Revision surgery for os odontoideum with irreducible atlantoaxial dislocation with a transoral approach is not commonly seen. Typically, management of this type of atlantoaxial dislocation is through posterior fixation and fusion or transoral decompression and posterior fusion. This report describes revision surgery in a patient with os odontoideum who was treated with a transoral approach. A 50-year-old man was diagnosed with os odontoideum and atlantoaxial dislocation in 2007 and was treated surgically with posterior occipitocervical internal fixation and fusion. In 2012, he had recurrence of neck pain and numbness of the limbs. Neurologic function was grade D according to the standard neurologic classification of spinal cord injury from the American Spinal Injury Association. Because this was a revision surgery, the internal fixation implant was removed through a posterior approach and a transoral approach was used for release, reduction, internal fixation, and fusion. Two 6-mm cages filled with autogenous bone were introduced into the lateral mass spaces for bony fusion and distraction, and 2 cervical compressive mini-frames were used for fixation. Complete atlantoaxial reduction and decompression of the spinal cord were achieved. The patient reported improvement of symptoms after surgery. Movement of the extremities increased from grade III force to grade V, and neurologic status improved from American Spinal Injury Association grade D to grade E. A transoral approach for release, reduction, bony fusion, and fixation could be an effective procedure for the treatment of os odontoideum with irreducible atlantoaxial dislocation. It provides a new option for bony fusion and internal fixation of the atlantoaxial joint.
A patient with os odontoideum was treated with posterior occipitocervical internal fixation and fusion. Six years after the first surgical treatment, breakage of the internal fixation and nonunion of the bone graft occurred. Therefore, revision surgery was performed with a transoral approach. This report describes the therapeutic options because few reports of such cases appear in the literature. Written informed consent was obtained from the patient for publication of the anonymous clinical data.

**Case Report**

A 50-year-old man with no history of trauma had continuous neck pain, dizziness, numbness, and gait disturbance in 2007. He was diagnosed with os odontoideum and atlantoaxial dislocation and was treated surgically with posterior occipitocervical internal fixation and fusion. After surgery, the patient noted improvement of neurologic symptoms. In 2012, he had recurrence of neck pain and numbness of the limbs. Initial physical examination showed limited range of motion of the neck, with tenderness over the posterior spinous process. Muscle strength was normal in the right upper and lower extremities, but weak in the left upper and lower extremities, with grade III force. Proprioception and vibratory sensation were impaired on the left side. Bilateral symmetric hyperreflexia of the upper and lower extremities was seen, but without Hoffmann and Babinski signs.

Anteroposterior and lateral plain radiographs of the neck showed an irregular ossicle at the tip of the odontoid process with atlantoaxial dislocation, C0-3, 4 internal fixation, and breakage of the rod on the right side. Computed tomography (CT) scan showed nonunion of the bone graft and extenuation of the C1 pedicle. Magnetic resonance imaging (MRI) scan confirmed the CT findings, indicating compression of the spinal cord (Figure 1).

Because this was a revision surgery, the authors removed the internal fixation implant through a posterior approach and used a transoral approach for release, reduction, internal fixation, and fusion. Skull traction was applied. After identifying the position of the atlantoaxial joint with a C-arm image intensifier, the mucosa and muscle layers were incised to reach the anterior aspect of the atlas and axial joints. The atlas was displaced anteriorly. Figure 1: Anteroposterior radiograph showing C0-3, 4 internal fixation and atlantoaxial dislocation after the first surgery (A). Lateral radiograph showing breakage of the rod on the right side (B). Computed tomography scan showing an irregular ossicle at the tip of the odontoid process with atlantoaxial dislocation (C). Magnetic resonance imaging scan showing ventral compression of the cervical spinal cord (D).

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**Figure 1:** Anteroposterior radiograph showing C0-3, 4 internal fixation and atlantoaxial dislocation after the first surgery (A). Lateral radiograph showing breakage of the rod on the right side (B). Computed tomography scan showing an irregular ossicle at the tip of the odontoid process with atlantoaxial dislocation (C). Magnetic resonance imaging scan showing ventral compression of the cervical spinal cord (D).
inferiorly, with excessive scar tissue formed between the atlantoaxial joint. The articular capsules of the bilateral atlantoaxial joints were resected completely. High-speed burr was used to strip the scar and articular cartilage between the bilateral atlantoaxial joints. Loosening and reduction of the atlantoaxial joint was identified. An intraoperative radiograph showed complete reduction of the atlantoaxial joint. Two 6-mm PEEK (polyetheretherketone) impacted cervical cages (Medicrea Technologies, La Rochelle, France) filled with autogenous cancellous bone were introduced into the bilateral lateral mass spaces for bony fusion and distraction. Holes were drilled at the midpoint of the lateral mass of the atlas and the vertebral body of the axis, and a cervical compressive mini-frame (C-Jaws; Medicrea Technologies) was placed into the desired position for fixation, with slight impaction applied at the holder. An intraoperative radiograph further confirmed reduction of the atlantoaxial joint. The wound was closed in layers. After the surgery, the patient was treated with immobilization in a cervical orthosis for 12 weeks.

At 4-month follow-up, movement of the extremities improved to grade V and neurologic status improved from D to E, according to standards from the American Spinal Injury Association. A CT scan showed bony union between the bilateral atlantoaxial joints. Postoperative radiograph and MRI scans showed ideal reduction of the atlantoaxial joint (Figure 2). Compression of the cervical cord disappeared.

**Discussion**

Os odontoideum is an ossicle with smooth circumferential cortical margins, indicating no osseous continuity of the odontoid process with the body of C2.1-3 The origin of os odontoideum is still a matter of debate, with evidence of both congenital4,5 and acquired6,7 causes reported in the literature. Salunke et al6 reported a case of os odontoideum that was fused to a hypertrophied C1 arch, suggesting a congenital mechanism. However, supporters of an acquired mechanism believed that os odontoideum developed after a traumatic event, as suggested by previously normal radiographic findings of imaging of the dens. For example, Wang and Wang6 reported that injury to the epiphysis between the 2 ossification centers of the dens could cause union failure, resulting in os odontoideum. Moreover, other authors noted that both congenital and acquired causes could lead to os odontoideum. Sankar et al8 reviewed a range of cervical spine radiographs obtained from 1991 to 2004 to identify os
odontoideum and concluded that there were 2 separate etiologies: posttraumatic and congenital. McHugh et al reported a 1-year-old girl with an incompletely ossified dens that was persistently attached to the anterior ring of the atlas and developed into a well-corticated ossicle 3 years later. This case suggested that the etiology of os odontoideum was multifactorial and related to embryology and the vascular supply to the odontoid process. Although the cause of os odontoideum remains controversial, the etiology would not affect the diagnosis.

Because os odontoideum can lead to instability of the atlantoaxial joint, patients are vulnerable to neurologic injury after minor trauma. Management of os odontoideum also remains controversial. Some authors reported that os odontoideum in itself was not an indication for surgical intervention. Fielding et al reported 4 patients with an average of 2 years of follow-up without surgical treatment who remained asymptomatic and neurologically intact. Hadley reported a series of 37 patients, 20 of whom were treated conservatively and did well at an average of 7 years of follow-up. They proposed the following: (1) Patients with os odontoideum, with or without C1-2 instability, who had neither symptoms nor neurologic signs could be managed with clinical and radiographic surveillance. (2) Most patients with os odontoideum who had neurologic symptoms and/or signs and C1-2 instability should be managed with posterior arthrodesis, such as C1-2 internal fixation or occipitocervical fusion. (3) Transoral decompression can be considered in patients with os odontoideum and irreducible ventral cervicomedullary compression. These principles were almost the same as the evidence-based clinical recommendations for the treatment of os odontoideum set out by the American Association of Neurological Surgeons and the Congress of Neurological Surgeons in 2001. Menezes reported a series of patients with symptoms of atlantoaxial or occipitocervical instability or extreme basilar invagination arising from os odontoideum and advocated stabilization surgery performed through a posterior approach if the lesion could be reduced preoperatively. Otherwise, they recommended decompressive surgery through a transoral approach, with subsequent fixation. In addition, Wang et al reported a case of a 52-year-old woman with os odontoideum and irreducible atlantoaxial dislocation who underwent surgery with a posterior approach for release, reduction, fusion, and stabilization, concluded that the transoral approach was anatraumatic and effective operative procedure.

For the current patient with os odontoideum and irreducible atlantoaxial dislocation, posterior surgery only for rod substitution was obviously inappropriate because the atlantoaxial joint could not be reduced by posterior occipitocervical fixation but through direct atlantoaxial fixation. Because the patient had been treated with a posterior approach, with formation of local scar tissue, exposure of the bilateral atlantoaxial joints could present great risk. Furthermore, the pedicles of the atlas were too thin for screw placement. Therefore, according to the new classification and treatment protocols for atlantoaxial dislocation, the authors decided to remove the internal fixation hardware with a posterior approach and use a transoral approach for release, reduction, internal fixation, and fusion.

In the authors’ clinical practice, the atlantoaxial joint could be restored by distraction of the atlantoaxial joint and posterior shift of the atlas. A special reductor would also be used for reduction of this irreducible atlantoaxial dislocation, and after reduction, transoral atlantoaxial reduction plate fixation would be applied. However, in the current case, neither the reductor nor transoral atlantoaxial reduction plate fixation was adopted. The transoral procedure is not without complications, especially those associated with internal fixation, such as wound infection, cerebrospinal fluid leakage, neurologic injury, and hardware loosening. Of these complications, wound infection is the most common. The authors believed that because the C-Jaws is much thinner and smaller than the transoral atlantoaxial reduction plate the tension at closure of the wound of the pharyngeal wall would be smaller to prevent the wound from opening. After complete release was obtained by scraping the scar between the atlantoaxial joints, the authors observed loosening and gradual reduction of the atlantoaxial joint intraoperatively. To maintain distraction of the atlantoaxial joint, 2 spacers (6 mm high) filled with autogenous bone graft were placed between the bilateral atlantoaxial joints. After the 2 cages were secured in position, 2 C-Jaws were used to stabilize the atlantoaxial joint, with the cephalad end fixed into the lateral mass of the atlas and the caudal end fixed into the vertebral body of the axis. As a device developed to stabilize an intersomatic cage or graft and increase bone fusion, the cervical compressive staple of C-Jaws leads to shorter surgical time and fewer neurologic injuries. After surgery, the patient was free of all symptoms. Bony fusion was achieved at the 4-month follow-up.

This technique provided instant release, reduction, bony fusion, and fixation in the treatment of os odontoideum with irreducible atlantoaxial dislocation. It also provided a new method for internal fixation through a transoral approach.

**CONCLUSION**

A transoral approach for release, reduction, bony fusion, and fixation could be an effective technique for the treatment of os odontoideum with irreducible atlantoaxial dislocation. It provides a new option for bony fusion and internal fixation of the atlantoaxial joint.

**REFERENCES**

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