



# Management of cervical fractures in ankylosing spondylitis: anterior, posterior or combined approach?

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## Abstract

**Introduction:** Ankylosing spondylitis (AS) can lead to an increased risk of cervical fractures.

**Sources of data:** A systematic review was undertaken using the keywords 'ankylosing spondylitis', 'spine fractures', 'cervical fractures', 'surgery' and 'postoperative outcomes' on Medline, Pubmed, Google Scholar, Ovid and Embase, and the quality of the studies included was evaluated according to the Coleman Methodology Score.

**Areas of agreement:** Surgery ameliorates neurological function in patients with unstable AS-related cervical fractures. The combined anterior/posterior and the posterior approaches are more effective than the anterior approach.

**Areas of controversy:** The optimal approach, anterior, posterior or combined anterior/posterior, for the management of AS related cervical fractures has not been defined.

**Growing points:** Open reduction and internal fixation allows avoiding worsening and enhances neurological function in AS patients with cervical fractures.

**Areas timely for developing research:** Adequately powered randomized trials with appropriate subjective and objective outcome measures are necessary to reach definitive conclusions.

**Key words:** ankylosing spondylitis, spine fractures, cervical fractures, surgery, management, postoperative outcomes

## Introduction

Ankylosing spondylitis (AS) is an autoimmune disease, involving spine, sacro-iliac joints and entheses.<sup>1,2</sup> Chronic inflammation determines degeneration of affected joints leading to fusion and ankylosis,<sup>3</sup> associated with pain and progressive joint stiffness. Severe AS, affecting the whole spine, leads to an increased risk of vertebral fractures.<sup>4</sup> The stiffened spine is not able to bear normal loads in comparison with a healthy flexible spine.<sup>5</sup> In addition, patients with AS develop marked bone mineral density loss in the early stages of the disease.<sup>6-9</sup>

The cervical spine, followed by the thoracic spine, is the most common site of fracture.<sup>1,10,11</sup> The diagnosis of cervical fractures is frequently delayed, because the clinical symptoms may not be severe.<sup>1,10,12,13</sup> Standard imaging is inadequate to detect osteoporosis-related shearing fractures, especially in the spine.<sup>7,14</sup> The comprehensive study of the spine using computer tomography (CT) and magnetic resonance (MR) is recommended even when a fracture is only suspected.<sup>7,15-17</sup>

Neurological deficits can be developed either at the time of the vertebral fracture or when it displaces, particularly in hyperextension injuries.<sup>18</sup>

Surgical management is currently indicated in patients with unstable vertebral fractures and progressive neurological involvement.<sup>15,16,19</sup> Surgery aims to avoid complications associated with conservative treatment, especially pin site infection and secondary loss of correction with neurological worsening.<sup>9,20-22</sup> Although several techniques have been described, the best procedure to manage patients with AS-related cervical fractures is still debated.

This systematic review reports the clinical outcomes and complications rate of anterior (AA), posterior (PA) and combined anterior/posterior (CA) approaches for the management of cervical fractures in patients with AS.

## Materials and methods

### Research protocol and literature search

Three authors performed a blind literature search in Medline, Pubmed, Google Scholar, Ovid and Embase databases using ‘ankylosing spondylitis’,

‘spine fractures’, ‘cervical fractures’, ‘surgery’ and ‘postoperative outcomes’ as keywords. The literature search was performed on October 15, 2014, with no limit based on the year of publication. All the studies published in Italian, English, Spanish and French were considered, given the language skills of the authors.

Eight hundred and twenty publications were identified. All the three reviewers evaluated the abstract and the bibliography of the collected studies to identify further relevant articles. Only papers published in peer-reviewed journals were considered eligible for inclusion; the articles had to report postoperative outcomes of patients with AS and concomitant cervical fractures managed by surgery.

Finally, all the included studies were reviewed, analyzed and discussed by all the authors to minimize selection bias.

### Inclusion and exclusion criteria

The inclusion criteria were: cervical spinal fracture occurring in patient with AS; imaging demonstrating the cervical fracture; clinical examination associated or not to a Frankel neurological score; detailed description of the neurological status; details of surgery; length of follow-up period; report of any peri/postoperative complication associated with surgery; statistical analysis of postoperative outcomes.

Technical notes, letters to editors, case reports, and narrative and quantitative reviews were excluded. Studies on conservative management of cervical fractures or surgical management of thoracic and lumbar fractures were also excluded.

Studies missing data about preoperative diagnosis, surgery, follow-up, imaging assessment of fracture, clinical examination, clinical outcomes and postoperative imaging were excluded. Patients included in the selected articles without a sufficient time of follow-up (1 month) were excluded from the study group.

### Quality assessment and data extraction

The quality of the included studies was evaluated according to the Coleman Methodology Score (CMS). The CMS includes 10 domains with a total score from 0 to 100 points. The quality was considered excellent for CMS values between 85 and 100 points,

good for values between 70 and 84 points, fair for values between 50 and 69 points, and poor for values under 50 points. The subsections of the CMS are designed on the basis of the CONSORT statement.<sup>23</sup>

Coleman criteria were modified to be reproducible and relevant for the present study investigating the surgical management of AS-related fractures of the cervical spine. Three reviewers scored independently all the studies to give a total CMS.

The data extracted from the articles are listed in Table 1. Outcomes of different procedures were evaluated with Frankel neurological score, imaging assessment and clinical examination. We also evaluated the percentage of fusion of the implants with each surgical procedure by imaging investigation.

Statistical analysis

Categorical variables were expressed as frequency with percentage. Continuous variables were expressed as average value with range. The comparison of the mean value of CMS calculated by the three examiners was performed with Student's *t*-test. A *P*-value under 0.05 was considered statistically significant. All statistical analyses were conducted with the SPSS 16.0 version.

Results

Initial database searches retrieved 820 studies. Eight articles<sup>5,7,9,24-28</sup> were considered eligible for the

study, published from 1990 to 2011. All the studies reported clinical and imaging outcomes after anterior, posterior or combined anterior and posterior surgery to treat cervical fractures in patients with AS.

Quality assessment

The mean value of the CMS score was 37.9 ± 12.8 points (range from 20<sup>27</sup> to 55<sup>28</sup>) (Table 2). No statistically significant difference was found between mean values of CMS calculated by the three examiners.

Demographics

The patients included were 110,<sup>5,7,9,24-28</sup> 103 (94%) males and 7 (6%) females, with an age from 32<sup>24</sup> to 82<sup>7</sup> years. They were assessed with a mean follow-up period of 21.9 months (range 1<sup>7</sup>–120<sup>5</sup> months). At the time of the index surgery, the age was 59.2 years (range 32<sup>24</sup>–82<sup>7</sup> years). The average age of women was 70.3 years (range 50–81<sup>7</sup> years), whereas the average age of men was 58.4 (range 32<sup>24</sup>–82<sup>7</sup> years). The male/total ratio was 0.93. No patients had previous operations in the cervical spine.

Diagnosis

The mechanism of injury was reported in 86 (78%) of 110 patients with a cervical fracture.<sup>5,7,9,24-28</sup> A low-energy trauma occurred in 76 (85%) patients,<sup>5,7,9,24-28</sup>

Table 1 Data extracted from the included articles

Data	Definition
Author, Journal and year of publication	
Study design	Prospective, retrospective, case study
Follow-up	Months and years
Mean age	Years
Coleman score	20–55 (RANGE)
Fracture level	Cervical
Type of treatment	Surgical: anterior procedure, posterior procedure, combined anterior and posterior procedure
Neurological status	Frankel score, clinical examination
Immobilization	Halo, cervical collar, SOMI, Minerva Philadelphia
Complications	Neurologic deterioration, epidural hematoma, DVT, infection, pneumonia, pulmonary embolism, ARDS, pulmonary insufficiency, lung edema, cerebral ischemia, intestinal obstruction and esophageal fistula.

**Table 2** Modified Coleman Methodology score

Items	Score (mean $\pm$ SD)	Range
Study size number of patient	1.5 $\pm$ 2.1	0–4
Mean follow-up	2.9 $\pm$ 1.9	0–5
Number of different surgical procedures	3.8 $\pm$ 5.2	0–10
Type of the study	3.8 $\pm$ 5.2	0–10
Diagnostic certain	5	5
Description of postoperative rehabilitation	3.4 $\pm$ 1.7	0–5
Outcome criteria	6.3 $\pm$ 5.2	0–10
Procedure for assessing outcomes	2.8 $\pm$ 0.5	2–3
Description of subject selection process	4.3 $\pm$ 1.1	3–5
Description of assessing outcomes	4.4 $\pm$ 1.8	0–5
Total Coleman score	37.9 $\pm$ 12.8	20–55

while a high energy trauma, including motor vehicle accident and fall from a height, was reported in only 10 (15%) patients.<sup>5,9,27</sup>

The concomitant intervertebral disc (IVD) or vertebral dislocation was evaluated in seven studies<sup>5,7,9,24–26,28</sup> reporting on 94 patients. Dislocation was confirmed with imaging assessment or during surgery, and it was reported in 68 (72%) of 94 patients,<sup>5,7,9,24–26,28</sup> while in 26 (28%) of 94 patients,<sup>7,24,26,28</sup> it was not specified.

The most common level of fracture was C6–C7, reported in 44 (40%) of 110 patients,<sup>5,7,9,24–26,28</sup> followed by C5–C6 in 21 (19%),<sup>7,9,24,26</sup> C4–C5<sup>7,25,26</sup> and C6<sup>24,27</sup> in 8 patients (7%), respectively. There were also four cases (4%) of C7 fracture,<sup>24,27</sup> and three cases (3%) of C6–C7 and C7–T1 combined fractures<sup>7</sup> such as C2 fracture.<sup>5,27</sup> Fractures of C7–T1 level or combined C5–C6 and C6–C7 or C4 and C2–C3 level were respectively reported in two patients (2%). Finally, fractures of C1<sup>27</sup> or C3<sup>27</sup> or multilevel C4–C7<sup>24</sup> or combined C4–C5 and C5–C6<sup>7</sup> were found in only one patient each.

### Surgical management and postoperative rehabilitation

Among the included studies, cervical fractures were managed with different surgical approaches. The AA was performed in 14 (13%) of 110 patients,<sup>7,27</sup> the PA in 34 (31%)<sup>5,7,9,24</sup> and a CA in the remaining 62 (56%).<sup>5,7,24–27</sup> The type of osteosynthesis performed

by each author is given in Table 3. The surgical procedure was adequately described in seven (88%) of eight selected studies, scoring the maximum CMS of 5 points in three (38%) studies,<sup>9,26,28</sup> and 3 points in four (50%) studies.<sup>5,7,24,25</sup> Postoperative rehabilitation was adequately described in five (62%) of eight studies, with the maximum CMS of 10 points,<sup>5,9,24,25,28</sup> whereas no description was found in three (38%) studies.<sup>7,26,27</sup>

### Patient selection and outcome assessment

Seven (88%) of the eight selected studies<sup>5,7,9,24–26,28</sup> reported clear selection criteria of patients, and all studies provided an outcome assessment by using validated scoring systems or clinical neurological examination, with good reliability and sensitivity.

Pre- and post-operative neurological functions were evaluated in all the included studies. The Frankel score was performed in 64 (58%) of 110 patients,<sup>5,7,25,27</sup> whereas a clinical neurological examination was used in the remaining 46 (42%) patients.<sup>9,24,26,28</sup>

In this population of 64 patients, the changes of Frankel score were evaluated independently by surgical procedure.<sup>5,7,25,27</sup> After surgery, the number of patients with Frankel A, B and C significantly decreased, whereas the number of patients with Frankel E increased (Table 4). The change of the Frankel score from pre-operatively to postoperatively according to each surgical procedure performed is reported in Tables 5 and 6.

**Table 3** Osteosynthesis and surgical approach

Author	Osteosynthesis	Approach
Cornefjord <i>et al.</i> <sup>24</sup>	Olerud Cervical Fixation System (OC), with cervical pedicle screws and rods; limited-contact-dynamic-compression plate (LCDCP) for fracture stabilization (Stratec Medical, Switzerland)	Posterior
Einsiedel <i>et al.</i> <sup>7</sup>	(CSLP cervical spine locking plate, Stratec Medical, Switzerland).	Anterior
	Wolter plate fixators (Litos), normal 1/3 tubular plates (AO/ASIF; Synthes/Zimmer), and flexible internal fixing systems (the NEON device [Ulrich, Ulm, Germany] and Cervifix [Synthes/Zimmer]).	Posterior
	Morscher plates (titanium-hollow-locking screw plate; Synthes/Zimmer, Solothurn, Switzerland) (locking screws/angle-stable screws were used after 2002) and Wolter plates (Litos, Hamburg, Germany)	Anterior
Sapkas <i>et al.</i> <sup>5</sup>	Not specified	Anterior/posterior
Lv <i>et al.</i> <sup>26</sup>	Not specified	Anterior/posterior
Olerud <i>et al.</i> <sup>27</sup>	Posteriorly reconstruction plates; In the C1–C2 region dens screws or facet joint screws were used, sometimes in combination with posterior cervico-occipital fusion	Posterior
Payer <sup>28</sup>	AO cervical spine locking plate	Anterior
	Posterior lateral mass and pedicle fixation; corpectomy and cage reconstruction or iliac crest autograft and a plate	Anterior/posterior
Duhem-Tonnelle <i>et al.</i> <sup>25</sup>	Not specified	Anterior/posterior
Taggard and Traynelis <sup>9</sup>	Lateral mass plating; rib graft harvested as described by Sawin and Traynelis and sized to the length of the instrumented levels. The rib graft was fixed to the lamina and spinous processes using titanium cable.	Posterior

Imaging assessment of fracture

Preoperative standardized plain radiographs were performed in 108 of 110 (98%) patients as the first imaging investigation.<sup>5,7,9,24–28</sup> MRI and CT scan were performed in 57 (52%)<sup>5,7,9,26</sup> and 40 (36%) patients,<sup>7,9,25,28</sup> respectively.

After surgery, healing of the fracture was assessed with imaging investigations in 73 (66%) of 110 patients.<sup>5,24–28</sup> Sixty-two (85%) of these patients received a radiographic evaluation,<sup>24,26–28</sup> while 11 (15%) of them were monitored with CT scan.<sup>5,25</sup> In all patients who had undergone imaging investigation healing of fracture was demonstrated.<sup>5,24–28</sup>

Complications

Post-surgical medical complications were recorded in 21 (19%) of 110 patients,<sup>5,7,24,26–28</sup> while intra-operative complications were recorded in 18 (16%) of 110 patients. Fifteen deaths occurred after a minimum of 2 months after surgery (Table 7). In the

**Table 4** Pre-operative and post-operative grades of Frankel score

Frankel grade	Preoperative	Postoperative
A	20% ( <i>n</i> = 13)	5% ( <i>n</i> = 3)
B	19% ( <i>n</i> = 12)	5% ( <i>n</i> = 3)
C	25% ( <i>n</i> = 16)	11% ( <i>n</i> = 7)
D	25% ( <i>n</i> = 16)	23% ( <i>n</i> = 15)
E	11% ( <i>n</i> = 7)	56% ( <i>n</i> = 36)
Tot. patients	100% ( <i>n</i> = 64)	100% ( <i>n</i> = 64)

subset of 62 patients who had undergone CA, post-surgical medical complications were reported in 10 (16%) patients, and in 7 of them death occurred after a minimum period of 3 months. Intra-operative complications were found in 11 (18%) patients. In the subset of 14 patients who had undergone AA, 2 (14%) developed post-surgical medical complications, and 2 (14%) intra-operative complications. Death occurred in three (21%) cases, two of them linked with the recorded post-surgical complications.

**Table 5** Pre-operative and post-operative grades of Frankel score for each surgical procedures

Surgery	Frankel grade	Preoperative	Postoperative
Combined approach	A	22% ( <i>n</i> = 9)	5% ( <i>n</i> = 2)
	B	13% ( <i>n</i> = 5)	2% ( <i>n</i> = 1)
	C	25% ( <i>n</i> = 10)	8% ( <i>n</i> = 3)
	D	30% ( <i>n</i> = 12)	25% ( <i>n</i> = 10)
	E	10% ( <i>n</i> = 4)	60% ( <i>n</i> = 24)
	Tot. patients	100% ( <i>n</i> = 40)	100% ( <i>n</i> = 40)
Anterior approach	A	21% ( <i>n</i> = 3)	0% ( <i>n</i> = 0)
	B	36% ( <i>n</i> = 5)	14% ( <i>n</i> = 2)
	C	29% ( <i>n</i> = 4)	21% ( <i>n</i> = 3)
	D	7% ( <i>n</i> = 1)	21% ( <i>n</i> = 3)
	E	7% ( <i>n</i> = 1)	43% ( <i>n</i> = 6)
	Tot. patients	100% ( <i>n</i> = 14)	100% ( <i>n</i> = 14)
Posterior approach	A	10% ( <i>n</i> = 1)	10% ( <i>n</i> = 1)
	B	20% ( <i>n</i> = 2)	0% ( <i>n</i> = 0)
	C	20% ( <i>n</i> = 2)	10% ( <i>n</i> = 1)
	D	30% ( <i>n</i> = 3)	20% ( <i>n</i> = 2)
	E	20% ( <i>n</i> = 2)	60% ( <i>n</i> = 6)
	Tot. patients	100% ( <i>n</i> = 10)	100% ( <i>n</i> = 10)

Finally, in the subset of 34 patients managed with PA, post-surgical medical complications were reported in nine (26%) patients, and in five of them death occurred. Intra-operative complications were found in five (15%) individuals.

## Discussion

In the present study AA, PA or CA procedures were considered to ascertain which provided the best surgical outcome in terms of effectiveness and safety.

The mean age of the patients included was lower than the mean age of the population of patients with cervical fractures, confirming the early onset of spine injuries in AS. Moreover, the mean age of men affected by AS-related fractures is significantly lower than those of women (58.4 years vs. 70.3 years), showing that cervical fractures occur earlier in men than in women.

In our study, the most frequently injured levels were C6-C7, followed by C5-C6, with >75% of all fractures involving C5, C6 and C7. Moreover, >70% of fractures were accompanied by an IVD or vertebral structure dislocation. In a healthy spine, the part most susceptible to traumatic injuries is the

cervical spine, because of small vertebral bodies, increased mobility, oblique articular facets and the mobility of the heavy skull on the cervical column.<sup>29,30</sup> When fusion of the spine occurs, as in AS, fractures usually occur adjacent to the fused spine and/or at the junction of mobile and fused spine.<sup>31</sup> For this reason, we can hypothesize that the cervico-thoracic junction has a higher risk to develop injuries if the ankylosis occurs.<sup>32</sup> On the other hand, the association between cervical fracture and IVD dislocation can be explained by the pathogenesis of AS, characterized by progressive degeneration of the IVD, with chondroid metaplasia, calcification of annulus fibrosus and nucleus pulposus leading to loss of elasticity and stability of the spine.<sup>33,34</sup>

The mechanism of injury was a low-energy trauma on the cervical spine in 85% of patients. Despite its features, this trauma often causes dislocation of the vertebral structures or IVD, leading to the unstable configuration of cervical fractures in patients with AS.

In the studies included, plain radiographs of the spine were performed in 98% of patients as the first imaging investigation. However, standard radiographs can be difficult to interpret in subjects with

Table 6 Improvement of Frankel score according to different surgical managements

Surgery	Frankel +1	Frankel +2	Frankel +3	Frankel +4	NO (Frankel: A-B-C-D)	NO (Frankel E)	Tot. patients
Combined approach	43% (n = 17)	23% (n = 9)	12% (n = 5)	5% (n = 2)	7% (n = 3)	10% (n = 4)	100% (n = 40)
Anterior approach	29% (n = 4)	29% (n = 4)	21% (n = 3)	0% (n = 0)	14% (n = 2)	7% (n = 1)	100% (n = 14)
Posterior approach	50% (n = 5)	20% (n = 2)	0% (n = 0)	0% (n = 0)	10% (n = 1)	20% (n = 2)	100% (n = 10)

Table 7 Complications

Complications	No. of patients (%)
Intraoperative	
Loosening of the implant	12 (11)
Epidural hematoma	2 (2)
Excessive intra-operative bleeding	2 (2)
Cerebrofluid leakage	1 (1)
Spinal process deformity	1 (1)
Post-surgical	
Pneumonia	5 (5)
Infection	4 (4)
ARDS	2 (2)
Deformity	2 (2)
Hoarseness	1 (1)
Transient swallowing difficulties	1 (1)
Cerebro-fluid leakage	1 (1)
Deep venous thrombosis	1 (1)
Cerebral ischemia	1 (1)
Pulmonary embolism	1 (1)
Intestinal obstruction	1 (1)
Death	
Infection	13 (87)
Cerebral ischemia	1 (7)
Deep venous thrombosis	1 (7)

AS, because of pre-existing pathologic bony changes, and ligamentous and discal calcification.<sup>35</sup> These features can lead to delay or miss the diagnosis of cervical fracture.<sup>5,7,36</sup> CT scan allows to detect the site of fracture and to assess fracture characteristics, particularly in patients with multiple spinal fractures or cervical spinal fractures. MRI is mandatory to detect spinal cord or nerve root injuries and epidural hematoma.<sup>35</sup>

All patients underwent clinical examination for the assessment of neurological function before surgery, and >90% of them had a cervical fracture associated with a clinical neurological impairment. Sixty-four (58%) of 110 included patients were also assessed with Frankel score. The preoperative evaluation showed that 39% of patients were Frankel A or Frankel B, 50% were Frankel C or D, and only 11% were Frankel E with normal neurology. These findings demonstrate that cervical fractures, in patients with AS, are severe events leading to marked neurological deficits. In these patients, neurological complications can arise from the level of vertebral



fracture, but also secondary events. Indeed, AS carries a higher risk to develop a secondary neurological impairment because of unstable fracture configuration occurring between the fused segments.<sup>37</sup>

In the included studies, cervical fractures were managed with different surgical approaches, including the AA in 14 (13%),<sup>7,27</sup> the PA in 34 (31%)<sup>5,7,9,24</sup> and the CA in 62 (56%)<sup>5,7,24–27</sup> of 110 patients.

The subpopulation of 64 patients assessed with Frankel score<sup>5,7,25,27</sup> showed a significant decrease of patients with Frankel A, B and C, and a significant increase of patients with Frankel E, independent of the surgical procedure performed. Similar results were found with the evaluation of Frankel score changes in each subset of patients who underwent different surgical procedures. All these findings support the critical role of surgery to improve neurological function after cervical spine fractures. However, CA and PA provide a higher rate of neurological improvement when compared with AA.

In terms of intra-operative and post-surgical complications, the safety of the different operative techniques was comparable. On the other hand, the post-surgical complication rate was significantly higher for PA when compared with AA (14 vs. 26%,  $P = 0.03$ ).

However, the isolated AA, as previously reported,<sup>38,39</sup> is frequently associated with screw and implant failure or displacements, which more often require revision surgery.

Although some deaths after surgery were reported, particularly in older patients, the mortality rate of surgery is lower than conservative management for patients with AS and concomitant cervical fractures.<sup>20–22,32,33</sup> Death usually occurred in patients with postoperative complications, particularly when the respiratory function was affected, as in pneumonia, pulmonary fibrosis, pulmonary embolism and ARDS.

One limitation of this article is its nature, and also that this systematic review included a small number of studies. However, it reflects the lack of a standardized surgical procedure for the management of traumatic cervical fractures in ankylosed spines. A further limitation is the poor quality of the included studies. Only three studies had a total CMS between

50 and 60 points. Furthermore, the studies did not report crucial information such as the type of fractures and displacement, and the specific type of osteosynthesis performed. We are aware that this could be considered a major weakness of the study, but unfortunately no adequately powered randomized trials have been performed on this topic.

## Conclusion

Surgical management consisting of open reduction and internal fixation allows to avoid worsening and to enhance improvement of neurological function in patients with AS with cervical spine fractures. Although each operative technique provides improvement in clinical outcomes, the combined anterior-posterior approach and the posterior approach are more effective than the anterior approach. The anterior approach is associated with higher risk of implant failure and subsequent revision surgery. However, adequately powered randomized trials with appropriate subjective and objective outcome measures are required to define the best surgical procedure for these patients.

## Conflict of Interest statement

The authors have no potential conflicts of interest.

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