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Surgical Outcomes of Subaxial Cervical Fractures in Patients with Ankylosing Spinal Disorder

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ABSTRACT

AIM: To evaluate the patient groups with ankylosing spinal disorder (ASD) in terms of patients' characteristics, applied surgical approaches, and the outcomes.

MATERIAL and METHODS: Between 2008-2019, 50 ASD patients (35–80 years) were operated on subaxial cervical fracture. The mean follow-up time was 48 months. According to the surgical approach, the patients were divided into three groups: patients who only underwent anterior fusion [AF], patients who only underwent posterior fusion [PF], and patients who underwent anteroposterior fusion [APF]. In this retrospective study, we examined the patients' files and outpatient checks to evaluate the history, operations, neurological results, and complications in cases.

RESULTS: After undergoing respective surgical interventions, 1 of the 7 patients in the AF group (14%), 2 of the 18 patients in the PF group (18%), and 3 of the 25 patients in the APF group (12%) died. The postoperative American Spinal Injury Association scores were statistically better in all groups than in the preoperative scores. Among the surgical interventions, improvement in the APF group was significantly better than in other groups.

CONCLUSION: Although there is a higher amount of surgery related complications in the APF group, the biomechanical and clinical results are better than the other two surgical interventions.

KEYWORDS: Ankylosing spinal disorder, Anterior fusion, Anteroposterior fusion, Cervical spine, Posterior fusion, Subaxial cervical fractures, Spinal Trauma

ABBREVIATIONS: AF: Anterior fusion, **APF:** Anteroposterior fusion, **AS:** Ankylosing spondylitis, **ASD:** Ankylosing spinal disorder, **ASIA:** American Spinal Injury Association, **CT:** Computed tomography, **DISH:** Diffuse idiopathic skeletal hyperostosis, **MRI:** Magnetic resonance imaging, **PF:** Posterior fusion, **F:** Female, **Inf:** Infection, **M:** Male

INTRODUCTION

Ankylosing spinal disorders (ASD) are a disease group that advances with the progressive ossification of the spinal column (3). The most common conditions observed in ASDs are ankylosing spondylitis (AS) and diffuse idiopathic skeletal hyperostosis (DISH). AS is also known as rheumatic spondyloarthropathy that is seronegative, progressive, and inflammatory and mainly affects the sacroiliac joints and spine (7). As a result, the patient develops kyphosis with a hard spine, which creates a high risk factor for spinal fracture. The risk of life-long vertebral fractures in patients with AS is three to five times higher than the general population (8). The prevalence

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is between 0.1 and 1.4%, and it is twice more common in males than females (7). Approximately 75% of the fractures are observed in the cervical spine, and this is followed by the thoracic and lumbar spine (6,14,23,26,29).

The reason it is observed more commonly in the cervical spine is explained by hypermobility of the cervical spine, weight of the skull, small vertebral bodies, and location of cervical facet joints (4,20,32,33,36). The diagnosis of cervical fractures is often delayed because clinical symptoms may not be severe (25,35). The delay is partially caused by the rarity of pain related to these fractures. Because patients have been receiving corticosteroid therapy for pre-existing spinal changes and AS (7).

AS-related fractures are often more serious than cervical fractures in the healthy population. It is highly instable due to the involvement of anterior and posterior structures in the fracture. In addition, the fracture in a bamboo-like spine structure that preserves the long level is extremely problematic with a high risk of neurological deterioration. In addition, kyphotic deformation does not provide the appropriate sagittal angle for primary stabilization. Despite these negative conditions, these bones show a good tendency for fusion (2,16).

Radiographs have been the key in the AS classification as well as a gold standard for the assessment of deformity. However, AS spinal fracture radiographs are difficult to interpret because of associated syndesmophytes, abnormal bone density, and deformity. Junctional areas, such as cervicothoracic spine, appear very weak due to associated kyphosis that makes them difficult to interpret (1,11). Fractures in the AS are, sometimes, observed as a cavity in the related bone continuity with a sharp or thin change in the spinal structure in the fractured area. These fractures can also be associated with osteolysis. High-resolution multidetector computed tomography (CT) is a reliable imaging method to identify the fine fracture lines as well as associated structural damages and erosions. Magnetic resonance imaging (MRI) scans are essential in the evaluation of soft tissue and spinal cord damage.

Although various techniques have been described in the research literature, the best method for patients with cervical fractures with AS is still under discussion. The purpose of this study is to evaluate the patient groups with ASD in terms of age, sex, surgical approaches, and their results.

MATERIAL and METHODS

In this study, the patients with ASD and subaxial cervical spine fractures were examined between the years 2008 and 2018. The data in this study were retrospectively obtained from the patient's files and outpatient records. MRI, CT, and plain radiographs were used as the imaging methods for the evaluation of patients. The patients with incomplete medical records were excluded from the study. In total, 50 patients with ASD were included in this study. According to the surgical approach, the patients were divided into three groups: patients who only underwent anterior surgery (anterior fusion [AF] group), patients who only underwent posterior surgery (posterior fusion [PF] group), and patients who only underwent a combined anterior and posterior surgery (anteroposterior fusion [APF] group). The type of surgical approach was mainly determined by the location and pattern of fractures, except in the following circumstances. Patients with conditions such as decreased lung capacity and/or overweight that are not appropriate for surgery in the prone position underwent AF surgery. However, patients with obesity and short neck and patients with excessively high rib cage underwent PF. APF was performed in the patients who did not carry the handicaps defined for both groups above. There were 7 patients in the AF group, 18 patients in the PF group, and 25 patients in the APF group (Table I). In the AF group, cages and plates were

Table I: Details of the Clinical Series

Case No	Age (in years)	Sex	AS	DISH	Fusion Types	Prepoerative ASIA Score	Postoperative 1st day/ Latest Follow-Up ASIA Score	Complications
1	37	M		+	APF	A	A (1 Mo)	Vertebral artery damage ->**Died**
2	39	M	+		PF	C	C / C (48 Mo)	
3	41	M	+		PF	C	C / C (48 Mo)	
4	43	M		+	APF	C	C / C (36 Mo)	Pulmonary infection
5	45	M	+		APF	C	D / D (60 Mo)	
6	47	M		+	AF	B	A (1 Mo)	Pulmonary inf->**Died**
7	49	M	+		APF	D	D / D (12 Mo)	
8	50	M	+		APF	D	D / D (48 Mo)	Pulmonary infection
9	51	M		+	APF	D	E / E (34 Mo)	
10	53	M	+		AF	E	D / E (120 Mo)	**Revision Surgery**
11	53	M	+		APF	D	D / E (110 Mo)	
12	56	M		+	PF	D	D / E (90 Mo)	Root injury
13	57	M	+		PF	D	D / D (52 Mo)	

Table I: Cont.

Case No	Age (in years)	Sex	AS	DISH	Fusion Types	Prepoerative Asia Score	Postoperative 1st day/ Latest Follow-Up Asia Score	Complications
14	58	M		+	PF	C	B / B (18 Mo)	
15	59	M	+		PF	C	B / B (48 Mo)	
16	62	M		+	PF	A	A (1 Mo)	Pulmonary inf ->**Died**
17	62	M	+		AF	D	D / D (48 Mo)	**Revision Surgery**
18	65	M		+	APF	D	D / E (64 Moo)	
19	65	M	+		PF	D	E / E (54 Mo)	
20	66	M	+		APF	D	D / D (48 Mo)	Pulmonary infection
21	66	M		+	APF	C	D / D (34 Mo)	Pulmonary infection
22	67	M	+		AF	E	B / B (24 Mo)	**Revision Surgery**
23	67	M	+		PF	E	D / E (88 Mo)	
24	68	M	+		AF	C	B / B (50 Mo)	
25	68	M	+		PF	C	D / D (48 Mo)	
26	69	M	+		APF	C	D / D (48 Mo)	
27	70	M		+	AF	C	C / C (10 Mo)	Esophagus rupture
28	71	M	+		APF	B	A (1 Mo)	Pulmonary inf->**Died**
29	71	M	+		PF	D	D / D (16 Mo)	Vertebral artery damage
30	72	M		+	PF	D	D / D (26 Mo)	Root injury
31	73	M	+		PF	D	E / E (34 Mo)	Pulmonary infection
32	74	M	+		APF	A	B / B (54 Mo)	
33	77	M		+	APF	A	B / B (52 Mo)	Pulmonary infection
34	80	M	+		APF	A	C / C (46 Mo)	
35	35	M		+	AF	C	D / D (58 Mo)	
36	41	F	+		PF	B	B / B (38 Mo)	Thromboembolic->**Died**
37	44	F	+		PF	C	D / D (38 Mo)	Root injury
38	47	F	+		PF	C	D / D (58 Mo)	
39	55	F	+		PF	E	D / E (48 Mo)	
40	57	F	+		PF	E	D / E (48 Mo)	
41	59	F		+	APF	E	E / E (48 Mo)	Vertebral artery damage
42	59	F	+		APF	B	B / B (48 Mo)	
43	61	F	+		APF	B	B / B (38 Mo)	Pulmonary infection
44	62	F	+		APF	B	B / B (48 Mo)	Pulmonary infection
45	62	F	+		APF	B	C / C (58 Mo)	Pulmonary infection
46	63	F	+		APF	A	B / B (95 Mo)	
47	65	F	+		APF	A	A (1 Mo)	Pulmonary inf->**Died**
48	65	F	+		APF	A	B / B (48 Mo)	
49	74	F		+	APF	A	B / B (38 Mo)	
50	75	F	+		APF	A	C / C (58 Mo)	Root injury

AF: *Anterior fusion,* ***APF:*** *Anteroposterior fusion,* ***AS:*** *Ankylosing spondylitis;* ***ASD:*** *Ankylosing spinal disorder,* ***CT:*** *Computed tomography,* ***DISH:*** *Diffuse idiopathic skeletal hyperostosis,* ***F:*** *Female,* ***Inf:*** *Infection,* ***M:*** *Male,* ***MRI:*** *Magnetic resonance imaging,* ***PF:*** *posterior fusion,* ***Mo:*** *Months.*

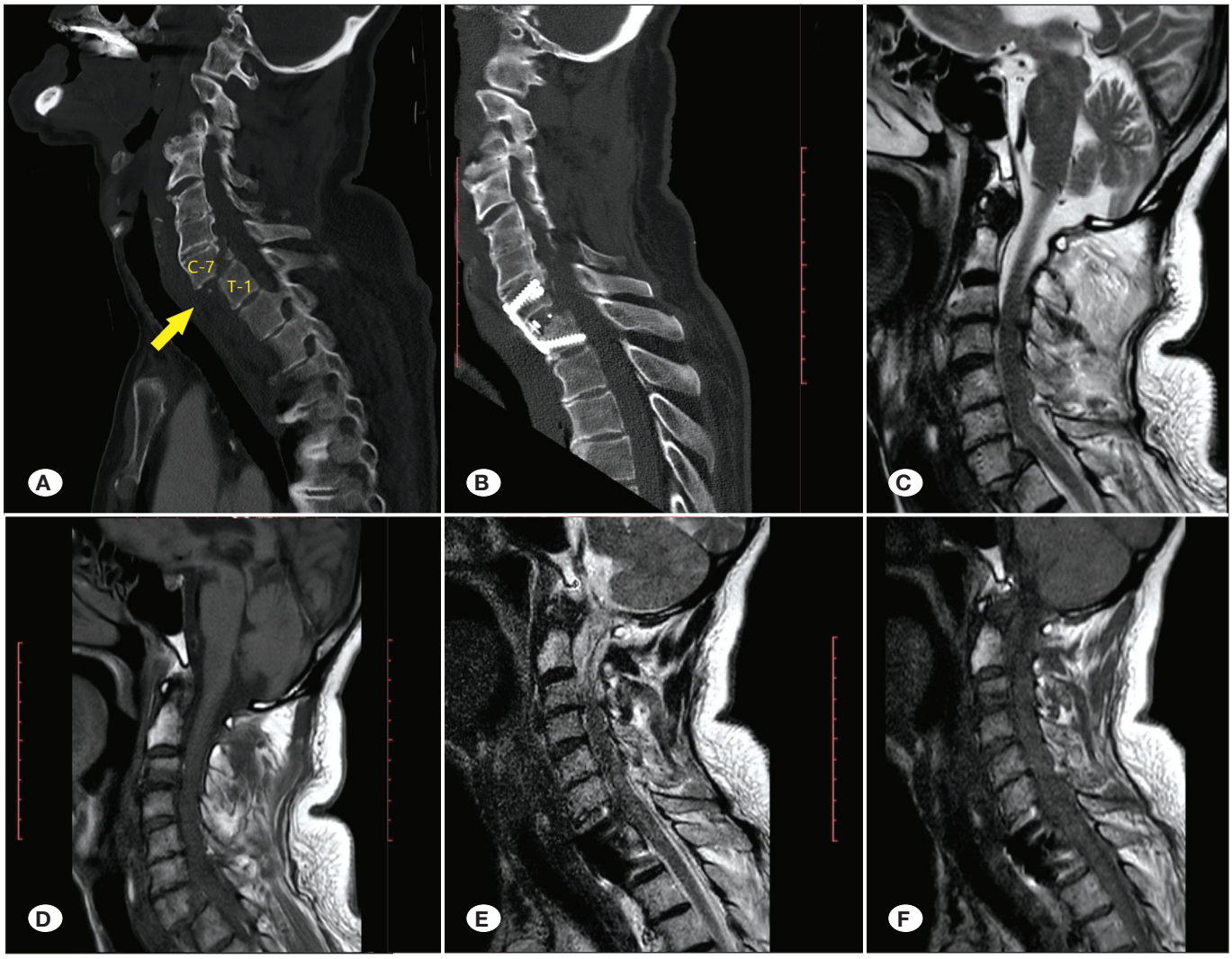

Figure 1: C7–T1 anterior dislocation + anterior fusion. **A)** Preoperative CT, **B)** postoperative CT, **C-D)** preoperative T2–T1 MRI, **E–F)** postoperative T2–T1 MRI. The yellow arrow indicates the fracture area. **CT:** Computed tomography, **MRI:** Magnetic resonance imaging.

employed to be fixed from the anterior region (Figure 1A-F). Transpedicular fixation was performed in all the patients of the PF group (Figure 2A-D). In the APF group, stabilization was performed first from the anterior region and then from the posterior region in the same session (Figure 3A-F).

The neurological status was evaluated according to the American Spinal Injury Association (ASIA) disorder scale. ASIA scores were taken preoperatively (neurological assessment at the time of arrival), postoperatively on day 1, and at the latest follow-up. These scores were taken separately for each case. The follow-up times ranged between 1 and 120 months **(mean: 48 months).**

Statistical Analysis

We used IBM SPSS 22 program for the statistical analysis in our study. Additionally, we employed Wilcoxon signed ranks test to compare the surgical treatment results. We compared the groups according to the Kruskal–Wallis test. The significance value for this study was considered as $p<0.05$. We used Mann–Whitney *U* test in the pairwise comparisons of groups (between AF and APF, between AF and PF, between patients with AS and DISH).

RESULTS

Among the 50 cases of cervical fracture (15 patients with DISH and 35 patients with AS), 40 of them were developed due to acute and high-energy trauma, 7 were developed during low-energy chronic process, and 3 were developed spontaneously. In total, 20 patients had C6–7 fracture and dislocation (40%), 5 had C7–T1 fracture and dislocation (10%), 3 had C4 (6%), 4 had C5 (%8), 10 had C7 (20%), and 8 had C7 and T1 fractures (16%). One patient with C6–7 fracture dislocation had an accompanying T12–L1 fracture dislocation.

The postoperative ASIA scores of the patients who underwent surgery in all the groups were found to be significantly better than the preoperative ASIA scores ($z=-2.485$, $p=0.013$) (Table II).

There was a mean difference in at least two groups in terms of post-treatment (p=0.029).

There was no significant difference on the outcomes between AF and PF (U=38, p=0.098). Moreover, there was a significant difference on the outcomes between AF and APF (U=38, p=0.015). Post-surgery clinical results in APF were better than other groups because of surgical treatment since the mean ranks in APF and AF were 18.48 and 9.43, respectively.

There was no significant difference between patients with AS and DISH in terms of clinical improvement (U=249, p=0.764).

As a statistical result, there was a significant improvement in all the groups via surgical treatment. However, the most beneficial surgical technique was APF.

In total, 1 of the 7 patients of the AF group died because of pulmonary infection (14%); 2 of the 18 patients who underwent

Table II: Summary Data for the Study Groups and Results

Variables	AF group n (%)	PF group n (%)	APF group n (%)
Number of patients	7 (14)	18 (36)	25 (50)
Neurological improvement	1/7 (14.2)	6/18 (33.3)	13/25 (52)*
Complications	2 (28)	7 (38)	13 (58)
Revision surgery	3 (42.8)	-	-
Death	1 (14.2)	2 (11.1)	3 (12)

p<0.05.* ***AF:* *Anterior fusion,* ***APF:*** *Anteroposterior fusion,* ***PF:*** *Posterior fusion.*

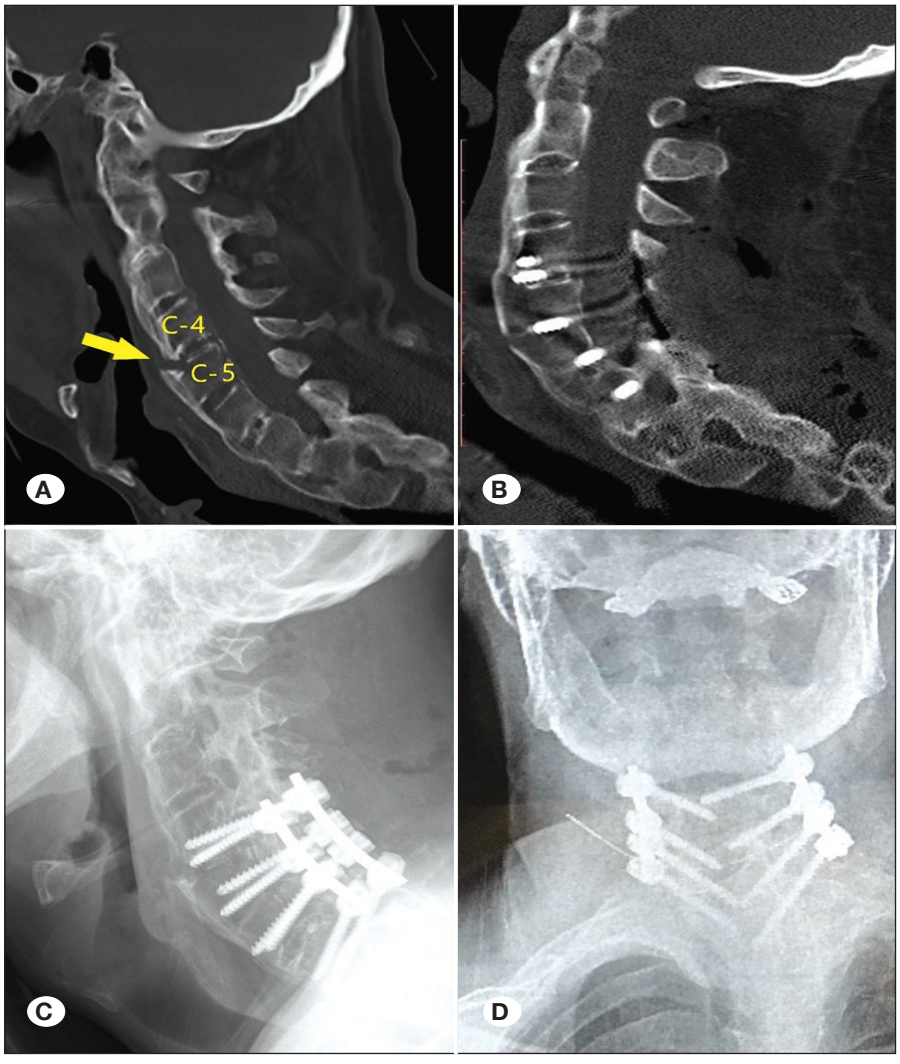

Figure 2: C4–5 Fracture + Posterior Fusion. **A)** Preoperative CT, **B)** postoperative CT, **C–D)** postoperative radiography. The yellow arrow indicates the fracture area. **CT:** Computed tomography.

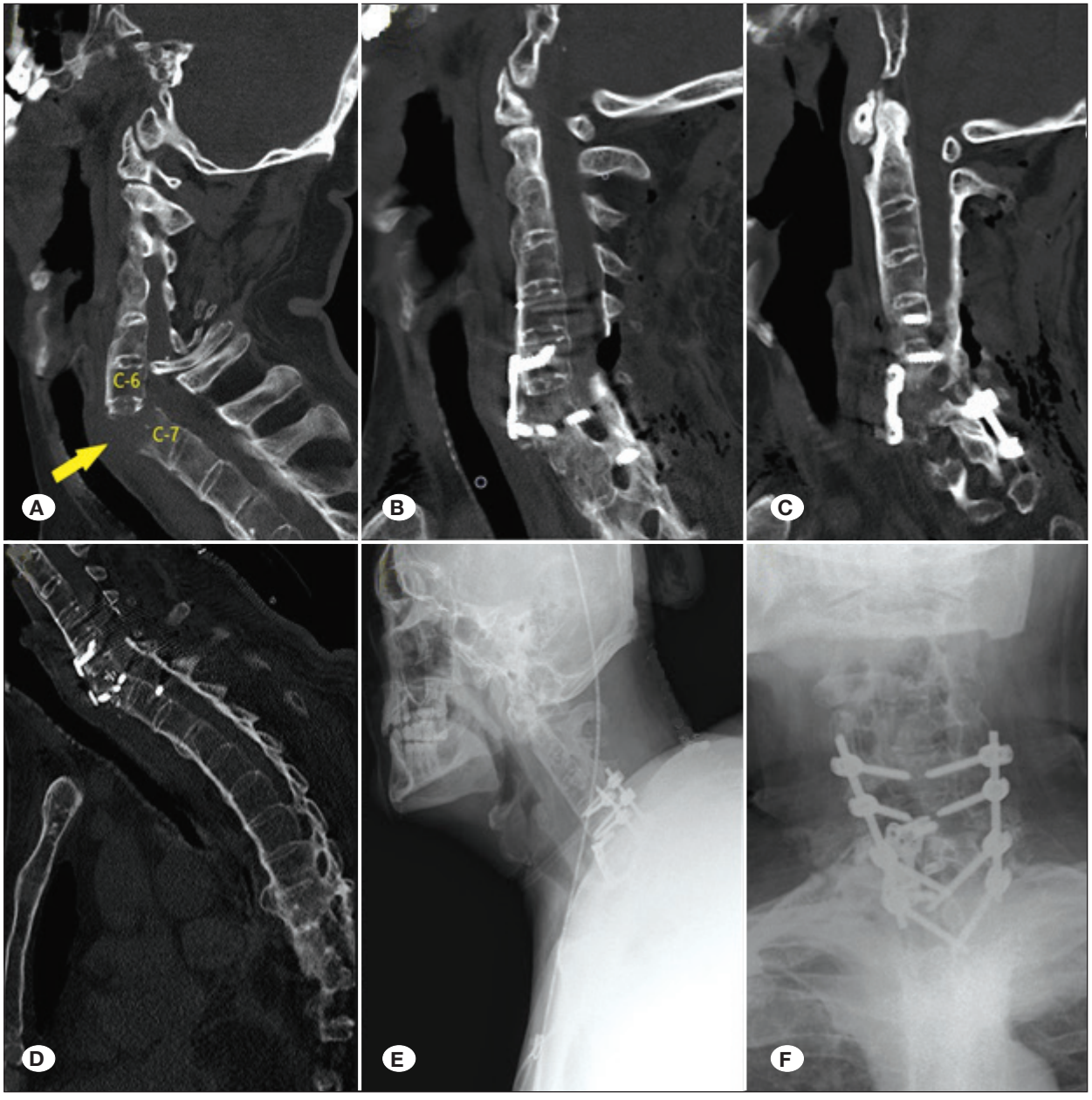

Figure 3: C6–7 Fracture and dislocation + clearance in anterior + anterior posterior fusion. **A)** Preoperative CT, **B–D)** postoperative CT, **E–F)** postoperative radiography. The yellow arrow indicates the angling and cavity in the fracture area. **CT:** Computed tomography.

PF died because of thromboembolism and pulmonary infection (11%); and 3 of 25 patients who underwent APF died. In the APF group, one patient died because of vertebral artery injury, and two patients died because of pulmonary infection (12%). In the AF group, one patient had esophageal rupture and one had pulmonary infection. In the PF group, one patient had vertebral artery damage and three patients had root damage. Two of the patients who underwent PF developed pulmonary infection and one patient developed thromboembolic complications. In total, ten patients who underwent APF developed pulmonary complications, two patients had vertebral artery damage, and one patient developed root damage.

DISCUSSION

Although many classifications related to ASDs have been developed on the spinal fractures, there is no optimization related to the treatment algorithm (4,5,21). The most frequent conditions in ASDs are AS and DISH. These two types of conditions share basic clinical features. Unlike AS, DISH is a non-inflammatory disease and is diagnosed if the bridged ossification of the anterior longitudinal ligament is present in at least four consecutive segments in radiographic scans. The prevalence of DISH is estimated to be between 2.9% and 25%, and it peaks between the ages of 60 and 69 years and gradually increases with age (15,30). The average age in our

series was 58.3 years. DISH tends to be more common in men; however, the etiology of the condition is still unknown (15,30,31). AS is diagnosed in the second and third decades of life. The average age of trauma exposure was 60.9 years in our series and was higher in males (62.9%). In that study, the mortality rates in patients with AS and DISH were 17.7% and 20%, respectively (32). In our series, the corresponding rates were 8.7% and 20%, respectively.

When the fusion of spine occurs because of ASDs, fractures usually occur adjacent to the missile spine or at the junction of the mobile and missile spines. For this reason, if ankylosis occurs, then we can assume that the risk of injury to the cervicothoracic junction is greater (10). In our case series, 86% of the existing fractures were at C6–7, T1 levels, followed by C4–5.

In patients with AS, pain occurs during inspiration due to the fusion of costovertebral joints and ankylosis of the thoracic spine or the involvement of anterior chest wall. A restrictive ventilation disorder may develop due to pain and ankylosis in the joints of chest wall. Fournié et al. described the inflammation and ankylosis of the manubriosternal symphysis and sternoclavicular joints in half of 50 patients, in which 35 patients had AS (9). In our case series, 13 of the patients (26%) developed pulmonary complications due to the lack of available lung capacity. Moreover, four patients who did not respond to treatment died due to pulmonary infection.

The anterior approach is less traumatic, minimizes the risk of displacement during positioning, provides quick stability, has a larger surface area for bone fusion, and has a lower incidence of postoperative infection (12). However, the biomechanical stability of the anterior approach is questionable (22). The commonly observed osteoporosis in the patients undergoing surgery from the anterior approach preferably affects the anterior column and reduces fusion. The failure in anterior fixation in patients with AS has been reported to be up to 50% (13). In a case series of Longo et al. involving 110 patients, 14 patients underwent anterior fixation (13%) (19). In our case series, anterior fixation was performed in 7 of the 50 patients (14%) and was preferred in patients with a weak structure and mild clearance in the anterior and compression regions from anterior to cord. Anterior approach was preferred in only two cases due to morbid obesity and pulmonary problems that would prevent them to be placed in a prone position. In the AF group, three (42%) of the cases were added with posterior fixation one week later due to the loosening of screws. These three cases were not included in the APF group. The patient, who had morbid obesity accompanied by pulmonary problems, was connected to the ventilator on the second postoperative day because of respiratory problems. He died at the end of the one month after surgery due to sepsis following pulmonary infection. The other patient who was morbidly obese with pulmonary problems was a patient with DISH. Esophageal rupture occurred in the anterior surgery. During surgery, the ruptured part was repaired by general surgery with flap rotation. After two weeks of postoperative follow-up, oral administration was provided. In their studies, Kurucan et al., and Longo et al. showed mortality rates of 13.7% and 21%, respectively, in the anterior fusion approach (17,19). The mortality rate in AF group was 14% in our series. The comparison of preoperative and postoperative ASIA scores of the patients in the AF group found a significant difference between them. This observation shows that the patients were benefited surgically.

The posterior approach can restore the alignment of the spine, stabilize the injured parts, and allow an extensive decompression of neurological structures. Multi-segmental posterior fixation with autologous fusion provides a biomechanical advantage as compared to anterior fixation, and has less morbidity as compared to the combined anterior–posterior fixation. However, the disadvantages of this method are as follows: it requires an extensive dissection of cervical muscles, increases the risk of wound infections, and cannot interfere with the anterior spinal cord compression. It is not suitable for cases with cavities in the anterior fracture line (28). In addition, posterior elements being ankylosed and the process-related vertebrae being more osteoporotic may cause difficulties in determining the localization of insertion points of the anatomical instrument. As a result, it can cause pedicle fractures during instrumentation, screw malpositions, neurodeficiency, and vertebral artery damage. Longo et al. applied posterior fixation to 34 patients in a series of 110 cases (31%) (19). In our series, 18 of the 50 patients were in the PF group (36%). Posterior fixation was preferred in patients without anterior compression or with advanced flexion (kyphosis) in the thoracic jaw line and without an adequate anterior clearance. Yan et al. stated that posterior transpedicular fixation is the most powerful technique biomechanically (34). We provided stabilization in the PF group by the transpedicular screw insertion technique. Despite the use of a neuromonitors in three patients, root damage occurred because of screw malposition (C5 in two patients and C6 in one patient). Screws were corrected by performing a revision surgery and root decompression. One patient had vertebral artery injuries at the C5 level during surgery. Screw position was changed by closing the part opened with Bone Wax. No additional neurological deficit was observed in the patient. Two patients with pulmonary infection were followed-up in the postoperative period. After three weeks of treatment, one patient recovered from infection. The other patient could not recover from infection and died at the fourth week after surgery. Moreover, one patient with quadriparesis due to thromboembolism died on the tenth postoperative day. In their studies, Kurucan et al. and Longo et al. showed mortality rates of 7.1% and 14.7%, respectively, in the anterior fusion (17,19). The mortality rate in the PF group was 11% in our series. The comparison of the preoperative and postoperative ASIA scores of the patients in the PF group revealed a statistically significant difference between them. Therefore, this observation shows that they have been benefited surgically.

The combined anteroposterior approach is the present treatment option in the cases with the instability of three columns (18). It is used in approximately 25% of patients with AS and cervical spine fractures (4). The primary indication of adding an anterior approach to the posterior surgery is the

presence of a permanent deformity, cavity, or displacement that damages the spinal cord after reduction with posterior instrumentation. Due to its prolonged surgery duration, blood loss and a high risk of morbidity associated with major trauma, it is less preferred for patients with weak structures (27). Longo et al. performed the combined anteroposterior approach in 62 patients in a series of 110 cases (56%) (19). In our case series, 25 of the 50 patients (50%) were in the APF group. In our series, the combined anteroposterior approach was preferred in the patients with anterior bone clearance or complete listhesis. In the APF group, two patients had injuries in the vertebral artery at the C5 level during posterior transpedicular screwing. Bone clearance was closed with Bone Wax, and the screw angles were changed. In one of these two cases, infarction was detected in the brainstem. The patient died despite the medical treatment. No additional neurological deficits were developed in the other patient. Kurucan et al. and Longo et al. showed mortality rates of 9.9% and 11.2%, respectively, in the patients who underwent combined anteroposterior surgery (17,19). In our series, the mortality rate in the APF group was 12%. In one patient, the screw direction was changed during surgery because of the stimulation of C5 root on the neuromonitor during the screwing process. The patient with postoperative deltoid weakness improved in the second week after medical treatment and physiotherapy. Pulmonary infection was developed in 10 patients postoperatively. In total, eight patients recovered with treatments between 14 days and 1 month, whereas 2 patients died. The comparison of preoperative and postoperative ASIA scores of the patients who underwent APF revealed a significant improvement between them. The comparison of APF group with the other two groups revealed that a statistically difference, there by confirming that the surgical results were better.

Robinson et al. reported no implant failure, non-union, or reoperation during the 52 months of follow-up in 41 patients (24). In our study, 3 patients underwent re-operation due to the early development of implant failure in the AF group. All three patients underwent APF surgery. We did not observe implant failure or nonunion during the follow up. One explanation for this observation might be that the high rate of ossification of the ankylosed spine structure improves fusion.

There may be some possible limitations in this study. First, it is a retrospective analysis. Second, the study was limited by a small sample size. Larger series with long-term follow-up are needed to confirm the effectiveness of the APF approach and to establish whether APF may represent a suitable alternative for selected patients referred to surgery.

CONCLUSION

Surgical management comprising open reduction and internal fixation in patients with ASD prevents neurological deterioration and ensures healing. Although each surgical technique provided significant improvement in clinical results, we found that APF had better neurological recovery than AF and PF in patients with ASD and subaxial cervical spine fractures. APF provides stronger biomechanical support and better spinal alignment. However, when choosing APF as a surgical technique due to better neurological outcomes, its comparatively longer duration surgery and a higher risk of complications as compared to AF and PF, should be actively considered.

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