



Case report

Odontoid fracture complicating ankylosing spondylitis presenting with cervical canal stenosis and quadriparesis: A case report with 5-year follow-up and review of the literature

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ABSTRACT

Introduction and importance: Spinal fractures, especially cervical fractures, are more common in patients with ankylosing spondylitis in comparison with unaffected patients. However, odontoid fractures are relatively rare in these patients. Also, neurological symptoms are not common in odontoid fractures due to the larger diameter of the spinal canal at this level.

Case presentation: Here, we presented a 41 year-old man known case of ankylosing spondylitis who develop odontoid fracture and severe cervical stenosis after falling trauma. Quadriparesis and positive Hoffman sign as well as significant thoracolumbar kyphosis were diagnosed in further investigation. Laminectomy and posterior fixation were executed primary and pedicular subtraction osteotomy was performed two years later to manage the kyphosis and sagittal imbalance. On the follow-up period of five years the patient was fully functional.

Clinical discussion: There are a few cases of odontoid fractures complicating ankylosing spondylitis in the literature. Neurological symptoms are relatively uncommon in odontoid fractures; however, cases with AS can present with neck pain, weakness, and hyperreflexia when having unstable fractures.

Conclusion: Although there is not any gold standard for the treatment of the odontoid fracture in AS, surgical intervention is preferred.

1. Background

Ankylosing spondylitis (AS) is a chronic systemic rheumatic disease affecting the sacroiliac joints and spine. Fourfold fracture risk is reported in some studies in patients with AS compared to the general population, which will eventually lead to low-energy fractures in the course of the disease progress [1]. The cervical spine is the most common site for spinal fractures in patients with AS. Although among cervical spine fractures in patients with AS, subaxial cervical fractures are the most common, a few cases of fractures involving the odontoid process in these patients are reported in the literature [2–6].

In general, odontoid fractures occur in the elderly by low-energy traumas, such as falls, or after motor vehicle accidents in the younger

patients, accounting for 9%–15% of cervical fractures [7]. Because of the larger diameter of the spinal canal at the level of the odontoid process, neurological injury is not common in odontoid fractures [8]; as a result, neurological manifestations are relatively uncommon in these cases which can delay the diagnosis.

The case presented here is a 41-year-old man with AS who was diagnosed with a fracture of the odontoid process following a falling trauma. We report clinical manifestations and neurological symptoms, which are not common among patients with odontoid fracture, in our patient. Also, we have reviewed some cases of odontoid fracture in AS.

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2. Case presentation

A 41-year-old man, salesman and known case of AS was referred to the trauma emergency center with walking disturbance and severe neck pain following falling two days ago. The AS was diagnosed for him at age of 30 years old following the back pain by imaging and clinical examinations. He had no family history of AS or any inflammatory joint disease. Any drug history except periodical use of nonsteroidal anti-inflammatory drugs (NSAIDs), Naproxen 250 mg pro re nata (PRN), was denied. Thoracolumbar global kyphosis in addition to cervical hyper lordosis was obvious during inspection. According to the preliminary physical exam, the vital signs were stable. Moreover, the patient suffered from severe cervical tenderness along with range of motion limitation. Regarding the neurological exam, upper extremity weakness with the score of 2 points of 5 and lower extremity weakness with the score of 3 points of 5 were detected. Upper motor neuron involvement was considered due to the hyperreflexia in both upper and lower extremities as well as positive Hoffman test in both upper extremities. Computerized tomography (CT) scan and magnetic resonance imaging (MRI) were done. Odontoid fracture, type 2 as well as posterior C1-C2 dislocation, were diagnosed in CT scan (Fig. 1A). Furthermore, the MRI revealed severe cervical stenosis along with substantial pressure on spinal cord (Fig. 1B, C).

Cervical spine decompression was done via laminectomy and posterior fixation (Fig. 2). Surgery was achieved by a posterior approach, using an incision along the midline made from the suboccipital area to the spinous process of C6. Posterior spinal instrumentation with occipitocervical fusion was done using a combination of lateral mass and pedicle screws followed by posterior fossa decompression using a laminectomy of C2. The patient was then placed on a collar neck postoperatively.

Two weeks after operation the symptoms were revealed gradually and the motor power improved. Two months later, following physiotherapy and rehabilitation, the patient regained the walking ability.

Two years later, pedicular subtraction osteotomy at the level of L3 as well as lumbar fixation were done due to the severe thoracolumbar kyphosis and sagittal imbalance (Fig. 3).

On the follow-up period of five years after the aforementioned trauma, patient had stable condition and normal physical and social activities (Fig. 4).

This work has been reported in line with the SCARE 2020 criteria [9].

3. Discussion

Patients with AS are more susceptible to spinal fractures, especially cervical fractures [2]. That is because of the ossification of the intervertebral discs and supportive tissues [1,10]. Also, in patients with AS, cervical fractures are considered unstable, resulting in neurological deficits; so, surgical management is the treatment of choice for this condition [2]. However, the odontoid fracture occurs rarely in these patients and infrequently causes neurological injury. Herein, we reported a case with AS that was diagnosed with a type II odontoid fracture - according to the Anderson and D'Alonzo classification - after a falling trauma [11]. Also, neurological symptoms, including quadriplegia and hyperreflexia in the extremities, were presented in our case. Eventually, he was treated with laminectomy and posterior fixation, which revealed the symptoms gradually.

Although odontoid fractures account for 9%–15% of cervical fractures and also, cervical fractures are common among patients with AS, there are relatively few cases of odontoid fractures in these patients in the literature - as mentioned, subaxial fractures are the most common cervical fractures among cases with AS [2,7]. Table 1 shows some of the cases of AS diagnosed with odontoid fractures. As demonstrated in that table, type II odontoid fracture, which was the most common type in the general population [12,13], was more common among the cases with AS relative to other types of odontoid fractures. Also, it is worth mentioning that low-energy traumas, especially falling, were the major cause of odontoid fractures in patients with AS.

The incidence of neurological injury in odontoid fractures is uncommon with a frequency of 2%–27% [7]. As mentioned, the infrequent incidence of neurological injury is due to the larger diameter of the spinal canal at this level [8]. In a study by Apuzzo et al. [14], 45 cases of acute odontoid fracture were analyzed, which showed that eight cases (17.8%) manifested myelopathy (3 hemiparesis, 2 modified Brown-Sequard syndrome, 1 central cord syndrome, 1 tetraplegia). In addition, in another work, Dunn et al. [15] reported 128 cases with odontoid fracture, that 25 (19.5%) of them presented neurological deficits. Also, they demonstrated that neurological deficits were more common in posterior subluxation of the odontoid process compared to anterior subluxation and non-subluxation cases. Goel et al. [16,17], in a more recent study, analyzed 124 surgically-treated odontoid fractures, reporting 110 (88.7%) cases with neck pain, 75 (60.5%) with weakness/spasticity, and 45 (36.3%) with paresthesia. However, there is not any case series study in the literature regarding odontoid fractures in AS due to its rarity. In Table 2, neurological signs and symptoms of 12 cases of

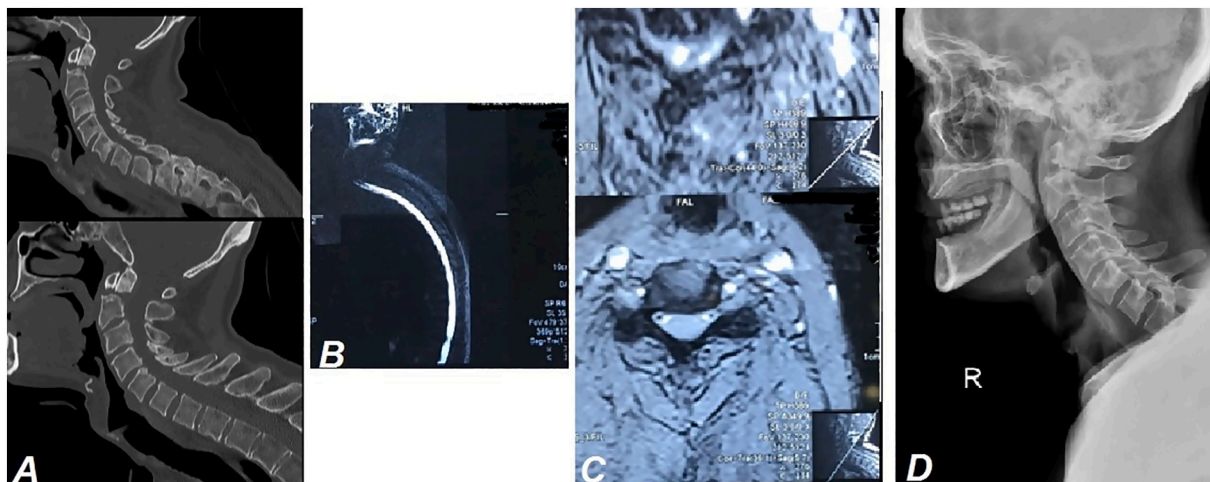


Fig. 1. A: The cervical CTS of the patient, sagittal view, demonstrates odontoid fracture with posterior displacement. B and C: The magnetic resonance imaging (MRI) demonstrates significant compression on cervical spinal cord and hyperintense lesion on T2-weighted Axial MRI images. D: The Lateral cervical radiograph demonstrates odontoid fracture with posterior displacement.

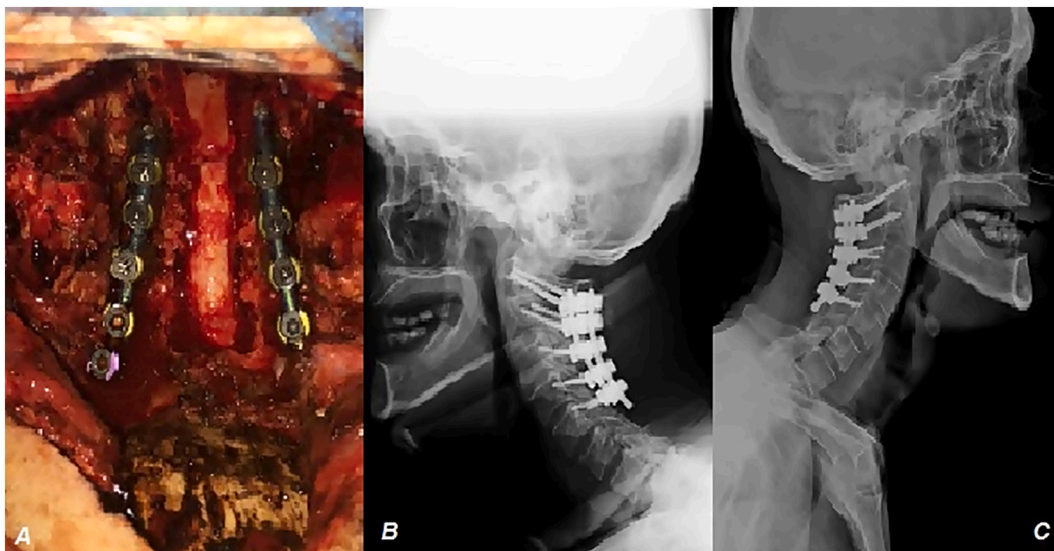


Fig. 2. A: Intraoperative image after decompression and posterior fixation. B: First postoperative X ray. C: Lateral cervical X ray 2 year after surgery.

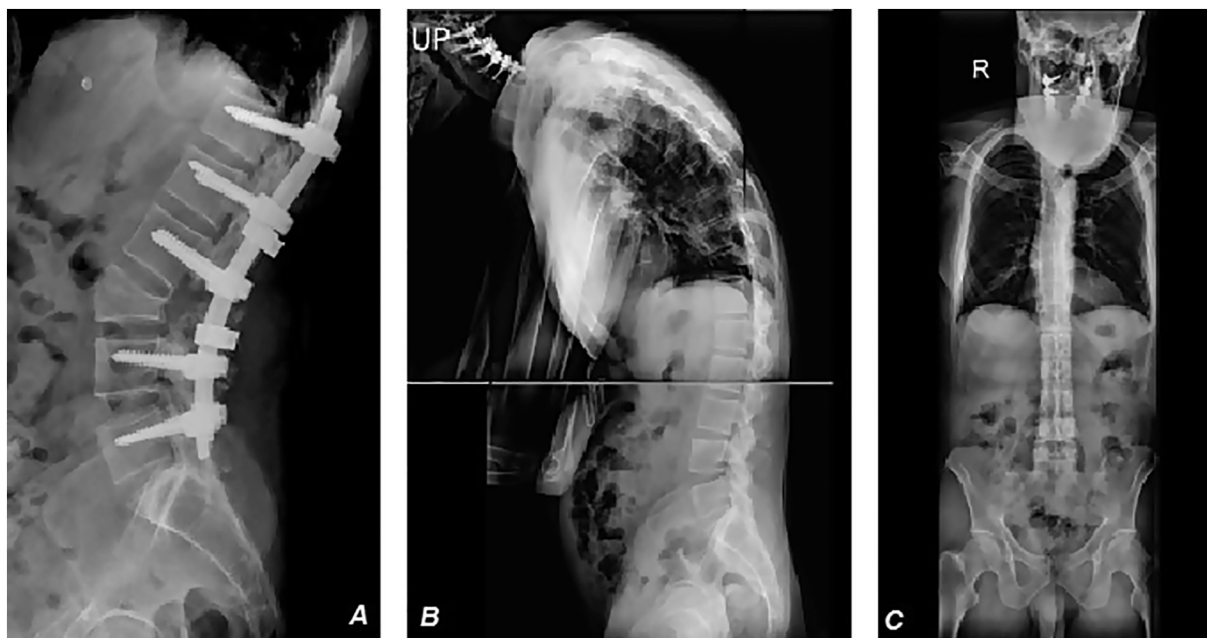


Fig. 3. A: Postoperative X ray after L3 pedicular subtraction osteotomy (PSO). B and C: lateral and AP views of spine profile x ray.

odontoid fracture with AS can be seen. As shown, neck pain (83.3%) and weakness (41.7%), as well as hyperreflexia (33.3%), were the most common symptoms among these cases. Besides, in our case, neck tenderness, weakness in extremities, hyperreflexia, and a positive Hoffman test was presented, as the result of the canal stenosis caused by the odontoid fracture and posterior C1-C2 dislocation.

In addition to AS, which increases the risk of spinal fractures, some other conditions have been reported leading to odontoid fractures. Martel et al. [18] reported a 53-year-old female with rheumatoid arthritis diagnosed with a fracture in the base of the odontoid fracture without any history of trauma. Also, neoplasms can cause pathologic odontoid fractures. These fractures may be secondary to primary tumors, such as benign fibrous histiocytoma of bone [19], or metastatic tumors, such as metastatic breast carcinomas [20] and meningioma [21]. In addition, there is a report of a 37-year-old woman diagnosed with an odontoid fracture because of spinal tuberculosis [22].

Finally, the optimal treatment approach for odontoid fracture in cases with AS is unknown. However, generally, the management of odontoid fractures is evaluated in several studies for comparison of conservative therapies with surgical treatments [23,24]. Currently, due to the risk of non-union and complications, surgical intervention is preferred over conservative treatment, especially for unstable fractures [25]. Besides, a study by Miao et al. [4] evaluated the best approach for odontoid fractures in AS. In the mentioned study, they suggested posterior occipitocervical fusion with internal fixation despite the restriction in head and neck movements.

4. Conclusion

The occurrence of the odontoid fracture is rare in patients with AS; however, a few cases have been reported in the literature. Although neurological symptoms are uncommon among odontoid fractures, some



Fig. 4. Spine profile X ray 5 years after cervical trauma.

of the cases can manifest with these symptoms, especially neck pain and weakness in the extremities, as well as hyperreflexia in cases with AS. In addition to AS, some other conditions can lead to pathologic odontoid fractures, such as neoplasms. The optimal treatment strategy for odontoid fractures in AS is lacking; however, surgical management is preferred.

Availability of data and material

All relevant data regarding this case report has been reported in the manuscript. Please contact the corresponding author for any further information.

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Ethics approval

The present study was approved by the Medical Ethics Committee of Shiraz University of Medical Sciences. The study's purpose was completely explained to the patient, and he was assured that his information would be kept confidential by the researchers. Also, a written

Table 2

Signs and neurological symptoms in 12 cases of odontoid fracture complicating ankylosing spondylitis.

Signs and symptoms	N (total = 12)	%
Neck pain	10	83.3%
Weakness	5	41.7%
Hyperesthesia	1	8.3%
Hypoesthesia/numbness	3	25.0%
Hyperreflexia	4	33.3%
Hoffman/Babinski sign	3	25.0%
Dysphagia/"plummy voice"	1	8.3%

Table 1

Review of 12 cases regarding odontoid fracture complicating ankylosing spondylitis.

Author, year	Age (years)	Odontoid fracture type	Etiology	Signs and symptoms	Treatment
Govender et al., 1987 [26]	52	II	Minor fall onto the forehead	Pain and tenderness in the upper part of the neck	Traction on skull tongs (6 weeks), modified Zimmer collar with a chin piece
Kaplan et al., 1990 [27]	53	II	Fall and motor vehicle accident	Neck pain, hyperesthesia, and hyperalgesia of hands, weakness in extremities, brisk deep tendon reflexes, extensor plantar responses	Halo-traction, soft collar
Ozgoemen et al., 2000 [28]	32	II	Fall	Severe neck pain, numbness, and weakness in extremities, brisk deep tendon reflexes, extensor plantar responses, ankle clonus	Posterior transarticular screw fixation and wiring, Philadelphia collar
Hilton et al., 2008 [29]	69	(not mentioned)	No history of trauma	"Plummy voice", dysphagia	(not mentioned)
Hadjicostas et al., 2010 [12]	51	II	Fall	Severe neck pain	Soft collar, plaster of paris
Albert et al., (2011) [30]	37	II	(not mentioned)	Quadriplegia, diffuse hyperreflexia and clonus, hypoesthesia	Transoral decompression
Moreno Martinez et al., 2016 [5]	44	(not mentioned)	Fall	Neck pain	(not mentioned)
Miao et al., 2018 [4]	50	II	Rear-end car collision	Pain in the occipitocervical junction, limb weakness	Skull traction, occipitocervical fusion with iliac autograft, neck collar
Miao et al., 2018 [4]	43	II	Falling from bicycle	Neck pain	Skull traction, occipitocervical fusion with iliac autograft, neck collar
Miao et al., 2018 [4]	39	III	Lost control of the motorcycle	Neck pain, decreased touch sensation in both hands	Neck collar, occipitocervical fusion with iliac autograft
Tang et al., 2019 [6]	60	II	Fall	Neck pain	Posterior bone graft fusion, screw-rod fixation, neck collar
Khatavi et al., 2020 [3]	28	II	Fall (a year before)	Neck pain, difficulty in balance while walking, weakness of right-hand grip, hyperreflexia of deep tendon reflexes, spastic extremities, positive Hoffman's reflex and Babinski sign	Anterior release and reduction and posterior fusion, cervical collar

consent form was obtained from the patient.

Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor of this journal.

Authors' contributions

K.E. interpreted the patient data and carried out the treatment. H.K., S.P.M, and H.J.M. drafted the manuscript and collected the data. A.S. prepared the figures. M.S. and K.E. revised the manuscript and acts as the guarantor of the manuscript. All authors read and approved the final manuscript.

Registration of research studies

Not applicable.

Guarantor

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Declaration of competing interest

The authors declare that they have no competing interests.

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