Posterior Screw-Rod Fixation and Selective Axial Loosening for the Treatment of Atlantoaxial Instability or Dislocation Caused by Os Odontoideum: A Case Series for a Single Posterior Approach

Jia Shao, Yan Zheng Gao, Kun Gao, Zheng Hong Yu

■ OBJECTIVE: **To evaluate the effect of screw-rod fixation and selective axial loosening in the treatment of atlantoaxial instability or dislocation (including reducible and irreducible) caused by os odontoideum (OO) via a single posterior approach.**

■ METHODS: **A consecutive series of patients with OO surgically treated in our hospital were retrospectively analyzed. For atlantoaxial instability and reducible atlantoaxial dislocation, C1-C2 screw-rod fixation and fusion were performed. OO combined with irreducible atlantoaxial dislocation was reduced after posterior axial loosening, followed by screw-rod fixation and fusion. The general information, clinical data, and radiographic data were compared between the 2 different procedures.**

■ RESULTS: **There were 41 patients with an average age of 40.6 ± 21.7 years. All the patients underwent posterior reduction and C1-2 screw rod fixation, 6 with axial loosening and 35 without axial loosening. The clinical manifestations and radiographic data significantly improved after the operation with a low rate of complications. Except for clivus-canal angle and visual analogue score of cervical pain, there were no differences in clinical and radiographic data between the 2 procedures.**

■ CONCLUSIONS: **Posterior screws-rod fixation and selective axial loosening is appropriate for treating OO complicated with atlantoaxial instability or dislocation (including reducible and irreducible) without the need for anterior decompression.**

INTRODUCTION

An os odontoideum (OO) appears as an isolated and smooth ossicle replacing the normal odontoid process, with smooth edges discontinuous with the base of the odontoid process or axis.[1] OO was first reported by Giacomini in 1886. Its etiology has been controversially explained with 2 different theories: congenital or posttraumatic lesion.[2] Each theory has their convincing evidence[3-9]; however, neither affect the choice of treatment.[1]

The clinical manifestation of OO ranges from no symptoms or only neck pain to severe myelopathy, disappearance of spontaneous breathing, and even sudden death.[10-12]

OO complicated with symptoms and signs of myelopathy or C1–2 instability requires C1–2 fixation. Clinical and radiologic surveillance or aggressive posterior C1–2 fixation and fusion can be selected for OO without myelopathy or instability.[1] OO complicated with irreducible atlantoaxial dislocation (AAD) is a candidate for occipitocervical fixation and fusion with selective C1 laminectomy or ventral decompression followed by posterior atlantoaxial fixation.[10,13] Transoral decompression surgery risks more serious complications,[14] whereas occipitocervical fixation

Key words
- Atlantoaxial dislocation
- Axial loosening
- Irreducible atlantoaxial dislocation
- Os odontoideum
- Posterior fixation

Abbreviations and Acronyms
AAD: Atlantoaxial dislocation
CCA: Clivus-canal angle
CMA: Cervicomedullary angle
CT: Computed tomography
JOA: Japanese Orthopaedic Association
OO: Os odontoideum
SAC: Space available for the cord
VAS: Visual analog scale

Department of Spinal Surgery, Henan Provincial People's Hospital, Henan, China

To whom correspondence should be addressed: Yan Zheng Gao, M.D. [E-mail: doctorgao63@126.com]

Citation: World Neurosurg. (2019) 132:e193-e201.
https://doi.org/10.1016/j.wneu.2019.08.208

Journal homepage: www.journals.elsevier.com/world-neurosurgery

Available online: www.sciencedirect.com


A
B
C
D
E
F
G
H
I

restricts the motion of the atlanto-occipital joint and has a greater impact on function in patients.[15]

We found that OO combined with irreducible AAD could be partially reduced by heavy-weight skull traction under general anesthesia. Therefore, we performed posterior screw-rod fixation in patients with atlantoaxial instability, and heavy-weight skull traction-assisted loosening of the lateral atlantoaxial joints (we named "axial loosening") in patients with OO complicated with irreducible AAD during a posterior procedure, followed by C1—2 screw-rod fixation for the purpose of treating OO via a single posterior approach.

METHODS

General Information

We retrospectively analyzed a consecutive series of patients diagnosed with OO and treated surgically in our hospital from January 2012 to March 2017. The inclusion criteria were patients diagnosed with OO with atlantoaxial instability or dislocation who underwent surgical treatment. The diagnosis of OO relied on x-ray and/or computed tomography (CT) scans to detect ossicles with smooth cortical margin separated from the axis, with hypertrophy of the atlas anterior arch[16] and a positive jigsaw sign[17] as references. Exclusion criteria were 1) a definite history of an odontoid process fracture; 2) imaging findings of an odontoid process fracture, which is characterized by normal size and shape of the odontoid process with a small separation from the axis and an irregular sclerotic margin of the old fracture[1,18]; and 3) incomplete imaging data and follow-up. This study was approved by the institutional review board of our hospital, and all patients signed written informed consent before treatment.

Operative Process

In the prone position under general anesthesia, Gardner-Wells skull traction tongs (Qingniu Company, Suzhou, China) were used to perform skull traction with a weight of one-sixth of the body weight of the patient. A posterior median incision was made from the base of the skull to C4. After the exposure of the posterior arch of C1 and lamina of C2, the Harms technique[19] was used for fixation. Two lateral mass and 2 pedicle screws (DePuy Synthes, Raynham, Massachusetts, USA) were implanted into the atlas and axis, respectively.

For the patients with complete reduction in dynamic x-ray or under heavy-weight skull traction, 2 rods of appropriate length were placed for connection of the C1—C2. The posterior arch of the atlas and lamina of the axis were decorticated for bone graft. Morselized cancellous bone harvested from the posterior superior iliac spinous was autografted onto the grafting bed. A typical case is illustrated in Figure 1.

For the patients who could not be completely reduced under heavy-weight skull traction, the dislocation was reduced by loosening the lateral atlantoaxial joints posteriorly. The posterior side of the lateral atlantoaxial joint was exposed via subperiosteal dissection from the isthmus of the axis toward the posterior articular capsule of the lateral atlantoaxial joints according to Goel.[20] The loosening of the joints was completed by the axial distraction of the joints with a 7-mm-wide osteotome (Qingniu Company) inserted into the facet joints. We named the loosening of the lateral atlantoaxial joint under heavy-weight skull traction via a posterior approach as "posterior axial loosening." The other steps were the same as previously mentioned. A typical case is illustrated in Figure 2.

The drainage tube was removed within 48 hours postoperatively and x-ray, CT, and magnetic resonance imaging of the cervical spine were ordered. External fixation with a collar was recommended for a period of 1 month. After removal of the collar, neck flexion, extension, and rotation exercises were permitted. X-ray or CT was used to check the reduction and evaluate the fusion status. The recovery of clinical symptoms was assessed at every follow-up evaluation.

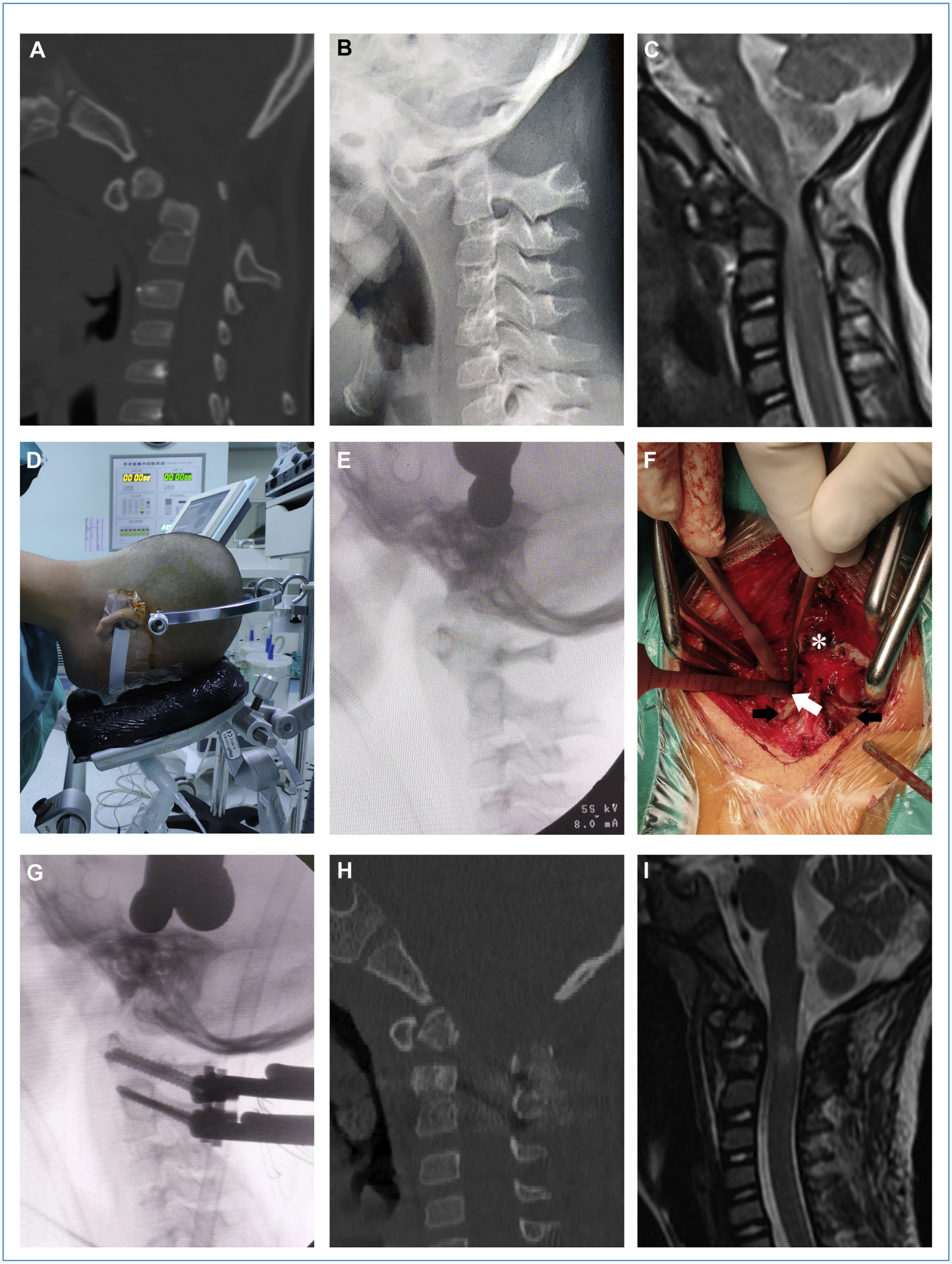
Clinical Manifestations and Surgical Data

According to the Rowland et al.[21] classification, clinical manifestations were recorded in the first group with only local neck pain and no myelopathy. The second group had transient myelopathy mainly caused by minor trauma. The third group had progressive myelopathy caused by spinal cord compression. The fourth group had dizziness or vertigo caused by posterior cerebral ischemia. Considering that the etiology of OO may be related to remote trauma and its clinical manifestations may be induced by trauma, remote and recent trauma are recorded respectively. Remote trauma is defined as a trauma occurring in the first 3 years of age,[5] and recent trauma as any trauma that caused the aforementioned manifestations. The operative time, blood loss volume, and complications for all cases in the series were recorded.

Radiographic Evaluation

Pre- and postoperative space available for the cord (SAC), clivus-canal angle (CCA), and cervicomedullary angle (CMA) were obtained by preoperative and most recent available images. The measurements were completed in the Carestream software (Carestream Health Inc., Rochester, New York, USA) by the first author. Each case was measured 3 times, and the average value was obtained for further analysis. Measurement methods are presented in Figure 3. Continuous callus observed on lateral x-ray or sagittal reconstructive CT scans of the cervical spine was defined as a successful fusion.

Figure 1. Case 1, a woman aged 63 years with atlantoaxial instability complicated with myelopathy caused by os odontoideum (OO). (**A**) Sagittal reconstructive computed tomography (CT) of cervical spine presented the OO with anterior atlantoaxial instability. (**B**) Posterior atlantoaxial instability demonstrated by hyperextension x-ray. (**C**) Upper cervical spinal cord was compressed by the superior posterior part of axis based on magnetic resonance imaging (MRI). (**D**) Postoperative reconstructive CT indicated an anatomic reduction of upper cervical spine and sufficient autograft bone. (**E**) Postoperative x-ray showed atlantoaxial fixation by screw and rod. (**F**) Postoperative MRI indicated release of upper cervical spinal cord compression. Solid fusion confirmed by CT scan (**G**) and hyperextension (**H**) and hyperflexation (**I**) x-ray 4 months after the C1-C2 fixation without axial loosening.


A
B
C
D
E
F
G
H
I
*
55 kV
8.0 mA

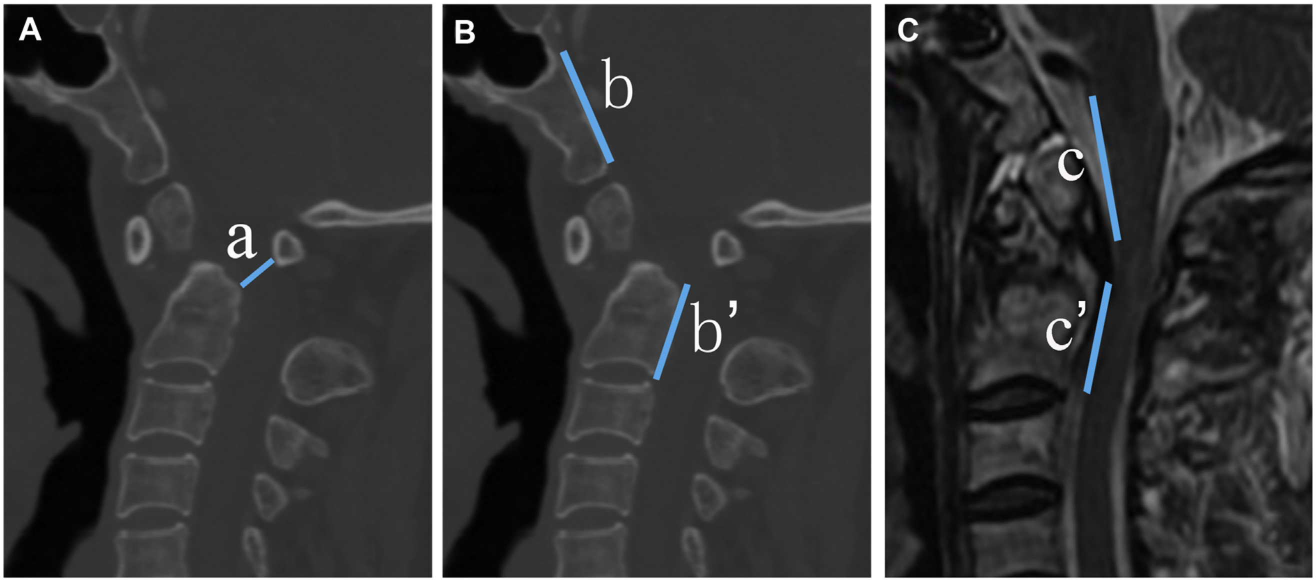


Figure 3. (**A**) The distance from the superior posterior point of axis to the nearest bony structure of posterior arch of atlas or lamina of axis (line a) was defined as space available for the cord. (**B**) The crossing angle of the line of clivus (line b) and the line extending the posterior border of the axis body (line b′) was defined as the clivus-canal angle. (**C**) The crossing angle of the line extending the anterior border of the ventral medulla (line c) and the line extending the anterior border of the ventral upper cervical spinal cord (line c′) in midline sagittal T2 magnetic resonance imaging was defined as the cervicomedullary angle.

Clinical Recovery

The visual analog scale (VAS) of cervical pain (cervical pain positive preoperatively) and the Japanese Orthopaedic Association (JOA) score (myelopathy positive preoperatively) were evaluated preoperatively and at the final follow-up. The improvement rate of the JOA score in the final follow-up was calculated using the formula as follows:

$$[(\text{postoperative JOA score} - \text{preoperative JOA score}) / (17 - \text{preoperative JOA score})] \times 100\%$$

Statistical Analysis

SPSS software version 20.0 (IBM Corp., Armonk, New York, USA) was used for statistical analysis. Descriptive statistics were used for enumeration data, means ± standard deviations were calculated for measurement data. Paired *t* tests were used for the statistical analysis of preoperative and postoperative imaging measurements and clinical recovery of the total series, and independent *t* tests were used for the statistical analysis of surgical data of the 2 different procedures. Two-way repeated measures analysis of variance tests were used to compare the preoperative and postoperative imaging data and clinical recovery data between the posterior screw-rod fixation with and without axial loosening. A level of $P < 0.05$ was established as the level of significance.

RESULTS

Clinical Data

There were 41 patients (15 men and 26 women) with an average age of 40.6 ± 21.7 years. The follow-up period ranged from 24–65 months, with an average of 40.7 ± 21.7 months. Nineteen patients (46.3%) had a certain history of trauma, 5 with remote trauma, 4 with no remote trauma, 32 with an unclear history, 15 (36.6%) with

Figure 2. Case 2, a girl aged 6 years with irreducible atlantoaxial dislocation (AAD) complicated with myelopathy caused by os odontoideum (OO) and Down syndrome. (**A**) Sagittal reconstructive computed tomography (CT) of cervical spine presented the OO with anterior atlantoaxial instability. (**B**) The dislocation was not reduced under hyperextension x-ray. (**C**) Upper cervical spinal cord was compressed by the superior posterior part of axis based on magnetic resonance imaging (MRI). (**D**) and (**E**) AAD was partially reduced under heavy-weight skull traction under general anesthesia. (**F**) Posterior loosening of the lateral atlantoaxial joints was completed by the axial distraction of the joints with a 7-mm-wide osteotome (*white arrow*), *represented the posterior arch of atlas, and black arrows represented the lamina of axis. (**G**) Screw and rod fixation after the axial loosening. (**H**) Postoperative reconstructive CT indicated an anatomic reduction of upper cervical spine and sufficient autograft bone. (**I**) Postoperative MRI indicated release of upper cervical spinal cord compression.

Table 1. General Data of the Patients (n = 41)

General Data	Value
Age, years	
Mean ± SD	40.6 ± 21.7
Sex, n (%)	
Male	15 (36.6%)
Female	26 (63.4%)
Trauma history, n (%)	
Trauma positive	19 (46.3%)
No trauma	22 (53.7%)
Remote trauma, n (%)	
Definite history	5 (12.2%)
No remote trauma	4 (9.8%)
Unclear	32 (78.0%)
Recent trauma, n (%)	
Recent trauma positive	15 (36.6%)
No recent trauma	26 (63.4%)
Symptoms, n (%)	
Local symptoms	33 (80.5%)
Transient myelopathy	13 (31.7%)
Progressive myelopathy	21 (51.2%)
Posterior circulation	12 (29.3%)

SD, standard deviation.

recent trauma, and 1 with a history of both remote and recent trauma.

Localized neck pain was found in 33 patients (80.5%), transient myelopathy in 13 (31.7%), progressive myelopathy in 21 (51.2%), and posterior cerebral symptoms in 12 (29.3%). Five cases (12.2%) had congenital dysplasia, including 2 with Down syndrome, 1 with vertebral dysplasia, 1 with posterior arch dysplasia of the atlas, and 1 with a right lateral mass dysplasia of the atlas. Details are shown in Table 1.

Surgical Results

All 41 patients underwent reduction and C1—C2 screw-rod fixation via a posterior approach. Thirty-two cases were defined as atlantoaxial instability based on preoperative dynamic x-ray, and 3 were defined as reducible AAD based on the successful reduction under heavy-weight skull traction in the operation.[22] For atlantoaxial instability and reducible AAD, screw-rod fixation and fusion without axial loosening was performed. Six cases of irreducible AAD were successfully reduced after axial loosening followed by C1—C2 screw-rod fixation and fusion.

Blood loss volume was 232.8 ± 61.9 mL, and operation time was 165.5 ± 28.1 minutes in the total series. Blood loss volume and operation time in the fixation with and without axial loosening were 247.3 ± 50.9 mL and 163.5 ± 32.7 minutes, and 232.8 ± 61.9 mL and 165.5 ± 28.1 minutes, respectively. Independent t tests confirm that there were no differences between the with and without axial loosening groups ($t = 0.618$ and $P = 0.540$ and $t = 0.183$ and $P = 0.856$, respectively). Detailed results are shown in Table 2.

Pain of the greater occipital nerve appeared in 2 cases of direct fixation on the second postoperative day, which was gradually relieved after oral gabapentin treatment for 1 week. Imperfect incision healing of the bone harvesting area appeared in 2 cases. One of the 2 cases was from the axial loosening group and was diagnosed with a superficial infection of *Staphylococcus aureus*. The wound healed after intravenous injection of cefazolin and a wound dressing change. The other case from the direct fixation group was diagnosed with deep cavity formation and healed after revision surgery for debridement and suturing. There were no cases of spinal cord injury, vertebral artery injury, or cerebrospinal fluid leakage.

Imaging Results

Preoperative SAC, CCA, and CMA were 10.56 ± 2.33 mm, 127.84 ± 9.82°, and 138.06 ± 7.01°, whereas the latest measurements of SAC, CCA, and CMA were 15.43 ± 1.70 mm, 145.37 ± 10.55°, and 154.21 ± 6.51°, with t values of 13.420, 9.026, and 11.892, respectively, based on paired t tests. Pre- and postoperative radiographic data showed a significant difference ($P < 0.001$). There were significant differences of pre- and postoperative SAC, CCA, and CMA in separate procedures (F value was 93.772, 31.942, and 89.898, respectively; $P < 0.001$), and there were no differences in the change of SAC and CMA (F value was 0.528 and 0.408,

Table 2. Comparison of the Clinical Data Between Posterior Fixation With (n = 6) and Without (n = 35) Axial Loosening

Groups	Age (years)	Blood Loss (mL)	Operative Time (minutes)	Fusion Time (month)	JOA Score Recovery Rate (%)
Fixation with axial loosening	46.0 ± 24.6	247.3 ± 50.9	163.5 ± 32.7	4.0 ± 1.1	83.11 ± 18.79
Fixation without axial loosening	39.7 ± 21.1	230.3 ± 63.8	165.8 ± 27.7	4.5 ± 1.1	70.74 ± 24.12, n = 28
t	0.648	0.618	0.183	1.078	1.177
P	0.521	0.540	0.856	0.288	0.248
Mean of total series, n = 41	40.6 ± 21.7	232.8 ± 61.9	165.5 ± 28.1	4.5 ± 1.1	72.92 ± 23.16

Independent *t* test was used to compare the operative data between the posterior fixation with and without axial loosening; $P < 0.05$ was defined as statistical difference.
JOA, Japanese Orthopaedic Association.

Table 3. Comparison of Radiographic Data Between Posterior Fixation With (n = 6) and Without (n = 35) Axial Loosening

Groups	Time Point	SAC (mm)	CCA (°)	CMA (°)
Fixation with axial loosening	Preoperation	10.84 ± 3.54	123.80 ± 9.10	137.07 ± 6.03
	Latest image	16.08 ± 1.78	136.90 ± 6.12	157.71 ± 8.75
Fixation without axial loosening	Preoperation	10.51 ± 2.13	127.83 ± 9.94	138.23 ± 7.23
	Latest image	15.32 ± 1.69	146.83 ± 10.52	153.61 ± 6.00
Intergroups comparison	F value	0.528	4.334	0.408
	P value	0.472	0.044	0.527
Pre- and postoperative comparison	F value	93.772	31.942	89.898
	P value	<0.001	<0.001	<0.001
Cross action between time point and groups	F value	0.175	1.079	1.918
	P value	0.678	0.305	0.174
Mean of total series, n = 41	Preoperation	10.56 ± 2.33	127.24 ± 9.82	138.06 ± 7.01
	Latest image	15.43 ± 1.70	145.37 ± 10.55	154.21 ± 6.51
Pre- and postoperative comparison	t value	13.420	9.026	11.892
	P value	<0.001	<0.001	<0.001

Two-way repeated measures analysis of variance was used to compare the radiographic data between the posterior fixation with and without axial loosening; $P < 0.05$ was defined as statistical difference.

SAC, space available for the cord; CCA, clivus-canal angle; CMA, cervicomedullary angle.

respectively; P = 0.472 and P = 0.527, respectively) and for the difference in the change of CCA (F value was 4.334; P = 0.044) between the fixation with and without axial loosening. No interactions of SAC, CCA, and CMA were detected in the groups of fixation with and without axial loosening (F values 0.175, 1.079, and 1.918, respectively; P = 0.678, P = 0.305, and P = 0.174, respectively). Detailed results are shown in Table 3.

The fusion time ranged from 3–6 months with an average of 4.5 ± 1.1 months. The fusion rate was 100%. There were 3 cases with partial resorption of posterior bone graft without failure of hardware or loss of reduction.

Clinical Recovery

The VAS scores of 33 patients with neck pain decreased from 2.3 ± 1.4 preoperatively to 1.3 ± 1.1 at the final follow-up, and paired t tests showed a significant difference (t = 2.966; P = 0.006). The JOA scores of 34 patients with myelopathy were 11.8 ± 2.4 preoperatively and 15.7 ± 1.1 at the final follow-up, and paired t tests showed a significant difference (t = 9.843; P < 0.001).

There were significant differences of pre- and postoperative VAS and JOA scores in separate procedures based on 2-way analysis of variance (F value was 13.770 and 73.226, respectively; P = 0.001 and P < 0.001, respectively), and there were no differences in the change of VAS and JOA scores (F value was 0.040 and 0.076, respectively; P = 0.843 and P = 0.784, respectively) between the fixation with and without axial loosening. Interaction of VAS score was detected (F = 4.458; P = 0.043) and no interaction of JOA score was detected in the groups of fixation with and without axial loosening (F = 2.073; P = 0.160). Detailed results are shown in Table 4.

The improvement rate of JOA score was 72.92 ± 23.16% in the total series, 70.74 ± 24.12% in the nonaxial loosening group, and 83.11 ± 18.79% in the axial loosening group. There was no difference between the 2 procedures by independent *t* test (t = 1.177; P = 0.248). Detailed results are shown in Table 2.

In 12 patients with posterior cerebral circulation symptoms, 9 patients (75%) had symptom resolution of the dizziness or vertigo, and 3 cases (25%) still had intermittent dizziness.

DISCUSSION

Etiology of OO

It has been elucidated that patients with OO have higher rates of congenital lesions such as Down syndrome, Klippel-Feil syndrome, epiphyseal dysplasia, and other bone dysplasia than the general population.[23] Kirlew et al.[8] studied female twins without trauma and found that there were partial fusions of C2–3, hypertrophy of the C1 anterior arch, and depression of the C2 spinous process. Straus et al.[24] studied identical twins with OO, patients with OO who were not twins, and 4 people without OO. By comparing differentially expressed genes, they found 6 differentially expressed genes, most of which were related to bone formation.

There is also literature supporting the trauma theory.[7,18,25,26] Fielding and Griffin[4] believed that the cause of OO is damage to the blood supply caused by trauma. It is noteworthy that the injuries in these patients with OO occurred before the age of 3 years, at which time the cartilage plate had not yet been closed.[5]

There were 5 cases of congenital malformation and 5 of remote trauma in our study. However, only 4 cases of remote trauma were confirmative, whereas the remaining 32 were uncertain as to whether they had experienced remote trauma or not. Therefore, we believe that the number of patients with remote trauma may be underestimated. Based on this series of cases, we believe that congenital development and remote trauma are independent causes of OO.

Treatment Methods and Surgical Indications

There is a consensus that patients with neurologic symptoms are candidates for surgical treatment. However, there is controversy about the treatment of incidentally discovered OO with or without instability.[10,14,27-29] Clinical and radiologic surveillance were recommended for OO without myelopathy according to the guidelines of the Congress of Neurological Surgeons for the diagnosis and treatment of OO in 2002.[13] However, the concept of treatment for this condition has changed in recent years. Klimo et al.[28,30] believe that surgical treatment should be performed to prevent catastrophic spinal cord injury. It has been reported that ligament relaxation with aging can lead to chronic instability,[31] and Ni et al.[32] believe that OO with chronic instability can cause hypertrophy of soft tissue around the odontoid process, which may compress the spinal cord ventrally. Therefore, the guidelines

Table 4. Comparison of Clinical Outcome Between Posterior Fixation With and Without Axial Loosening

Groups	Time Point	VAS Score	JOA Score
Fixation with axial loosening	Preoperation	3.2 ± 1.8, n = 5	11.0 ± 3.3, n = 6
	Final follow-up	0.6 ± 0.9, n = 5	16.2 ± 0.8, n = 6
Fixation without axial loosening	Preoperation	2.2 ± 1.3, n = 28	11.9 ± 2.2, n = 28
	Final follow-up	1.5 ± 1.1, n = 28	15.6 ± 1.1, n = 28
Intergroups comparison	F value *P* value	0.040 0.843	0.076 0.784
Pre- and postoperative comparison	F value *P* value	13.770 0.001	73.226 <0.001
Cross action between time point and groups	F value *P* value	4.458 0.043	2.073 0.160
Mean of total series	Preoperation	2.2 ± 1.4, n = 33	11.8 ± 2.4, n = 34
	Final follow-up	1.3 ± 1.1, n = 33	15.7 ± 1.1, n = 34
Pre- and postoperative comparison	t value	2.966	9.843
	P value	0.006	<0.001

Two-way repeated measures analysis of variance was used to compare the clinical outcomes between the posterior fixation with and without axial loosening. Paired *t* test was used to compare preoperative and postoperative VAS and JOA scores of the total series; $P < 0.05$ was defined as statistical difference.
VAS, visual analog scale; JOA, Japanese Orthopaedic Association.

of the Congress of Neurological Surgeons in 2013[1] suggested that for incidentally discovered OO, both surveillance and surgical treatment are appropriate.

In this group, 15 cases had received trauma, which caused neck pain, transient myelopathy, and progressive myelopathy. Because of worsening symptoms after trauma and the concern for progressive instability with aging, we recommend surgical treatment once OO is diagnosed.

Surgical Treatment of OO

Surgical treatment of OO was variable depending on the type of instability or dislocation and the location of compression. Posterior fixation was recommended for atlantoaxial instability, and anterior decompression followed by posterior fixation can be considered if there is ventral compression in irreducible AAD.[1,23] Anterior reduction and fixation for irreducible AAD have also been reported.[33] However, the anterior reduction is technique demanding[14] and includes a high risk of infection when followed by anterior fixation.

Most of OO cases present with atlantoaxial instability and can be considered for a posterior fixation without anterior reduction before surgery. Patients with AAD that can be reduced under skull traction are defined as reducible AAD, and as irreducible AAD if they cannot be reduced under skull traction.[34] However, the former defined as irreducible AAD under conscious skull traction may be reduced under general anesthesia,[35] and should be defined as reducible AAD according to Shenglin et al.[22] We define irreducible AAD according to this more rigorous classification system in our case series.

The true proportion of OO complicated with irreducible AAD remains unclear. Klimo et al.[28] reported 78 cases of OO in 2008 and did not find "fixed" dislocation according to dynamic x-ray. The study with a series of 32 patients by Huang et al.[36] was aimed at atlantoaxial instability. The study with a series of 279 cases of OO,[23] the largest series published to date, reported transoral reduction and posterior fixation for the treatment of irreducible AAD; however, the authors did not present the proportion of irreducible AAD cases. According to our series of 41 patients, 6 patients (14.6%) were defined as irreducible AAD referring to the Shenglin et al.[22] classification.

Posterior reduction and fixation for treating irreducible AAD had been reported before[20,37]; however, we insist that the posterior technique is not suitable for all types of irreducible AAD. According to the Shenglin et al.[22] classification, failure of reduction under heavy-weight skull traction under general anesthesia is defined as irreducible dislocation. We found distinctive characteristics of OO complicated with irreducible AAD, which could be partially reduced (reduction rate >50% based on atlantodental interval) under heavy-weight skull traction under general anesthesia. This type of irreducible AAD could be anatomically reduced by axial loosening via a single posterior approach as we presented. We would recommend transoral or anterior retropharyngeal reduction and posterior fixation for AAD, which cannot be reduced or reduced <50% under heavy-weight skull traction for obtaining anatomic reduction. However, this kind of irreducible AAD is mostly found in occipitalization of the atlas or basilar invagination.[38]

For most patients with OO, atlantoaxial fixation is appropriate, and occipitocervical fusion is recommended in the cases with upper cervical stenosis in which posterior atlantoaxial decompression is needed, or in cases with dystopic OO that is characterized by the fusion of OO and the basion.[39] This series is consistent with the previous study by Huang et al,[36] in which no upper cervical stenosis or dystopic OO was found, thus all patients were treated with atlantoaxial fixation, which maximally preserves the motion of the spine, even in the case of lateral mass dysplasia of the atlas, in which the implantation of a lateral mass screw was successfully completed under the guidance of navigation.

Limitations

This study is a retrospective analysis, and because of the characteristics of OO, only 6 cases with irreducible AAD underwent axial loosening. It is permissible to compare the selective axial loosening technique with anterior reduction and posterior fixation with screws and rod for treating OO complicated with irreducible AAD in the future.

CONCLUSIONS

Most AAD caused by OO are characterized as atlantoaxial instability or reducible AAD. Posterior screw-rod fixation and selective axial loosening are appropriate for treating OO complicated with atlantoaxial instability or dislocation (including reducible and irreducible variants) without the need for anterior decompression.

REFERENCES

1. Rozzelle CJ, Aarabi B, Dhall SS, et al. Os odontoideum. *Neurosurgery*. 2013;72:159-169.
2. McHugh BJ, Grant RA, Zupon AB, DiLuna ML. Congenital os odontoideum arising from the secondary ossification center without prior fracture. *J Neurosurg Spine*. 2012;17:594-597.
3. Flemming C, Hodson CJ. Os odontoideum; a congenital abnormality of the axis; case report. *J Bone Joint Surg Br*. 1955;37-B:622-623.
4. Fielding JW, Griffin PP. Os odontoideum: an acquired lesion. *J Bone Joint Surg Am*. 1974;56:187-190.
5. Hawkins RJ, Fielding JW, Thompson WJ. Os odontoideum: congenital or acquired. A case report. *J Bone Joint Surg Am*. 1976;58:413-414.
6. Ricciardi JE, Kaufer H, Louis DS. Acquired os odontoideum following acute ligament injury. Report of a case. *J Bone Joint Surg Am*. 1976;58:410-412.
7. Schuler TC, Kurz L, Thompson DE, et al. Natural history of os odontoideum. *J Pediatr Orthoped*. 1991;11:222-225.
8. Kirlew KA, Hathout GM, Reiter SD, Gold RH. Os odontoideum in identical twins: perspectives on etiology. *Skeletal Radiol*. 1993;22:525-527.
9. Matsui H, Imada K, Tsuji H. Radiographic classification of os odontoideum and its clinical significance. *Spine (Phila Pa 1976)*. 1997;22:1706-1709.
10. Spierings EL, Braakman R. The management of os odontoideum. Analysis of 37 cases. *J Bone Joint Surg Br*. 1982;64:422-428.
11. Shirasaki N, Okada K, Oka S, et al. Os odontoideum with posterior atlantoaxial instability. *Spine (Phila Pa 1976)*. 1991;16:706-715.
12. Kikuchi K, Nakagawa H, Watanabe K, Kowada M. Bilateral vertebral artery occlusion secondary to atlantoaxial dislocation with os odontoideum: implication for prophylactic cervical stabilization by fusion-case report. *Neurol Med Chir*. 1993;33:769-773.
13. Hadley MN, Walters BC, Grabb PA, et al. Os odontoideum. *Neurosurgery*. 2002;50:S148-S155.
14. Menezes AH. Pathogenesis, dynamics, and management of os odontoideum. *Neurosurg Focus*. 1999;6:e2.
15. Kim IS, Hong JT, Jang WY, et al. Surgical treatment of os odontoideum. *J Clin Neurosci*. 2011;18:481-484.
16. Holt RG, Helms CA, Munk PL, Gillespy TR. Hypertrophy of C-1 anterior arch: useful sign to distinguish os odontoideum from acute dens fracture. *Radiology*. 1989;173:207-209.
17. Fagan A, Askin G, Earwaker JS. The jigsaw sign. A reliable indicator of congenital aetiology in os odontoideum. *Eur Spine J*. 2004;13:295-300.
18. Verska JM, Anderson PA. Os odontoideum. A case report of one identical twin. *Spine (Phila Pa 1976)*. 1997;22:706-709.
19. Harms J, Melcher RP. Posterior C1-C2 fusion with polyaxial screw and rod fixation. *Spine (Phila Pa 1976)*. 2001;26:2467-2471.
20. Goel A. Treatment of basilar invagination by atlantoaxial joint distraction and direct lateral mass fixation. *J Neurosurg Spine*. 2004;1:281-286.
21. Rowland LP, Shapiro JH, Jacobson HG. Neurological syndromes associated with congenital absence of the odontoid process. *AMA Arch Neurol Psychiatry*. 1958;80:286-291.
22. Shenglin W, Chao W, Ming Y, Haitao Z, Gengting D. Novel surgical classification and treatment strategy for atlantoaxial dislocations. *Spine*. 2013;38:1348-1356.
23. Deng Z, Shenglin W, Passias PG, Chao W. Craniocervical instability in the setting of os odontoideum: assessment of cause, presentation, and surgical outcomes in a series of 279 cases. *Neurosurgery*. 2015;76:514-521.
24. Straus D, Xu S, Traynelis V. Os odontoideum in identical twins: comparative gene expression analysis. *Surg Neurol Int*. 2014;5:37.
25. Zygourakis CC, Cahill KS, Proctor MR. Delayed development of os odontoideum after traumatic cervical injury: support for a vascular etiology. *J Neurosurg Pediatr*. 2011;7:201-204.
26. Wada E, Matsuoka T, Kawai H. Os odontoideum as a consequence of a posttraumatic displaced ossiculum terminale. *J Bone Joint Surg Am*. 2009;91:1750-1754.
27. Wilson JR, Dettori JR, Vanalstyne EM, Fehlings MG. Addressing the challenges and controversies of managing os odontoideum: results of a systematic review. *Evid Based Spine Care J*. 2010;1:67-74.
28. Klimo PJ, Kan P, Rao G, Apfelbaum R, Brockmeyer D. Os odontoideum: presentation, diagnosis, and treatment in a series of 78 patients. *J Neurosurg Spine*. 2008;9:332-342.
29. Arvin B, Fournier-Gosselin M, Fehlings MG. Os odontoideum: etiology and surgical management. *Neurosurgery*. 2010;66:A22-A31.
30. Klimo JP, Coon V, Brockmeyer D. Incidental os odontoideum: current management strategies. *Neurosurg Focus*. 2011;31:E10.
31. Dyck P. Os odontoideum in children: neurological manifestations and surgical management. *Neurosurgery*. 1978;2:93-99.
32. Ni B, Zhou F, Xie N, et al. Transarticular screw and C1 hook fixation for os odontoideum with atlantoaxial dislocation. *World Neurosurg*. 2011;75:540-546.
33. Wang X, Fan C, Liu Z. The single transoral approach for os odontoideum with irreducible atlantoaxial dislocation. *Eur Spine J*. 2010;19:91-95.
34. Bach CM, Arbab D, Thaler M. Treatment strategies for severe C1C2 luxation due to congenital os odontoideum causing tetraplegia. *Eur Spine J*. 2013;22:29-35.
35. Visocchi M, Fernandez E, Ciampini A, Di Rocco C. Reducible and irreducible os odontoideum in childhood treated with posterior wiring, instrumentation and fusion. Past or present? *Acta Neurochir*. 2009;151:1265-1274.
36. Huang DG, Wang T, Hao DJ, et al. Posterior C1-C2 screw-rod fixation and autograft fusion for the treatment of os odontoideum with C1-C2 instability. *Clin Neurol Neurosurg*. 2017;163:71-75.
37. Jian FZ, Chen Z, Wrede KH, Samii M, Ling F. Direct posterior reduction and fixation for the treatment of basilar invagination with atlantoaxial dislocation. *Neurosurgery*. 2010;66:678-687 [discussion: 687].
38. Srivastava SK, Aggarwal RA, Nemade PS, Bhosale SK. Single-stage anterior release and posterior instrumented fusion for irreducible atlantoaxial dislocation with basilar invagination. *Spine J*. 2016;16:1-9.
39. Jumah F, Alkhdour S, Mansour S, et al. Os odontoideum: a comprehensive clinical and surgical review. *Cureus*. 2017;9:e1551.

Conflict of interest statement: This work is supported by Overseas Research Project for Health Care and Family Planning Talents of Henan Province (grant number 2018090).

Received 10 May 2019; accepted 26 August 2019

Citation: World Neurosurg. (2019) 132:e193-e201.
https://doi.org/10.1016/j.wneu.2019.08.208

Journal homepage: ***www.journals.elsevier.com/world-neurosurgery***

Available online: ***www.sciencedirect.com***