Management of Irreducible Atlantoaxial Dislocation with Horizontal Screw-Rod Construct and Technical Note

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ABSTRACT
Os odontoideum is an independent ossicle of variable size with smooth circumferential cortical margins separated from the axis. The natural course of this anomaly is atlantoaxial dislocation either gradually or after a trauma. Atlantoaxial dislocation due to os odontoideum is mostly reducible, visible in dynamic lateral cervical radiographs or with cranial traction and the term reducible atlantoaxial dislocation or RAAD is used for this condition. Less frequently, the dislocation might be neglected and become chronic and fixed, designated as irreducible atlantoaxial dislocation or IAAD.

Two different strategies exist for management of IAAD: a resection strategy mainly with odontoidectomy and a releasing strategy that is focused on the release of the C1-C2 facet joints. Recently, a releasing strategy has found a definite role in treatment of irreducible cases.

Various techniques for reduction of irreducible atlantoaxial dislocation (IAAD) have been described in the literature. In all these methods, the release of C1 and C2 facet joints from the scar tissues is the priority. The releasing strategy can be used either anteriorly via the transoral, retropharyngeal, transnasal and transcervical corridors with the aid of a microscope and endoscope or posteriorly via the conventional midline posterior approach.

Herein, the authors describe a 20-year old man with progressive quadriparesis. Plain dynamic radiographs, computerized tomography and MRI disclosed displaced an orthotopic os odontoideum with fixed forward atlantoaxial subluxation. One week of skeletal traction was ineffective and the patient underwent surgery via a posterior midline incision. After resection of C2 nerve root and releasing of the C1-C2 facet joints of scar tissues, reduction was achieved with application of C1-C1 and C2-C2 horizontal rod construct. Finally after reduction, C1-C2 osteosynthesis was performed with the Harms technique followed by transfacet arthrodesis. The patient showed marked recovery at 3-month follow-up. At 3-year follow up he is doing very well.

KEY WORDS: Atlantoaxial dislocation, surgical technique

INTRODUCTION
Os odontoideum (OO) is an oval or round-shaped ossicle of variable size with a smooth cortical border (2, 3, 7, 23, 31, 33). It may be located in the position of the odontoid process without any bony connection (orthotopic) or near the base of the occipital bone in the region of the foramen magnum (dystopic) (2,3,7,23,31).

This abnormality frequently leads to atlantoaxial dislocation (AAD) (33). Most of these dislocations are reducible where the condition can be managed with posterior C1-C2 fusion and stabilization via different posterior wiring techniques, clawing, transarticular C1-C2 screw, screw plate and the Harms technique (4, 5, 8, 1, 40). In a minority, atlantoaxial dislocation might be missed and gradually become
irreducible (IAAD) (3, 16). The treatment of irreducible atlantoaxial dislocation (IAAD) presents a considerable challenge to the treating surgeon. Traditionally, the most commonly employed procedure was occipitocervical fusion in association with atlas arch laminectomy without trying to reduce the condition that had variable results (9). Later, transoral transpharyngeal, transnasal and transcervical odontoideectomy followed by occipitocervical fusion became popular (12,17,24,29,31,32,41,43,44). However, trying to release the C1-C2 facet joints either anteriorly or posteriorly captured attention as an alternative procedure. Releasing strategies for reduction of such irreducible conditions followed by stabilization via different corridors and techniques are increasingly described and employed (1, 20, 25-28, 31, 35, 38, 39, 45, 46).

In releasing procedures, C1-C2 joints are cleared from obliterating granulation and scar tissues that hamper the reduction. Anterior releasing methods are more challenging with a more complicated learning curve where posterior releasing methods have a fast and short learning curve due to the familiarity of the neurosurgeons. In posterior releasing strategy, the releases of C1-C2 facet joints are done via the posterior midline approach that is mostly based on the experience of Goel (14).

Herein, we describe the horizontal rod technique as an alternative accomplished via a posterior corridor in a 20-year-old male with irreducible atlantoaxial fixation.

**CASE REPORT**

A 20-year-old male was referred to our clinic in March 2011 because of progressive weakness of all four extremities of 6 months duration. Examination demonstrated findings that were consistent with cervical myelopathy such as difficulty in manual fine motor skills, bilateral Hoffmann signs and diffuse hyperreflexia in the upper and lower extremities compatible with quadriplegia. The MJOA score used for the patient's neurological status was 13. Lateral neutral cervical radiograph revealed atlantoaxial dislocation with forward displacement of atlas with respect to axis. Dynamic cervical X-ray confirmed that we were faced with a fixed atlantoaxial dislocation (Figure 1). MRI of the cervical spine revealed cervicomedullary compression at the level of atlas (Figure 2). Axial and reconstructed 2- and 3-dimensional computerized tomography revealed os odontoideum with displacement of C1 on C2 and fixed atlantoaxial dislocation (Figure 3).

Cranial traction was started with 2 kg and was increased gradually to 6 kg. This was continued for a week without any reduction. Surgical intervention was therefore recommended.

The procedure required the patient to be placed in a prone position. After induction of general anesthesia, he was turned into the prone position with the head positioned in the neutral position on a horseshoe headrest while cranial traction was maintained. Lateral fluoroscopy was performed after appropriate positioning and was repeated at intervals during the surgery where the pathology remained constantly irreducible.

Subsequently, after a midline incision, the exposure of the posterior aspects of C1–C2 was obtained, and self-retaining retractors were used to maintain the exposure.

Later, C2 nerve roots were exposed on both sides and transected. Subsequently, C1 lateral mass and C2 pedicle screws were inserted in both sides. Then, with distraction applied to the C1 and C2 screws on the right side, the intervening facet was identified. A blunt osteotome-like instrument was inserted in the facet joint in anterocaudal direction and with its rotation the joint was properly opened. Thereafter, the joint was cleared from scar tissues. The same scenario was repeated on the contralateral side. After releasing of the facet joints, a manual maneuver was tried which had no effect on reduction. At this time, two C1 polyaxial screws were connected with a horizontal rod and the nuts were tightened (Figure 4). The same was repeated in C2 screws and they were connected with an appropriate size rod.

Later, with the aid of a rod holder, the C1-C1 horizontal rod construct was pulled backward and cranially while the C2-C2 horizontal rod was pushed forward and caudally by the assistance. This was continued in a gentle manner for a few seconds till a pop indicating the reduction and realignment was heard. Thereafter, arthrodesis of C1–C2 facet joints was accomplished after decortication with a small high-speed drill and their packing with demineralized allograft bone chips mixed with bone marrow aspirate. Then, the temporary horizontal rods were removed and C1-C2 stabilization was done appropriately with two longitudinal rods placed in the polyaxial screw heads and secured in position on both sides according to the Harms technique (Figure 5).

The patient tolerated the procedure without any postoperative complication. AP and lateral radiographs of
Figure 1: The lateral dynamic radiographs taken in (A) flexion and (B) extension show irreducible dislocation in a patient with os odontoideum.

Figure 2: The preoperative magnetic resonance image of the same patient. (A) It can be seen that the body of the axis is compressing the cord at cervicomedullary junction. (B) Axial slice at the level of C2 shows the facet joints of atlas are located anterior to the axis.
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Figure 3: The preoperative computed tomographic scan of the same patient. (A) Axial cuts show both facets of C1 are in front of the body of C2. (B) The sagittal reconstructive image demonstrates an os odontoideum and marked forward displacement of atlas on C2 (C,D,E). The preoperative 3D reconstructive image shows marked forward atlantoaxial displacement.

Figure 4: An intraoperative photograph taken before reduction. Note forward displacement of atlas and C1-C2-screw-horizontal rod assembled.

Figure 5: A lateral fluoroscopic image was taken after reduction and replacement of the final rods.
the cervical spine obtained postoperatively showed optimal alignment (Figure 6). At 2-month follow-up, in May 2011, the MJOA score of the patient became 16. Reformatted CT scan obtained at this time revealed persistence of alignment with the bone chips at the C1-C2 facet joints (Figure 7). Now at 3-year follow-up, a telephone call revealed that the patient is doing well.

**DISCUSSION**

The atlantoaxial joint has the greatest mobility of any spinal motion segment in flexion, extension and rotation. Stability at this joint is conferred by the dens and the ligamentous structures surrounding it, including the transverse, apical and alar ligaments. Forward or backward displacement of an os odontoideum with its partner atlas arch with respect to axis result in anterior or posterior atlantoaxial dislocation respectively (2,3,7,23,31). Atlantoaxial dislocations are classified as reducible (RAAD) and irreducible (IAAD). This classification dates back to Arvin Greenberg in 1968 (16). Atlantoaxial dislocations due to this pathology are mostly reducible but less frequently might become fixed or irreducible.

**Reducible Atlanto-Axial Dislocation (RAAD):** A lesion is considered to be “reducible” when traction or change in head position allows C1-C2 realignment so as to relieve compression of the CMJ. This means that before using the term irreducible, cranial traction should be tried first and continued for at least a week emphasizing the fact that even long-standing fixed atlantoaxial dislocations might be reduced by preoperative conscious traction as well as intraoperative cranial traction under general anesthesia. Furthermore, response to traction also has prognostic value since radiological alignment and clinical improvement achieved following traction suggests that cervicomedullary myelopathy is probably reversible.

Reducible atlantoaxial dislocations can be best treated with posterior osteosynthesis and fusion. Various techniques have been described for fixation of reducible atlantoaxial dislocations. Historically, this type of operation was based on Gallie’s posterior wiring technique and its modifications described by Brooks & Jenkins, Dickman et al and Brockmeyer (4,5,8,11). In all these techniques, mechanical stability is achieved with structural tricortical iliac bone blocks fixed in compression between C1 and C2 with wire or cable (4,5,8,11,36). Later, a C1-C2 interlaminar clamp was used as a fixation device for this type of dislocation (30, 36).

Subsequently, in an effort to enhance the fusion rate at C1–C2, Magerl and Seemann developed a fixation procedure designated as C1-C2 transarticular screw technique which was used frequently for management of RAAD in os odontoideum (13,18). Combination of this technique and posterior wiring produced the strongest construct introduced to that date.

A decade later, rigid reconstruction, previously unattainable in this problematic region of the spinal column, could be achieved with application of plate and screw by Goel and Lehari (10,15). After a few years, Harms and Melcher proposed C1 lateral mass and C2 pedicle screw osteosynthesis with application of polyaxial screw-rod construct (19,22,30). Nowadays, C1 lateral mass and C2 pars/pedicle poly axial screw-rod fixation is the most accepted technique employed for stabilization of reducible atlantoaxial dislocations (22, 30). Both autogenous and allograft can be used for arthrodesis (21).

**Irreducible Atlantoaxial Dislocation (IAAD):** An atlantoaxial dislocation where the dislocation is not reduced by neck movement or traction is designated irreducible.

In patients with os odontoideum, the atlantoaxial joint remains stable for a variable time with the support of ligamentous structures. However, daily activities and
repetitive flexion movements of the head gradually result in forward displacement of the C1 facet joints on horizontal C2 facet joints. With further anterior slippage of the atlas, the ligamentous support fails and will finally be lost. Simultaneously, remodeling of C1-and C2 facet joints occurs and facilitates forward translation while preventing reverse backward motion. The synovial infolding, scar tissues and contracture of capsules of the C1-C2 facet joints and their remodeling ultimately result in IAAD.

Surgical management of irreducible atlantoaxial instability in os odontoideum with irreducible atlantoaxial dislocation poses considerable difficulties due to the highly variable anatomy of the upper cervical spine and surrounding neurovascular structures, making it a challenging scenario.

Through a review of the literature, we found that it might be helpful to classify all contemporary techniques used in management of IAAD due to os odontoideum in two main

Figure 7: Reconstructed CT scans taken 2 months after surgery (A) show a well-maintained ADI and good alignment. (B) Note intraarticular bone graft.
strategies: (1) Resection strategy that aims for resection of os odontoideum ventrally, (2) Releasing strategy focusing on the release of the C1-C2 facet joints either anteriorly or posteriorly. However, the decision paradigm should be tailored on an individual basis.

Resection strategy: Removal of the os odontoideum to get rid of its persistent compressive effect on the cervicomedullary region has always been an option in irreducible conditions. This can be done through the transoral-transpharyngeal, transnasal and transcervical corridors. Via these techniques, the odontoid, the coexisting offending anterior bony masses, redundant ligaments, granulation and hypertrophic scar tissues should be removed till adequate ventral decompression can be achieved. Transoral odontoidectomy for os odontoideum dates back to 1968, when Greenberg and Scoville accomplished this procedure in two patients with IAAD due to hypoplastic separate odontoid successfully (17) in the largest series, 28 out of 134 patients operated by Menezes for os odontoideum had irreducible atlantoaxial dislocation with cervicomedullary compromise in whom a transoral decompressive procedure was performed (31).

For several years, this technique was recommended as the best option and remained the procedure of choice for irreducible conditions affecting the atlantoaxial joints for many authors.

However, because of the drawbacks and morbidities encountered in the transoral-transpharyngeal corridor such as local infection, retropharyngeal abscess, meningitis, palatal dehiscence, pharyngeal dehiscence, delayed pharyngeal bleeding, velopalatine incompetence, and persistent hoarseness, the transnasal endoscopic odontoidectomy was introduced. Leng et al described endonasal endoscopic resection of os odontoideum with fewer complications in comparison to the former routes (24). Magrini et al described an endoscopic endonasal odontoidectomy in a patient with Down syndrome suffering irreducible IAAD due to os odontoideum (29). Visocchi et al in 2011 described their experience with endoscopic assisted microsurgical odontoidectomy in 7 patients where one of them had os odontoideum (37). In 2011, Gempt et al used the same procedure in a 52-year-old female with os odontoideum (12). Recently, Yen et al operated on 13 cases suffering from IAAD via transnasal endoscopic odontoidectomy without resection of nasal turbinates. 2 out of these 13 cases had os odontoideum (44). All these patient underwent occipitocervical stabilization thereafter (29, 44).

A much safer route is endoscopic odontoidectomy via transcervical corridor that has been described by Wolinsky et al (41). This route is the same as has been described by Smith and Robinson and the only difference is that the incision is at the high cervical region.

However, in fixed dislocation due to os odontoideum, mainly of the dystopic variety, the cervicomedullary compromise may not necessarily and mainly be from the os odontoideum but the assimilated atlas or the body of axis might be the cause of further compression. Therefore, resection of the caudal part of clivus, arch of atlas and the cranial part of axis might be necessary.

Regardless of the corridor used for resection of the os odontoideum and other offending tissues, instability of the atlantoaxial joints exists and might become even worse. Therefore, after achievement of adequate decompression for avoiding serious sequels of further instability and in order allow early mobilization and rehabilitation, a second operation for stabilization is mandatory (32, 44). For this purpose, occipitocervical instrumentation and fusion are recommended by most authors. Odontoidectomy for irreducible AAD due to os odontoideum has currently become confined either to dystopic os odontoideum, in which there is an apparent non-reducible compression of the foramen magnum or in those orthotopic type that the deformity cannot be reduced by intrafacet releasing methods. This means that the necessity of odontoidectomy in all orthotopic os odontoideum cases might be re-visited. This especially applies for odontoidectomy via the transoral corridor where risks and complications exist despite refinements of the surgical procedure.

Releasing strategy: This strategy is composed of different challenging procedures either ventrally or posteriorly that aim to clear C1-C2 facet joints from the scar tissues hampering reduction. During recent years, our understanding of releasing strategies in irreducible atlantoaxial dislocation has been enhanced by several contributions. These releasing methods seems very effective, particularly if we consider that in longstanding subluxations, failure of reduction is almost certainly due to damage of the joint capsule, synovial infolding, interposition or invagination of the granulation tissues into the joint aggravated by muscle spasm or ligamentous contracture and even bony fusion. The decision to release from the front or the back is largely based on the surgeon’s preference, experience, and comfort level with the particular approach. Nonetheless, following
any releasing procedure via open reduction, stabilization of the corresponding joints is mandatory either in the same session or in staged manner.

**Anterior releasing methods**: Anterior release of the C1-C2 facet joints under skull traction is an attractive procedure, which can be accomplished through the transoral, retropharyngeal, endoscopic endonasal and transcervical corridors.

In fact, transoral release of the C1-C2 facet joints under skull traction should be ascribed to Subin in 1995 who released the locked joints transorally for the first time. He subsequently and after interfacet fusion kept the patient in skull traction for a few days and with further application of Minerva cast, solid fusion could be achieved (34). C. Wang et al. were one of the first to introduce this technique, and after transoral C1-C2 facet joint release and reduction by traction, preferred posterior C1-C2 transarticular screw fixation.

The transoral route remained the accepted corridor, till retropharyngeal approach for release of the corresponding joints with sequential posterior internal fixation was introduced, for IAAD. This corridor has some proponents such as Hao et al, who described their experience with the same technique in 2013 (20). Later Lu et al. preformed the same technique though the same route but endoscopically in 21 cases, seven out of these cases had os odontoideum (27).

However, Yin et al. in 2005 released C1-C2 articular joints' capsule and performed reduction of atlantoaxial dislocation transorally, but stabilized the joints with supplementing plate and screw through the same corridor in five patients with irreducible atlantoaxial dislocation, two of which had a dysplastic odontoid (45). In 2010, Xiang Wang et al. described transoral one stage anterior C1-C2 joint release followed by anterior C1-C2 osteosynthesis in a 52-year-old female with os odontoideum who was suffering from irreducible atlantoaxial dislocation (39). In 2011, Ai et al. introduced a novel one stage transoral technique. In their technique after resection of the articular capsules, the granulation and the scar tissues inside the atlantoaxial facet interspace, reduction was achieved with transoral atlantoaxial reduction plate (TARP) (1).

Release of C1-C2 facet joints via conventional high cervical approach is not impossible but very difficult. Liu et al. could accomplish this via high cervical incision and using conventional self-retaining cervical retractors where simultaneous retraction of the mandible was maintained with S-shaped hand held retractor. The corresponding facet release was done with the aide of a fiberoptic endoscope and light source. After release of the facet joints at both sides, reduction became possible with traction and manipulation (25). Subsequent to alignment, the pathology was stabilized via posterior internal fixation. The same technique was employed in 2014, by Ma et al., for irreducible AAD (28). However, Wu et al. in 2010 could access the corresponding field with tubular approach (42). The entry incision was high cervical, medical to sternocleidomastoid. A K-wire was aimed to C1-C2 facet joint and after enlarging the route with different size dilators, a tubular retractor if fixed in place, via this working channel, release of the facet joints could be achieved with the aide of endoscope where reduction became possible with cranial traction. Subsequently, the corresponding joints could be stabilized with percutaneous anterior transarticular screws. It seems that retropharyngeal, transnasal endoscopic and transcervical endoscopic release of C1-C2 facet joints might become more popular in the future without facing the disadvantages that might be seen in transoral route.

In the majority of the latter three releasing corridors, stabilization can be achieved via posterior route in the same session or in staged manner.

**Posterior releasing methods**: Posterior techniques of reduction are more attractive than demanding anterior corridors because of the familiarity of most neurosurgeons. The technique of posterior C1-C2 facet release and their subsequent reduction dates back to Goel. In this technique, after sectioning of C2 ganglion bilaterally, C1-C2 joints are opened by direct manual facetal distraction by the aid of a small osteotom, Thereafter, reduction by means of Goel plate and screws can be accomplished. Initially, a plate of appropriate size is placed on the facet joints of C1 and C2 vertebrae and the plate is fixed by tightening the C2 screw. Then, in order to pull the C1 facet back, the corresponding C1 screw is tightened. This is similar to reduction of spondylolisthesis where sequential tightening pulls back the second vertebra.

In 2013, with consideration of independent acquisition of C1 lateral mass and C2 pedicle polyaxial screws in Harms technique, Yin et al. described their experience in reduction of IAAD by a maneuver similar to Goel’s (46). Their proposed mechanism of reduction is that the implanted rods between C1 and C2 act as a lever system. After tightening C2 nut on
one side, the corresponding rod draws C1 backward. With accomplishment of the same maneuver on the contralateral side, the optimal reduction and alignment might be obtained. In the same year, Suh et al introduced their technique where a temporary T shape tool was used instead of rod acting as a lever in order to pull back C1 lateral mass screw. In 2011, Liu et al described their heroic technique named posterior cable-dragged reduction/cantilever beam internal fixation surgery where the atlas arm is pulled back with the aid of a cable tightened on a U shape rod already attached to bilateral C2 C3 lateral mass screws.

Another interesting method is posterior rotating rod strategy introduced by Chang–Wei et al. In this method, C1 lateral mass screws, C2 and C3 pedicle screws are inserted at appropriate points, subsequently a curved rod with its convex side posteriorly are placed in the tulips of the screws and the nuts are tighten but not fully. Then with two rod rotators, both rods simultaneously are rotated till the rod become in lordotic position.

It should be noted that direct facet joint opening and wide removal of the articular cartilage and scar tissues are prerequisite of all releasing techniques either anteriorly or posteriorly.

In summary, the treatment of irreducible AAD is still a demanding procedure. Posterior releasing/reduction techniques are less challenging than ventral procedures for majority of the neurosurgeons. Horizontal screw-rod technique described in this paper might provide a stronger and more effective operative maneuver than the previous ones in the treatment of IAAD. In previous techniques, the lever arm pulls back single C1 lateral mass screw where the force derived by a strong rod holder on horizontal rod technique pulls back two C1 screws simultaneously. Therefore, greater force can be applied with more chance of reduction and less possibility of screw pull out. Furthermore, the application of this horizontal construct should not be confined to atlantoaxial joints and its efficacy in other irreducible conditions throughout the vertebral column can be tried.

REFERENCES

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