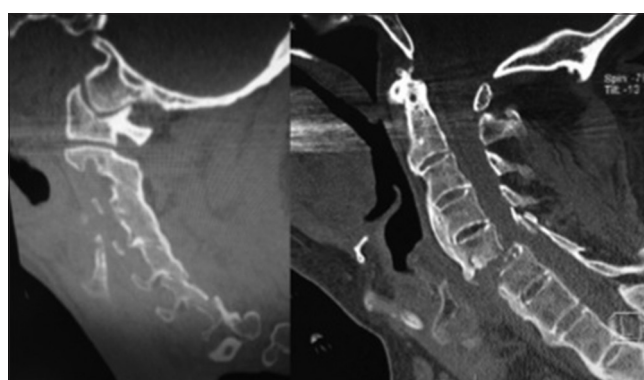


Figure 3: All patients had subaxial ankylosis (from C3 to T1). The most frequently affected area was C5–C6

Table 2: Demographic data

Demographic characteristics	Value
Patient number	86
Male	67 (78%)
Age	69 ± 12 years
Weight	75 ± 12 kg
Height	169 ± 8 cm
Level of fracture	
C2	4 (5%)
C3	2 (2%)
C4	7 (8%)
C5	14 (16%)
C6	33 (38%)
C7	26 (30%)
Frankel score	
A	11 (13%)
B	3 (4%)
C	6 (7%)
D	2 (2%)
E	64 (74%)

Table 3: Health-related quality of life (EQ5D) and disability (ODI) from baseline to 2 years after fracture

	<i>n</i>	EQ-5D 0 y	EQ-5D 1 y	EQ-5D 2 y	ODI 0 y	ODI 1 y	ODI 2 y
C-type	39	0.68	0.60	0.54	27	28	26
AB-type	47	0.36	0.55	0.55	44	30	24

cohort group, all the 86 operated patients had a complete facet ankylosis from C3 to T1 due to the AS, 24 of them also had a fused C0–C1 joint and in 15 patients C1–C2 joint was fused. We did not found any differences in the QOL (EQ5D) and disability (ODI) at 2 years between the group that had C0–C1–C2 free and the group that had ankylosis at C0–C1/ C1–C2 or both articulations.

CONCLUSIONS

X-angle is a reliable way to measure joint integrity C0–C1–C2 in patients with AS. In AS patients operated of a cervical

fracture, total cervical ankylosis including the C0–C1–C2 segments is not related to poorer QOL and disability.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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