International Medical Case Reports Journal

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CASE REPORT

# Atlantoaxial Spondyloptosis: Report of a Case and Review of the Literature

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**Objective:** To present an extremely rare case of atlantoaxial spondyloptosis in a neglected case of odontoid fracture, type-2, with complete anterior displacement. The surgical approach in such cases are always challenging.

**Background:** C2 fractures are the most common type of spinal traumatic injury. Among them, odontoid fracture represents the most variable and complicated C2 fractures. When non-operative management is planned, close follow-up and patient compliance are mandatory. Neglecting these cases can lead to complications such as malunion, nonunion and subluxation, of which, atlantoaxial spondyloptosis is the most severe form.

**Presentation of Case:** The patient was a 20-year-old male with the chief complaint of neck pain, progressive quadriparesis, and gait disturbance since 8 months prior to presentation, with Japanese orthopedic association score of 11. Imaging studies showed anterior non-reducible, malunited atlantoaxial spondyloptosis, associated with a type-II odontoid fracture.

**Methods:** The patient underwent open reduction and posterior atlantoaxial instrumented fusion using Harm's technique. One-year Follow-up examination and imaging evaluations revealed complete neurological recovery, with Japanese orthopedic association score of 15, and complete fusion in normal alignment.

**Conclusion:** A posterior-only approach for the management of atlantoaxial spondyloptosis with malunited type-2 odontoid fracture is associated with lower morbidity and fewer complications than anterior or combined approaches, while still achieving all surgical goals. The integrity of the transverse ligament is mandatory in such cases.

**Keywords:** atlantoaxial dislocation, odontoid fracture, spondyloptosis

## Introduction

Spondyloptosis is the most severe form of spondylolisthesis and is defined as complete slippage of one vertebral body (VB) over another, to the extent that the two facing end plates do not meet at all.[1] Axial load is transferred through occipital condyle, C1 and C2 lateral masses from the skull to the C2 VB; then through the VBs of the rest of the spinal column down to the pelvis.[2] The ligaments of the craniovertebral junction play an important role in stability and transferring the load of the skull to the spinal column. The capsule of the facet joints play the keyrole in keeping alignment while moving head and neck in different directions. transverse ligament (TL), beside the capsule of the facet joints, not only restricts flexion of the neck, binds the movement of axis with the atlantocranial complex.[3] Alar ligaments, also, add more restriction to the rotation of the head alongside the capsule of the first two facet joints of the craniovertebral junction.[2,3] Atlantoaxial spondyloptosis (AAS) is therefore characterized by a complete mismatch and no opposition of the atlantoaxial joint surfaces.[1,2] A hyperflexion injury at the atlantoaxial region may result in a rupture of the TL, a type I or type II odontoid fracture, and loss of restraince and anterior atlantoaxial translation.[1–3] When the TL is still intact, the fractured odontoid process, moves with the atlantocranial complex. Thus, in such cases, remodeling and reduction of the facet joints, brings about the reduction of the dense process and its proper alignment.[4] The majority of patients with atlantoaxial dislocation have a rotational component, as in patients with Down's

Received: 12 July 2025
Accepted: 29 October 2025
Published: 31 October 2025

syndrome, rheumatoid arthritis, or cervical vertebral anomalies.[1–4] AAS is an extremely rare presentation and there are a few reports in the literature. Because a locked dislocated facet component is always present in AAS, management necessitates surgical intervention to achieve reduction, realignment, and decompression of neural tissue.[1–3,5] In cases of AAS with intact TL, posterior approach with joint remodeling and reduction has shown good results; while in case of ruptured TL, anterior approach might be necessary for odontoidectomy.[6–8] In this manuscript, we present a 20-year-old male patient who presented 8 months after trauma, with diagnosis of post-traumatic, malunited anterior AAS associated with a neglected type-II OF. The surgical challenge in such cases is taking the proper approach with least risk of complications and maximum chance of realignment, decompression and fusion.

# Presentation of Case

## History and Physical Examination

The patient was a 20-year-old man who fell from his bicycle 8 months prior. He had been evaluated with a cervical x-ray and discharged without further evaluation or management. He presented with neck pain, progressive weakness in all four limbs, minimal sensory loss in upper extremities, and walking difficulty following the incident. Neurologic examination revealed quadriparesis of 3–4/5, deep tendon hyperreflexia, positive Hoffman sign, positive Babinski sign, spastic gait, and torticollis. There were no sphincter problems. The Japanese Orthopedics Association (JOA) score was 11.

## Preoperative Imaging

Preoperative dynamic cervical spine radiography showed frank instability at the atlantoaxial level with AAS and a radiolucent bony fragment of the fractured odontoid at the space between the body of the axis and the anterior arch of the atlas. Preoperative non-contrast computed tomography of the cervical spine revealed a complete, anteriorly displaced, malunited type-II OF. On the right side, the foramen transversalium of C1 and C2 were far apart from each other in an axial cut of the CT-scan, while on the left side, significantly less space was observed between these two foramina. This indicated unilateral complete AAS on the right side and left-sided atlantoaxial spondylolisthesis (Figure 1). Preoperative non-contrast cervical spine MRI revealed severe spinal canal stenosis at the cervicomedullary junction, with spinal cord kinking in association with signal change (Figure 2). A preoperative CT angiography of the vertebral arteries (VA) was performed to investigate its course, thrombosis or dissection, which was normal (Figure 3).

## Operation and Postoperative Course

After the induction of general anesthesia, orotracheal intubation was performed in a neutral position with the assistance of fiber optics. Neuromonitoring of all extremities was applied prior to positioning for baseline motor and somatosensory evoked potential. The Gardner head clamp was then applied, with the pins fixed 1 inch superior and anterior to the pinna. The patient was then positioned in a standard prone position, with his head fixed in horseshoe head holder, and 3 kgs of traction was applied.

After midline exposure from suboccipital bone to the C3 spinous process, lateral dissection of muscles and soft tissue was carefully performed, till posterior arch of atlas, C2 lamina and superior articular process of axis became visible. The first step was decompression of the craniovertebral junction by removing the posterior aspect of the foramen magnum, up to 1 cm of suboccipital bone, the posterior arc of the atlas, and approximately 5 mms of the superior aspect of C2 lamina. The C2 ganglion was identified, dissected medially, and mobilized laterally to improve visualization of the atlantoaxial joint while protecting the laterally placed VAs.

The posterosuperior aspect of the superior articular process of the axis was the first structure encountered during this step. Beginning on the right side, the next step involved drilling the anterosuperior aspect of the C2-superior articular process, and the posteroinferior aspect of C1-inferior articular process. This procedure was continued under continuous fluoroscopic guidance, until the blade of a small osteotome could be inserted into the atlantoaxial joint (AAJ). After bilateral joint remodeling, reduction maneuver was performed. This process included bilateral placement of the blade of a small osteotome in the AAJ, enhancing traction by 1.5 kgs each time, and clockwise rotation

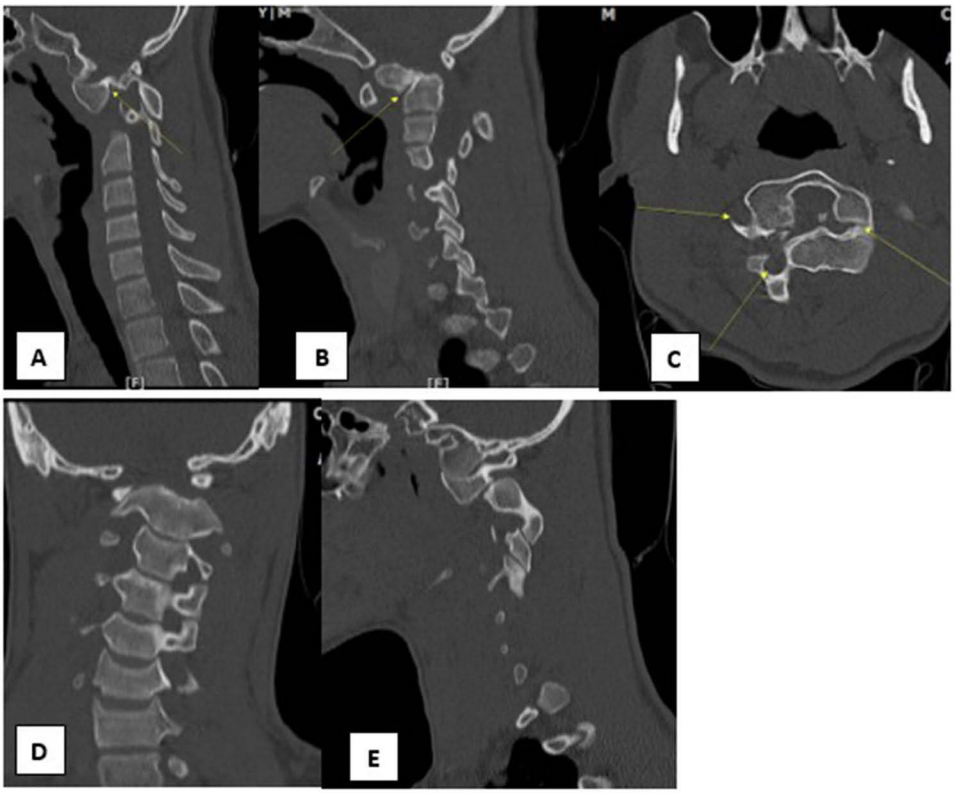


**Figure 1** Preoperative CT scan of cervical spine sagittal, coronal, and axial view; malunited complete anteriorly slipped right atlantoaxial facet (**A**), malunited complete anteriorly slipped of (**B**), departed foramen transversalium of C1 and C2 on the right side, indicating unilateral AAS (**C**), right torticollis as a result of right AAS (white arrows) (**D**) and near complete atlantoaxial subluxation on the left side (**E**).

of the osteotomes. This maneuver was attempted three times (with up to 8kgs of traction applied) and failed to reduce AAJ.

In the next step, the abnormal bony fusion between the odontoid process and C2 vertebral body was carefully removed bilaterally, using 1-millimeter Kerrison punch, following C2 pedicles under fluoroscopic guidance. The reduction was successful after this procedure. Bilateral C1 lateral mass screw and C2 pedicular screw were inserted, and autologous bone chips from decompression procedure were inserted in the AAJ space; A rod was then applied and fixed. Complete reduction was confirmed using fluoroscopy.

The patient was transferred to the ICU and extubated the day after surgery. After 2 days of ICU care and 3 days of ward admission, he was discharged. A Philadelphia collar was prescribed for 4 months.

Post-operative CT and CT-angiographic evaluations revealed complete reduction of AAS and OF, fixed construct in normal alignment, no violation of VAs or internal carotid arteries, and sufficient expansion of the spinal canal and foramen magnum (Figures 4 and 5).

At the 3-month follow-up, the patient reported improvement in neck pain and a progressive decrease in weakness, allowing for normal walking. MRI evaluation showed complete resolution of signal changes in the spinal cord, with no pressure on neural tissue observed (Figure 6). The JOA score was measured 15 at this time.

At the one-year follow-up, CT evaluations revealed complete fusion of AAJ bilaterally, complete fusion of the OF in normal alignment, and posterolateral fusion in atlantoaxial region (Figure 7). Neurological status was assessed as completely normal, and JOA score was 17.

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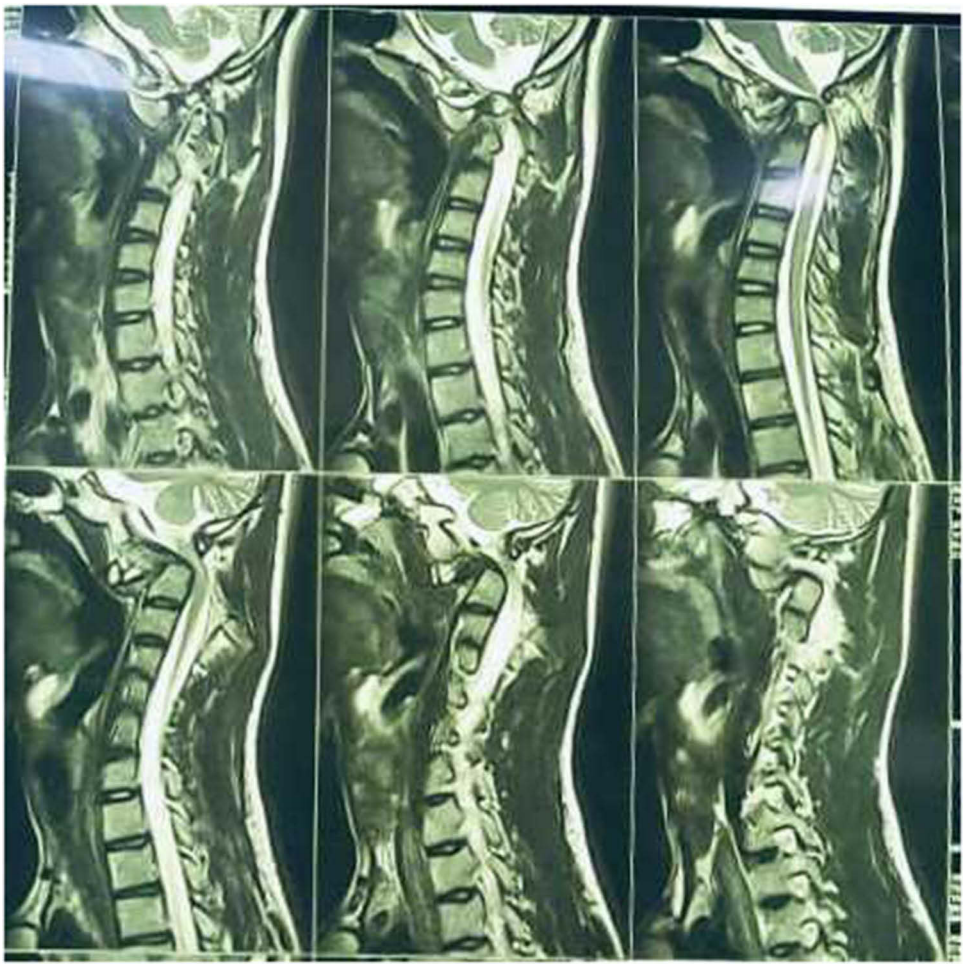

**Figure 2** Preoperative MRI (T2-Weighted) of the cervical spine, showing significant canal stenosis and signal change of the spinal cord at the cervicomedullary junction due to spinal cord kinking.

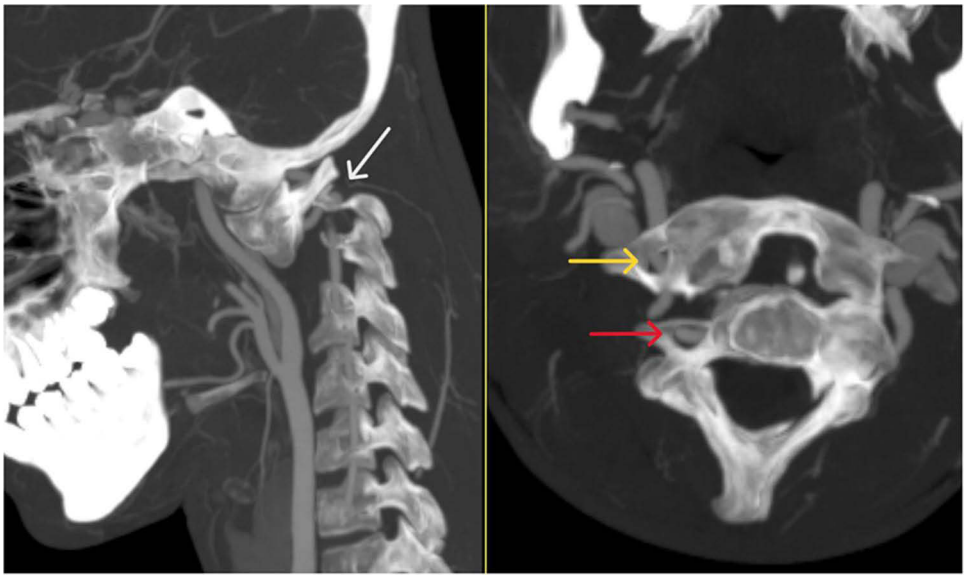

**Figure 3** Preoperative CT angiography of the vertebral and carotid arteries, showing no malformation, thrombosis, dissection or pseudoaneurysm. The white arrow in the left side of the picture shows the competency of VA in its atlantoaxial course on the right side. The yellow arrow on the right side of the picture shows the VA in the foramen transversalium of the Atlas and the red arrow shows the VA in the same foramen of the Axis.

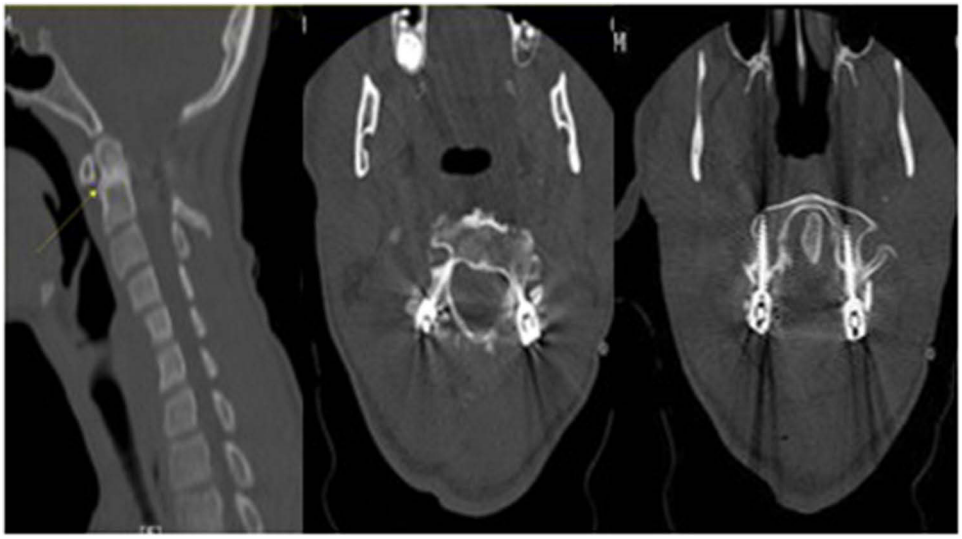

**Figure 4** Postoperative CT scan of the cervical spine showed atlantoaxial PSF with complete reduction and fusion with normal alignment of AAJ and fractured odontoid (white arrow).

**Figure 5** Postoperative CT angiography of carotid and vertebral arteries, without any significant pathology.


https://doi.org/10.2147/IMCRJ.S542587

**Figure 6** MRI (midsagittal T2W) at 3-month follow-up, showing complete release of spinal cord in cervicomedullary junction and resolution of the signal changes in spinal cord.

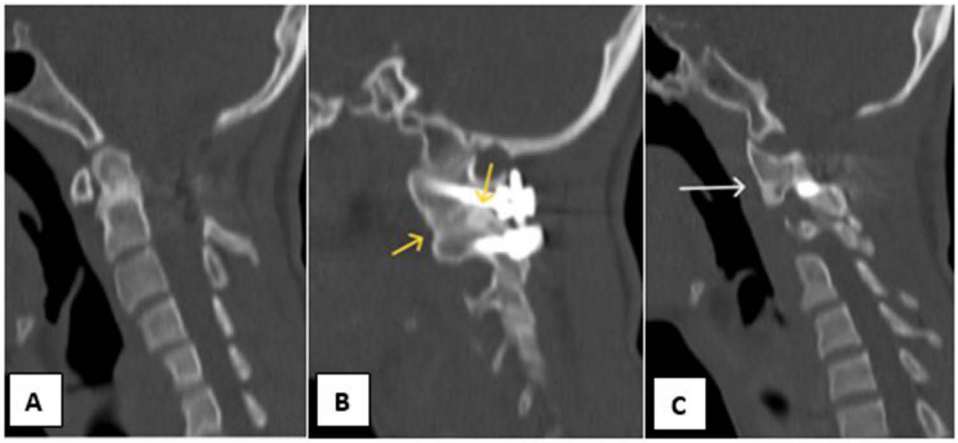


**Figure 7** CT evaluation at one-year follow-up. Complete fusion of OF (**A**), normal alignment, anterior and posterior fusion mass (yellow arrows) on the left AAJ (**B**) and complete fusion (white arrow) in normally aligned position of the right AAJ (**C**).

## Discussion

The craniovertebral junction (CVJ) acts as a transition zone between the cranium and cervical spine region.[1] This axis of gravity originates from the skull, passing through the atlantooccipital and atlantoaxial joints to the subaxial cervical spine, down to the sacroiliac joints, and then to the heels.[1,2] The atlantoaxial relations consist of AAJ and atlantodental components. The AAJ is a unique joint in the spinal column. It not only plays a remarkable role in stability, similar to the facet joints in the posterior column of other spinal regions, but also as the weight bearing vertebral body in the anterior columns in other regions.[4] Thus, any insult to the bony structures or soft tissue in this region should be taken seriously.

The biomechanical characteristics of the impact predominantly predict the injuries in CVJ. When hyperflexion occurs, a dense fracture with anterior displacement, rupture of the AAJ capsule, and subsequent anterior subluxation are anticipated.[1,8] In such cases, cervicomedullary junction of the spinal cord, which contains the most important tracts of vital organ innervation, may kink and lead to instantaneous death. The transverse atlas ligament (TAL) is among the strongest ligaments in the human body. One important point in surviving such circumstances is the competence of the TAL, which supports the fractured odontoid process together with the anterior arc of the atlas, away from the spinal cord.[7,8] Thus, the presentation of atlantoaxial instability is rare in the acute phase, but more common in the chronic phase after the trauma, due to progressive instability in less severe cases.

AAS, although previously described as atlantoaxial dislocation with locked facets,[6,9] was first termed spondyloptosis by Salunke in 2015.[10] Goel subsequently described the management of atlantoaxial dislocation with locked facets – referred to as AAS – utilizing a spacer in the AAJ for reduction and C1-C2 fusion via a posterior-only approach for the first time in 2006.[6]

Table 1 presents the reported AAS cases in the literature. Nearly all of them have a delayed presentation (3–9 months after the trauma). Therefore, the OF, rupture of AAJ capsule, and intact TAL are the three common building blocks of AAS. Repeated motion of the fracture segments leads to disorganized osteogenesis in the region, and causes the accumulation of granulation tissue in AAJ. These two conditions are the main reasons for failure of reduction and non-operative management in AAS.[1,7,11–13] Anterior approach for resection of the fractured dense process is kept for circumstances of ruptured TAL and failure of the posterior approach.[6,7,11]

Traction prior to surgery has often been ineffective in AAS, except in one report.[15] Traction should be applied more anteriorly on the skull to induce sight extension in the reduction process.[6] The most important part of the reduction process in the posterior-only approach is the release of the adhesions and malunions. It consists of complete mobilization of AAJ by removing joint capsule and granulation tissue and may involve articular process drilling or removal of malunions on the surface of the OF.[6,9,13] The reduction maneuver should include not only upward, vertical and posterior directed traction but, in cases with rotatory component, manual rotation.[6,7,9,12] The rotational component of the AAS is

**Table 1** Reports of AAS in the Literature

| Ref | Year | Pathology | No. of Patients | Anterior/ Posterior | Reduction Method | Fixation Construct | Follow-Up |
|---|---|---|---|---|---|---|---|
| [11] | 2006 | Congenital | 4 | Anterior | Joint manipulation and titanium joint spacer | C1 lateral mass and C2 pars screw and plate | 45 months |
| [6] | 2011 | Trauma | 14 | Anterior, Posterior | Joint manipulation and bone chips | C1 lateral mass and C2 transarticular screw and plate | 23 months |
| [10] | 2014 | Trauma | 6 | Anterior, Posterior | Joint manipulation and remodelling | C1 lateral mass and C2 pedicular screw and rod | 4 months |
| [9] | 2018 | Trauma | 1 | Anterior | Joint manipulation and bone chips | C1 lateral mass and C2 pedicular screw and rod | 6 months |
| [7] | 2019 | Trauma, Congenital | 10 | Posterior | Joint manipulation and titanium joint spacer | C1 lateral mass and C2 pedicular screw and rod | 6–60 months |
| [1] | 2019 | Trauma | 1 | Anterior | Joint manipulation and bone graft | C1 lateral mass and C2 pars screw and rod | 6 months |
| [8] | 2022 | Trauma, Congenital | 4 | Anterior | Joint manipulation and bone graft | C1 lateral mass and C2 pedicular screw and rod | 1–4 years |
| [14] | 2022 | Trauma | 2 | Anterior | Close reduction | C1 lateral mass and C2 laminar screw and rod | 3 years |


https://doi.org/10.2147/IMCRJ.S542587

defined as the difference between the distances of C1-C2 foramen transversaria on both sides.[7] Varshney reported the first cases of unilateral AAS and suggested the DCER (distraction, compression, extension, and reduction) procedure in cases of unilateral AAS with rotatory component.[7] The DCER procedure is mainly utilized in cases with basilar invagination, when the insertion of the spacer and traction cannot reduce and align the AAJ and the odontoid process.[16,17]

Although the anterior approach for resection of the fractured odontoid process and stabilization through anterior/posterior approach is always an option, in recent years, posterior approach has received greater attention. In our center, we have significant experience with CVJ surgeries through the posterior-only approach.[16,18–20] Therefore, we chose to perform AAJ remodeling and reduction of OF through posterior-only approach. At the 1-year follow-up, complete reduction and fusion of the OF and the AAS were observed, indicating that the blood supply to the region was maintained and alignment was normal.

## Conclusion

AAS associated with anteriorly displaced odontoid fracture is a rare entity, with trauma to the CVJ being the most common cause. Irreducible atlantoaxial dislocation and even displaced odontoid process can be managed with direct posterior reduction and fusion by distraction of C1-C2 joints in association with drilling and remodeling the parts of the atlantoaxial facets and ossified OF to facilitate open reduction. The integrity of the TAL and anatomy of the vertebral artery are the main limitations in this process. Intraoperative neuromonitoring and step by step fluoroscopic guidance are mandatory.

## Ethical Approval and Institutional Permission

This study was approved by the Ethics Committee of Shiraz University of Medical Sciences. All necessary institutional permissions for data access and usage were obtained in accordance with the approved protocol.

## Patient Consent Statement

Informed consent was obtained from the patient for the publication of this case report and any accompanying information.

## Acknowledgment

The authors would like to thank Dr. Kamran Hosseini (Ph.D. in Molecular Medicine) and Ms. Hosseini for reviewing and editing, in Shiraz Neuroscience Research Center, Shiraz University of Medical Sciences, Shiraz, Iran.

## Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

## Funding

This article has no source of funding.

## Disclosure

The authors declare that they have no competing interests in this work.

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