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ABOUT COVER

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Single door laminoplasty plus posterior fusion for posterior atlantoaxial dislocation with congenital malformation: A case report and review of literature

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Abstract

BACKGROUND

Posterior atlantoaxial dislocation (PAD) is a rare type of upper cervical spine disease. We sought to describe a unreported case of old PAD with os odontoideum (OO) and atlas hypoplasia (AH) and our unique treatment approach consisting of C1 single door laminoplasty with C1-3 posterior fixation and fusion.

CASE SUMMARY

A 70-year-old male patient who suffered from progressive aggravating numbness and limb weakness for 4 years without trauma, was diagnosed with old PAD with OO and AH. The patient underwent closed reduction and C1 single door laminoplasty with C1-3 posterior fixation and fusion instead of C1 laminectomy with occipitocervical fusion. During the 3-year follow-up, he was able to walk by himself instead of using a wheelchair and with a $\pm 25^\circ$ range of head rotation as well as a $\pm 10^\circ$ range of flexion-extension. Three-year follow-up images showed satisfactory reduction and fusion.

CONCLUSION

C1 single door laminoplasty with cervical fusion in PAD combined with spinal cord compression could be a suitable and effective surgical option. Compared with laminectomy and occipitocervical fusion, it retains more cervical range of motion, has a smaller incision and provides an adequate bone grafting space for atlantoaxial fusion.

Key Words: Posterior atlantoaxial dislocation; C1 laminoplasty; Os odontoideum; Atlas hypoplasia; Case report

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Core Tip: This article describes an unreported case of old posterior atlantoaxial dislocation with os odontoideum and atlas hypoplasia. C1 single door laminoplasty with C1-3 posterior fixation and fusion was performed as surgical treatment, which retained partial range of motion, decreased operative trauma and provided an adequate bone grafting space for atlantoaxial fusion compared with laminectomy and occipitocervical fusion.

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INTRODUCTION

Posterior atlantoaxial dislocation (PAD) is a rare disease which only accounts for approximately 0.3% of all cervical spine injuries and 1.7% of upper cervical spine injuries^[1]. To date, only 29 cases of PAD of several types have been reported, all of which have been due to traumatic accidents^[2-29]. Herein, a unreported case of old PAD with os odontoideum (OO) and atlas hypoplasia (AH) is introduced, as well as our unique treatment approach consisting of C1 single door laminoplasty with C1-3 posterior fixation and fusion.

CASE PRESENTATION

Chief complaints

A 70-year-old male patient suffered from progressive aggravating numbness and weakness of limbs.

History of present illness

The patient's symptoms started 5 years ago without any trauma or accident. When admitted to our hospital, he could only move using a wheelchair.

History of past illness

The patient had no previous relevant medical history.

Personal and family history

The patient had no previous relevant family history.

Physical examination

The results of physical examination revealed that the grip strength in both hands was grade 3, the muscle tension of both lower limbs was high, bilateral Hoffman sign was positive, knee and ankle reflexes were hyperactive, and ankle clonus was positive.

Laboratory examinations

Laboratory examinations showed no obvious deficits.

Imaging examinations

Considering that there was no history of trauma, and the borders of the odontoid free body and basis were smooth and rounded, an old PAD combined with OO was diagnosed based on computed tomography (CT) results (Figure 1A). The C1 inner sagittal diameter was 22.51 mm (Figure 1B) which supported the diagnosis of AH^[30]. The results of magnetic resonance imaging (MRI) (Figure 1C) showed that the spine cord was constricted by the odontoid process and the signal of the spine had changed at the level of atlantoaxial dislocation.

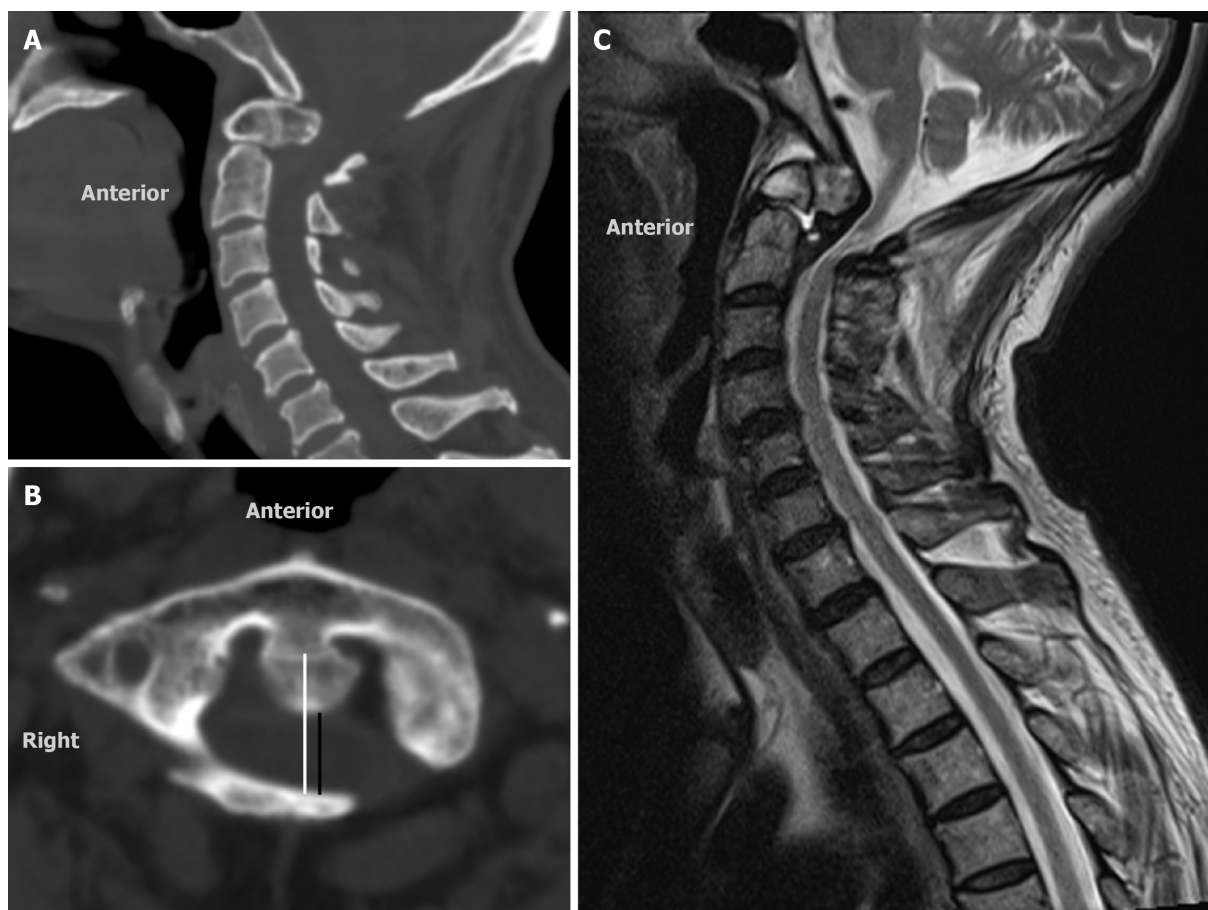


Figure 1 Preoperative images. A: Parasagittal computed tomography (CT) showing the posterior atlantoaxial dislocation combined with os odontoideum; B: Axial CT scan, demonstrating the C1 inner sagittal diameter (white line) = 22.51 mm and the canal sagittal diameter (black line) = 12.00 mm; C: Parasagittal magnetic resonance imaging.

FINAL DIAGNOSIS

The final diagnosis in the presented case was an old PAD combined with OO and AH.

TREATMENT

Due to the mechanism of posterior dislocation, skull traction was performed under a flexed position of the cervical spine with an initial weight of 3.5 kg and gradually increased to 7.5 kg, and the flexed angle also increased synchronously. Lateral radiographs (Figure 2A-C) were obtained on the fourth day, the eighth day and the twelfth day after skull traction. MRI (Figure 2D) after skull traction demonstrated satisfactory reduction of the dislocation, but the cervical canal was compressed again by the posterior arch of C1 due to AH. Therefore, decompression surgery was necessary.

The surgery was performed under general anesthesia and in the prone position. Cortical somatosensory evoked potential (CSEP) was utilized to monitor the neurological condition. The reference frame of the 3-dimensional navigation system was placed on the spinous process of C4 through an isolated small incision. Then the surgeons inserted 5 pedicle screws in C1, C2 (right side) and C3 under the guidance of the O-arm machine and navigation. The C1 single door laminoplasty was performed with the assistance of a surgical electric grinder and a piezosurgery osteotomy. Bone autograft mixed with bone allograft was placed between C1 and C2 posterior arch as an atlantoaxial fusion. There was no specific medication except routine methylprednisolone, omeprazole, antibiotics and dehydration.

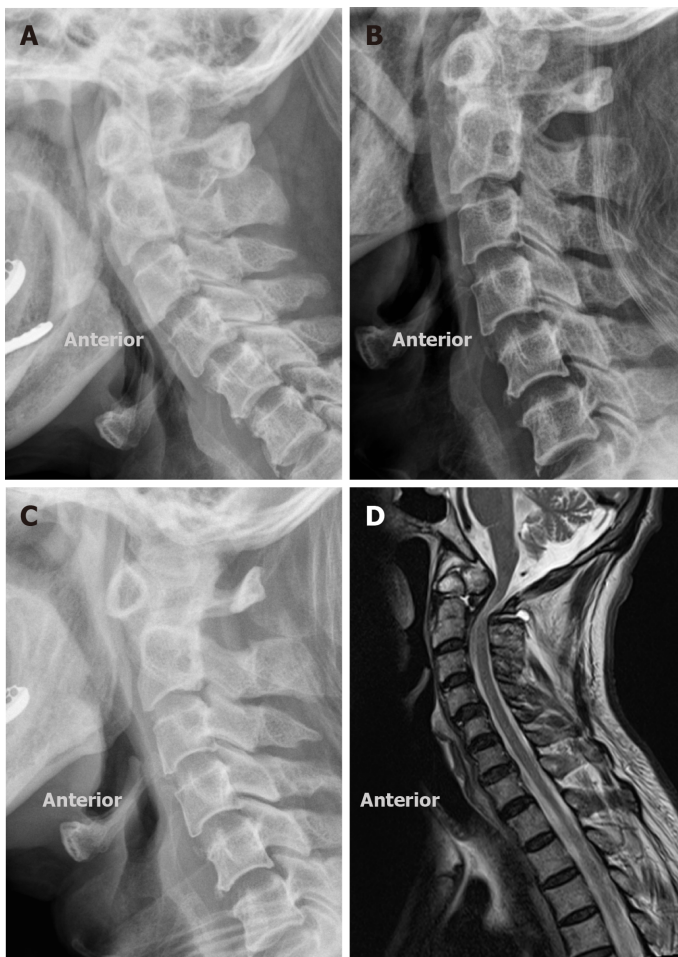


Figure 2 Images of reduction. A: Lateral radiographs 4 d after traction; B: Lateral radiographs 8 d after traction; C: Lateral radiographs 12 d after traction, showing satisfactory closed reduction; D: Post-reductional parasagittal magnetic resonance imaging, demonstrating that the spinal cord was still compressed by C1 posterior arch.

OUTCOME AND FOLLOW-UP

The grip strength in both hands was improved after skull traction and surgery, as well as muscle tension of the lower limbs. Neurological signs including Hoffman, Babinski and ankle clonus returned to normal. During the 3-year follow-up, he was able to walk by himself instead of using a wheelchair and with a $\pm 25^\circ$ range of head rotation as well as a $\pm 10^\circ$ range of flexion-extension. Three-year follow-up images showed satisfactory reduction and fusion (Figure 3).

DISCUSSION

Literature review

A total of 29 PAD patients with or without fracture and neurological deficit were found following a thorough literature review of PubMed, Elsevier, MEDLINE and Web of Science (Table 1). The rarity of this dislocation is mainly due to the anatomical locking structure as the dorsal ligament of the osseous-ligamentous ring is easily damaged compared to the odontoid process and hence anterior atlantoaxial dislocation occurs more frequently^[31]. The treatment methods for PAD include closed reduction (mostly skull traction), open reduction (mostly odontoidectomy), anterior odontoid screw fixation and posterior fusion. Skull traction was first reported in a case of PAD without fracture or neurological deficit by Haralson *et al*^[2] and gradually became the first choice for conservative treatment and preoperative reduction. However, closed reduction was considered to be a risky procedure especially when the patient was under anesthesia. Sud *et al*^[11] reported a case who developed quadriplegia during traction which might lead to transient over-distraction of the spinal cord. Open reduction was considered a second choice after ineffective closed reduction^[13,15-19,23-25,28].

Table 1 Review of previously reported posterior atlantoaxial dislocation cases

No.	Ref.	Patient	Fracture	Neurological deficit	Treatment
1	Haralson <i>et al</i> ^[2] , 1969	30, M	N	N	CR + WF
2	Sassard <i>et al</i> ^[3] , 1974	20, F	N	Y	CR
3	Patzakis <i>et al</i> ^[4] , 1974	37, M	N	N	CR
4	Fox <i>et al</i> ^[5] , 1977	65, M	Y	Y	OR + WF
5	Jamshidi <i>et al</i> ^[6] , 1983	22, M	N	N	CR + WF
6	Autricque <i>et al</i> ^[7] , 1986	45, M	Y	Y	CR
7	Autricque <i>et al</i> ^[7] , 1986	63, M	Y	N	CR + OCF
8	Wong <i>et al</i> ^[8] , 1991	23, M	N	Y	CR + AAF
9	Fujimura <i>et al</i> ^[9] , 1997	54, M	N	N	CR
10	Carroll <i>et al</i> ^[10] , 2001	19, F	N	Y	CR
11	Sud <i>et al</i> ^[11] , 2002	38, M	N	N	OR + SF
12	Neumann <i>et al</i> ^[120] , 2003	22, M	N	N	CR
13	Yoon <i>et al</i> ^[13] , 2003	64, M	N	Y	OR + AAF
14	Chaudhary <i>et al</i> ^[14] , 2008	35, F	N	Y	CR
15	Amirjamshidi <i>et al</i> ^[15] , 2009	31, M	N	Y	OR + AAF
16	Jiang <i>et al</i> ^[16] , 2010	48, M	N	N	OR + SF
17	Zhen <i>et al</i> ^[17] , 2011	44, M	N	N	OR + AAF
18	Zhang <i>et al</i> ^[18] , 2012	38, M	Y	Y	OR ¹
19	Moreau <i>et al</i> ^[19] , 2012	65, M	Y	N	OR + OCF
20	Kambali <i>et al</i> ^[20] , 2013	32, M	N	Y	CR + AAF
21	Riouallon <i>et al</i> ^[21] , 2014	25, M	Y	N	CR + SF
22	Meng <i>et al</i> ^[22] , 2014	47, F	Y	N	CR + WF
23	Xu <i>et al</i> ^[23] , 2015	54, M	N	Y	OR + AAF
24	Hu <i>et al</i> ^[24] , 2015	50, M	N	Y	OR + AAF
25	He <i>et al</i> ^[25] , 2016	72, M	Y	Y	OR + C1-3 F
26	Minyu <i>et al</i> ^[26] , 2018	30, M	Y	N	CR + AAF
27	Ghailane <i>et al</i> ^[27] , 2019	89, M	Y (C1 fracture)	N	CR
28	Ning <i>et al</i> ^[28] , 2019	52, M	N	N	OR + AAF
29	Nowell <i>et al</i> ^[29] , 2019	71, M	Y (Jefferson fracture)	N	CR + AAF
30	Our case	70, M	Y (OO)	Y (AH)	CR + C1-3 F

¹Transoral atlantoaxial reduction plate. M: Male; F: Female; Y: Yes (Type II Odontoid fracture); N: No; CR: Closed reduction; OR: Open reduction; WF: Wiring fusion; OCF: Occipitocervical fusion; AAF: Atlantoaxial fusion; SF: Screw fixation; OO: Os odontoides; AH: Atlas hypoplasia; F: Fusion.

To date, there is no consensus as to whether surgical fixation and fusion should be performed. An atypical case of PAD with C1 anterior arch fracture was treated by closed reduction under anesthesia without fusion because it was believed that there was no ligamentous instability due to the mechanism of PAD^[27]. Hu *et al*^[24] concluded that the need for fusion after successful closed reduction depends on the integrity of the transverse ligament and the stability of the cervical spine. However, evaluation of the condition of ligaments and stability of the cervical spine remains to be solved. Of these previously published cases, 21/29 underwent fixation or fusion, which indicated that most of the doctors were inclined to strengthen the stability by fusion.

Innovation points

What distinguishes our case from previous cases is the existence of OO and AH, as

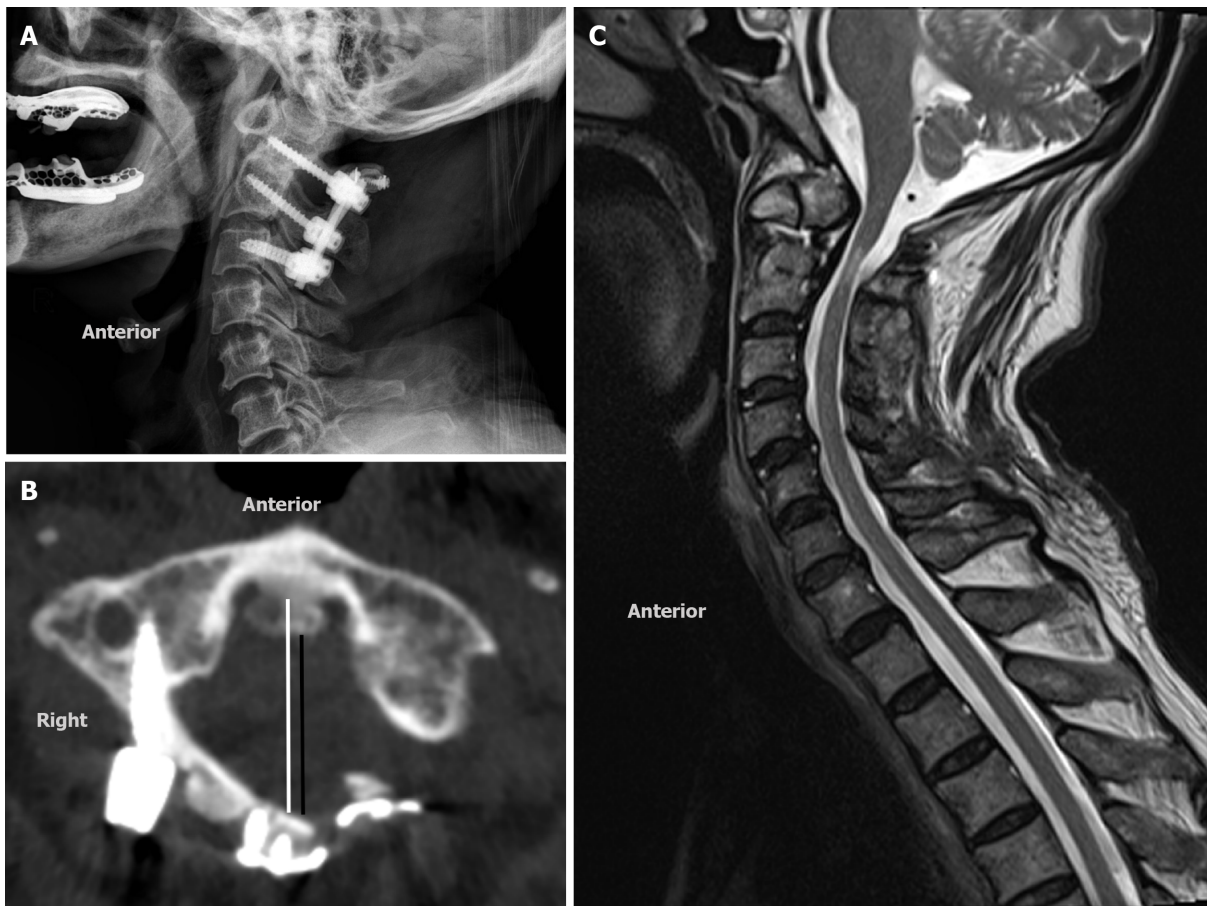


Figure 3 Postoperative images during the 3-yr follow up. A: Lateral radiograph indicating stable fixation and fusion; B: Axial computed tomography scan, demonstrating the C1 inner sagittal diameter (white line) = 32.75 mm and the canal sagittal diameter (black line) = 22.24 mm; C: Parasagittal magnetic resonance imaging, demonstrating satisfactory release of compression caused by the odontoid process and C1 posterior arch.

well as the adoption of decompression and fusion as the surgical strategy. Hypoplasia occurs when the sagittal diameter of the atlas is less than or equal to 26 mm^[30]. Fareed *et al*^[32] indicated that PAD was more likely to occur in OO patients due to deficient ossification of the odontoid and hyperlaxity of the ligament ring.

During the treatment of our patient, two important points should be noted. One was the preoperative flexed-positional skull traction. Soft and scar tissue adhesion caused by an old PAD could form a strong counterforce against rapid reduction, and long-term high weight traction could stretch the bound tissue gently and gradually. Additionally, the mildness and progressivity of sustained traction might effectively reduce the risk of transient over-distraction and stimulation of the spinal cord. Preoperative closed reduction is safer than intraoperative closed reduction under anesthesia, especially for old dislocations.

The other point is the unique surgical procedure. The conventional surgical procedure might be C1 laminectomy with occipitocervical fusion^[33], which has the disadvantages of larger surgical injury and less range of head motion compared with C1 laminoplasty with cervical fusion that has not been reported so far. Boniello *et al*^[34] suggested that laminoplasty results in decreased length of stay, readmissions and complications compared with laminectomy. Noguchi *et al*^[35] indicated that laminoplasty was a safe and useful procedure for AH. To the best of our knowledge, C1 laminoplasty can retain one side of the pedicle, which makes atlantoaxial fusion possible and results in an adequate bone grafting space to increase the success rate of fusion. Yang *et al*^[36] demonstrated that bone grafting of the atlantoaxial joint plays an important role in increasing the fusion rate of atlantoaxial fusion. Conversely, C1 laminectomy would inevitably lead to occipitocervical fusion, which would lead to the loss of more cervical range of motion with increased operative injury.

CONCLUSION

C1 single door laminoplasty with fusion for PAD combined with spinal cord compression could be a suitable and effective surgical option. Compared with laminectomy and occipitocervical fusion, it retains partial range of motion, decreases operative injury and provides an adequate bone grafting space for atlantoaxial fusion. An effective flexed-positional skull traction is vital in patients diagnosed with PAD. These findings require verification by further prospective, randomized studies.

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