



# A modified microsurgical interfacet release and direct distraction technique for management of congenital atlantoaxial dislocation: technical note

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## Abstract

Various techniques have been used for management of congenital atlantoaxial dislocation. Recently, the reduction of atlantoaxial dislocation through a single posterior approach has attracted more and more attention. Here, we present a modified technique including direct interfacet release and distraction between C1 and C2 by a specially designed distractor, posterior internal fixation and bone graft fusion. The illustrated technique was performed in 15 consecutive patients, and the outcomes were recorded and analyzed. Follow-up ranged from 12 to 26 months. Clinical symptoms improved in 14 patients (93.3%) and were stable in 1 patient (6.7%). Radiologically, 60–100% reduction was achieved in 13 patients (86.6%). Bone fusion was obtained in all patients at 12 months after the operation. The two-tailed Wilcoxon signed-rank test was used to analyze the preoperative and postoperative Japanese Orthopedic Association scores (JOA), atlas-dens interval (ADI), and cervicomedullary angle (CMA) ( $P < 0.001$ ). Our results suggested that this direct interfacet release and distraction technique with a specially designed C1–2 distractor can provide a definite effective C1–2 facet distraction and odontoid process restoration through a single posterior approach.

**Keywords** Atlantoaxial dislocation · Odontoid process · Interspace distraction

## Introduction

Atlantoaxial dislocation and odontoid process protrusion are the prominent pathological mechanisms of congenital basilar invagination. The surgical management of atlantoaxial dislocation is

focused on the reduction of the odontoid process. Current techniques could be divided as follows: (1) A single anterior approach was representative of the transoral atlantoaxial reduction plate (TARP) fixation technique introduced by Yin [13] in 2005; Yadav [12] described an endoscopic technique for single-stage anterior decompression and anterior fusion by transcervical approach in 2017. (2) An anterior combined posterior approach was representative of the transoral anterior decompression with posterior fusion for treatment of atlantoaxial dislocations introduced by Lee [8] in 1985; Wang et al. [11] detailed a modified technique which was one-stage anterior transoral release and posterior fixation in 2006. (3) A single posterior approach was representative of the transarticular method of fixation introduced by Magerl et al. [9] in 1987; Jain VK [5] also present a technique of occipital-axis posterior wiring and fusion for atlantoaxial dislocation associated with an occipitalized atlas in 1993 year; the insertion of screws into the lateral masses of the atlas and axis introduced by Goel and Lheri [4] in 1994; and the direct intraoperative distraction between the occiput and C2 pedicle screws, which was introduced by Jian et al. [7] in 2010. All these techniques have been clinically proven to be effective and have their various indications, advantages, and disadvantages.

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In this study, we introduce a direct posterior interfacet release and distraction technique with a specially designed C1–2 distractor (Fig. 1) for the management of congenital atlantoaxial dislocation (CAAD). The brief operating steps, critical technique points, the odontoid process restoration assessment method, and surgical outcomes are described and evaluated in detail.

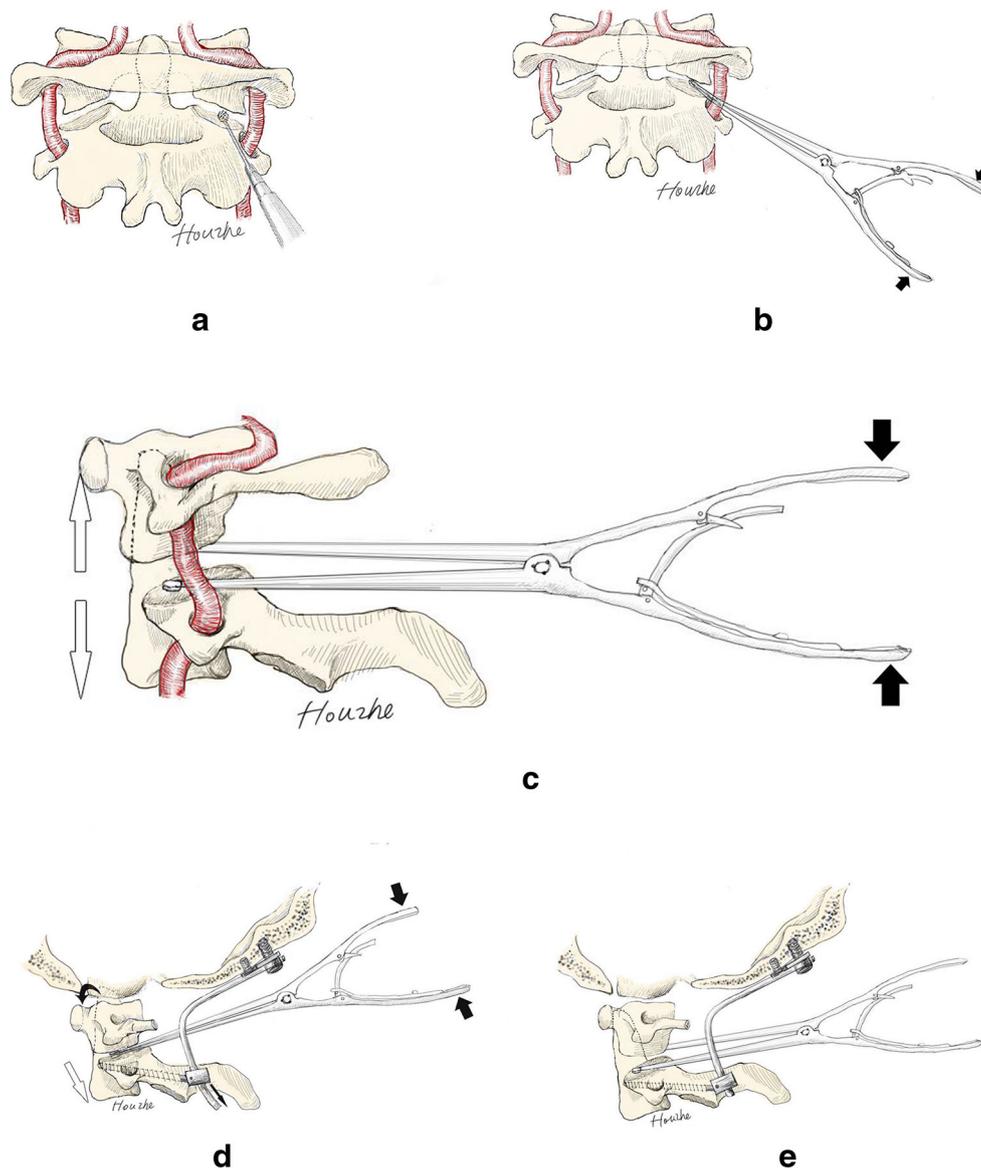
### Surgical procedure

The patients were placed in the prone position under general anesthesia, with the neck in neutral position and the head fixed in a pin head-holder device. A routine posterior suboccipital approach was used, exposing the region

from the posterior of the occipital foramen to the C2 spinous process. Malformed posterior arch of C1 was removed based on its compression on the dural sac. And, autogenous bone was obtained from the removed C1 arch. Subsequently, exploring and opening the C1–2 lateral joints, we performed a posterior release of the facet on both sides (Fig. 2a). The C2 ganglion was stretched upward to increase the visual field of a right-handed surgeon. After fully denuding the articular cartilage and bony spur, a specially designed C1–2 distractor was put into the interface of the facets (Fig. 2b), and then a pressurized force was placed on the hand rein of the distractor. The facets were distracted by the plate which was in direct contact with the facets (Fig. 2c). To gain a sufficient distraction space between the facets, this interfacet

**Fig. 1** A specially designed interfacet distractor between C1 and C2 is our independent research and design (China patent no. 201621427099.4)





**Fig. 2** Exploration and release of the C1–2 facets (**a**). We explored the articular surface of C1 and C2 and fully denuded the articular cartilage and bony spur and then inserted the specially designed instrument into the released facets of C1–2 (**b**). The specially designed C1–2 distractor was placed into the interface of the facet. Distracting the C1–2 facets moved the odontoid process downward (**c**). The facets were distracted by placing a force on the handle of the specially designed distractor. This step could be repeated multiple times until the odontoid process had moved downward a satisfactory distance on vertical level. The gliding of the

C2 screw on the angled rod permitted the odontoid process to move forward and downward (**d**). A screw was placed on either side of C2 but not tightened. After molding the connecting rods to a specific angle, we implanted four screws into the occipital bone to fix the rods. Then, the C2 screw would be able to glide along with the predetermined trajectory and angle by distracting the released articular surface. Thus, the odontoid process could move forward and downward a satisfactory distance on the horizontal and vertical levels. Finally, we tightened the screws on the C2, and the odontoid process was restored to a normal anatomical position (**e**)

distraction process was performed several times and gradually until the odontoid process had moved downward to a satisfactory distance on the vertical level.

Based on the occipitalization of the atlas occurred in all patients, we decided to employ occipital-cervical (C2) fixation. A multiaxial screw was placed on either side of C2 pedicle, and four occipital screws were fixed into the occipital bone. Two titanium rods were bent to have a certain angle and then connected between C2 pedicle screws and occipital

screws bilaterally. The connected rods were fixed by tightening occipital screws heads and only tightening C2 screw heads loosely. With the C0/1–C2 interfacet distraction at one side, the sloping of the C2 pedicle screw and C2 body on the previously angled rod would allow horizontal and vertical movement of the odontoid process to be restored to a satisfactory level (Fig. 2d, e). The interfacet distraction was performed again and matched a same level as the previous step at the other side. Then, the reduction of the odontoid process was

achieved. The C2 screw heads were tightened. The reduction effect of the odontoid process was verified by intraoperative radiography. Finally, all of the screws were fastened and mixed autologous, and artificial particulate bone was used to fill in the space between C1 and C2 facets to obtain postoperative bone fusion.

Preoperative CT angiography of vertebral artery (VA) was carried out routinely on all our patients. Sometimes, the 3-D model was also made to provide important clues for the course of the VA and its adjacent structures. Intraoperative neurophysiological monitoring (IONM), such as somatosensory evoked potential (SEP), was used to monitor the functional changes of the spinal cord. Any sign for decrease of amplitude and latency present by IONM may provide important hints to adjust the degree of decompression and distraction.

## Illustrate cases

**Case 1** A 39-year-old woman was referred to us with major clinical symptoms of limbs numbness and hypesthesia, dizziness, and progressive muscle weakness. In addition, she had developed unsteady gait in the last month. The result of the

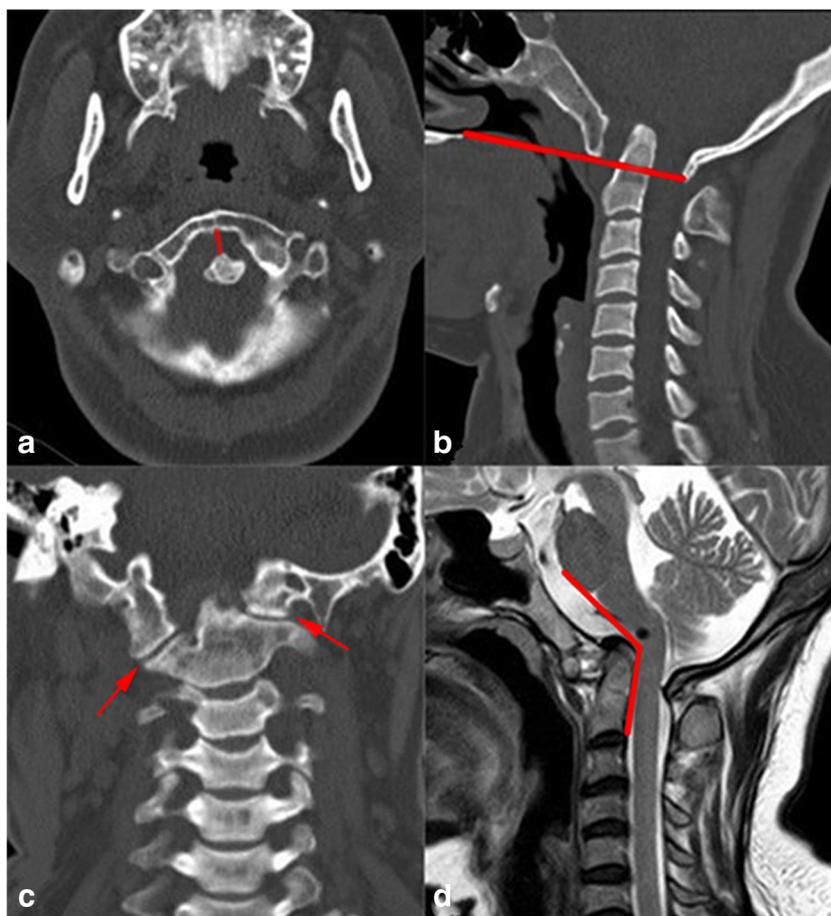
physical examination showed that limb myodynamia was grade 4, and heel–knee–tibia test and Rombergism were positive. The Japanese Orthopedic Association (JOA) score was 9. The preoperative MRI and CT scans were obtained and measured (Fig. 3)

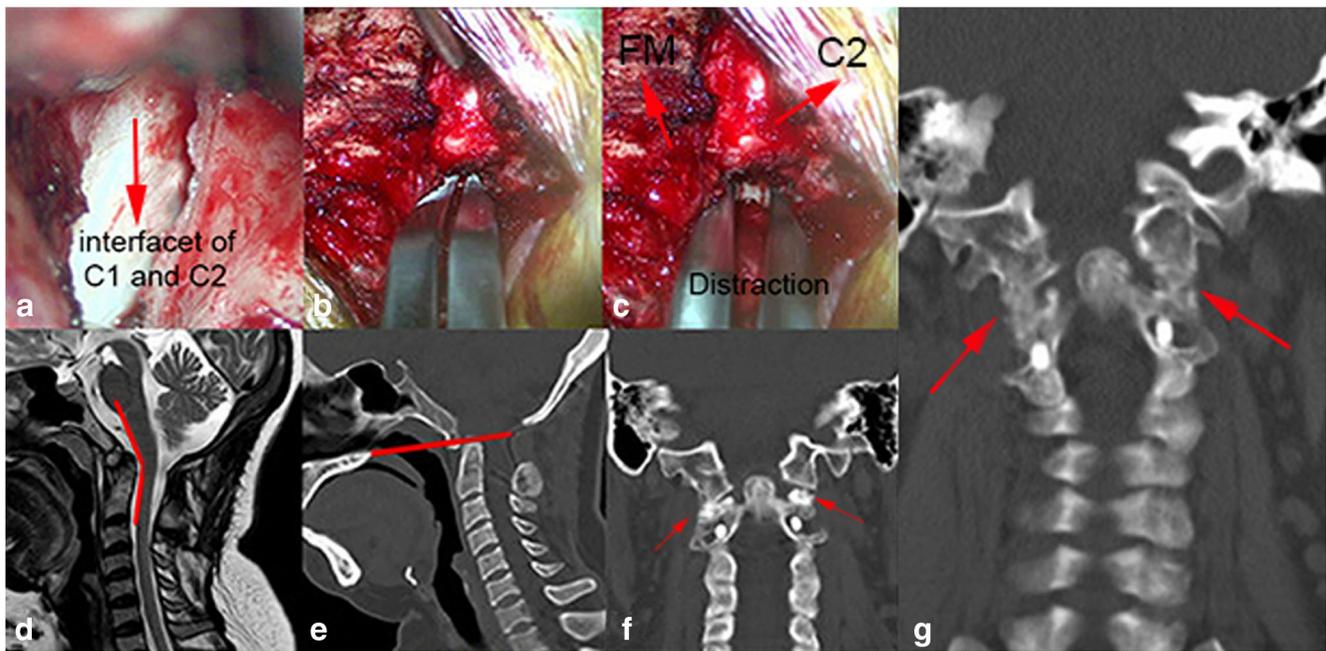
Eighteen months after the operation, adequate relief of symptoms including dizziness, numbness, and upper limb muscle weakness was achieved. Additionally, the myodynamia of both upper extremities was restored to normal, and the JOA score was improved to 12. MR and CT scans were available for review for this patient, at 2 weeks postoperative and at the 12 months follow-up after the operation (Fig. 4).

**Case 2** The patient was a 45-year-old woman with discontinuous headache and right limb numbness for the past 2 years, which became aggravated in the previous 7 months. The physical examination revealed that the limb myodynamia was grade 4, and the reflection of Babinski's sign was induced. Additionally, the JOA score was 12. The preoperative MRI and CT scans were obtained and recorded (Fig. 5).

On a follow-up survey, the patient indicated that the symptoms of headache and numbness had improved dramatically.

**Fig. 3** Preoperative radiological images from case 1. The axial CT scan (a) showed the atlas-dens interval extended, ADI = 6 mm (the red line). The sagittal CT (b) revealed that the odontoid process exceeded 15 mm from the CL line (the red line). In addition, the dysplastic C1 showed bone fusion with the foramen magnum, indicative of C1 assimilation. The red arrow is positioned at the preoperative articular surface of C1 and C2 (c). The sagittal MRI (d) showed that the odontoid process was compressing the brainstem, and the CMA = 127°





**Fig. 4** Details of the operation and postoperative radiological images from case 1. We made a release between the facets of C1 and C2 (a). Afterwards, the specially designed C1-C2 distractor was integrated into the interface of the facets (b). The facets were distracted by pressurizing the distractor (c). The sagittal MRI obtained 2 weeks after the operation (d) showed that the compression of the anterior brainstem had been

relieved, and the CMA = 150°. The CT scan (e) revealed that the odontoid process had been restored to a normal anatomical position, with an ADI = 0 mm. The facets of C1 and C2 were released, and mixed autologous and artificial particulate bone was implanted (f). Bone bridge formation between C1 and C2 had been obtained by 12 months after the operation (g)

In addition, the myodynamia of both lower extremities was restored to normal, and the JOA score increased to 14, by 18 months after the operation. The patient's MRI and CT were obtained 2 weeks postoperative and 6 months after the procedure (Fig. 5).

## Results

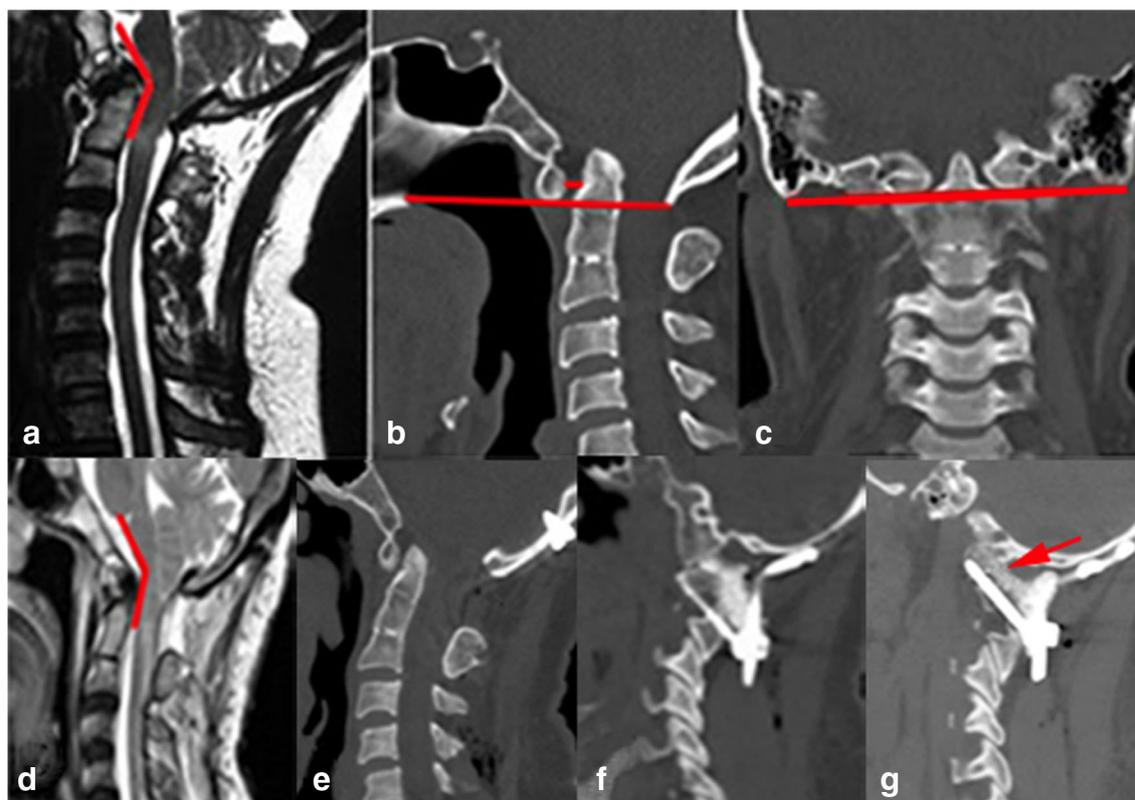
Between April 2016 and June 2017, 15 patients who had CAAD underwent a modified microsurgical interfacet release and direct distraction between C1 and C2, associated with posterior internal fixation and bone graft fusion, at Sanbo Brain Hospital Capital Medical University. The study was approved by the Institutional Review Board of Sanbo Brain Hospital. The cases are composed of 4 men and 11 women with an average age of  $41.4 \pm 11.5$  years (range from 24 to 62 years). The duration of symptoms ranged from 3 months to 20 years. The main clinical features are presented in Table 1.

All patients were diagnosed as CAAD with basilar invagination (BI) and partial or complete occipitalization of the atlas by the preoperative CT and MRI scans. C2–3 fusion was demonstrated in three cases. Os odontoides was not present in any case. The cervicomedullary angle (CMA) was measured to assess the extent of ventral compression of the spinal cord and medulla by the odontoid process. The measurement of atlas-dens interval (ADI) was used to evaluate the degree of

dens dislocation. Neurological function was assessed in all patients with the JOA scale. A two-tailed Wilcoxon signed-rank test (IBM SPSS Statistics 19.0) was performed for preoperative and postoperative JOA, ADI, and CMA.

The follow-up duration ranged from 12 months to 26 months ( $18.5 \pm 4.8$  months) for 15 patients (Table 2). No death, infection, or delayed neurological worsening case was reported in this series. Clinical symptoms improved in 14 patients (93.3%) and were stable in 1 patient (6.7%). There was no injury of vertebral artery and C2 nerve root during operation. Neither side of the C2 root resection was performed. Only one patient underwent postoperative right occipital mild numbness and which may be on account of the C2 nerve root harassment during the operation. The duration of surgery ranged from 60 to 90 min (mean 80 min), and the blood loss was 50 to 200 ml (mean 126 ml). The CT and MRI scans were obtained in all patients at 2 weeks and 12 months after the surgery. Sixty to 100% reduction of odontoid process was achieved in 13 patients, and the effective fixation was observed in all patients at 2 weeks after the operation. Varying degrees of bone fusion were achieved in all 15 patients at 12 months after the operation.

The postoperative average value of ADI decreased from  $5.1 \pm 1.5$  to  $0.9 \pm 1.2$  mm ( $p < 0.001$ ). Thirteen patients' value of ADI returned to normal range, revealing satisfactory reduction of the horizontal dislocation of the odontoid process. Compared with the preoperative measurement results, the



**Fig. 5** The preoperative radiological images from case 2. The MRI showed (a) that the ventral side of the brainstem was pressed by the odontoid process, the CMA = 131°. The sagittal CT revealed (b) that odontoid process exceeded 14 mm from the CL line (the red line), ADI = 4 mm. The dysplastic atlas showed bone fusion with the foramen magnum. The coronary CT (c) showed that the odontoid process exceeded the FL (Fishgold line, the red line) by more than 5 mm. Compared with preoperative radiological images, postoperative images

showed that the pressure on the front of the brainstem had been removed, and the CMA had increased to 145° at 2 weeks after the operation (d). In addition, the position of odontoid process had been satisfactorily restored, with an ADI = 0 mm (e). Finally, the mixed autologous and artificial particulate bone were implanted into the distracted facets of C1 and C2 (f). Six months after the operation, bone fusion had formed between both articular surfaces of C1–C2 (g)

mean value of CMA increased from  $126.6 \pm 6.9^\circ$  to  $143.5 \pm 5.7^\circ$  ( $p < 0.001$ ), indicating an indirect relief of ventral compression of the spinal cord and medulla. There were no changes for the ADI and CMA value of each patient from 2 weeks after the operation to the end of follow-up. Neurological

functions were further improved in three patients on sensory or motor function from 2 weeks to 6 months after the operation. The neurological functions were stable in all patients from 6 months to the end of the follow-up. The patients' JOA scores increased from the preoperative value of  $10.6 \pm 1.4$  (range from 8 to 13) to  $12.9 \pm 1.6$  (range from 10 to 15) at the end of follow-up,  $p < 0.001$ , showing a statistical significance between the above groups (Table 3).

**Table 1** Clinical features in 15 patients

Variable	All patients (n = 15)
<b>Demographics</b>	
Mean age, years (range)	41.4 ± 11.5(24–62)
Sex (M/F)	4:11
<b>Clinical symptoms, n (%)</b>	
Neck pain	5 (33.3)
Sensory disturbance	9 (60.0)
Motor dysfunction	7 (46.6)
Ataxia	2 (13.3)
<b>Radiologic features, n (%)</b>	
Basilar invagination	15 (100.0)
C1 assimilation	15 (100.0)
C2–3 fusion	3 (20.0)
Os odontoideum	0 (0.0)

## Discussion

Beginning in early 2016, we performed a microsurgical interfacet release technique by using a specially designed interfacet distractor. This method is derived from the commonly accepted posterior distraction and fixation technique, but is quite different from the previous method of intraoperative external cranial skull traction and intraoperative distract between the occipital bone and the C2 spinous process or C2 pedicle screw. The operating principle of this C1–C2 distractor may be similar to the arthroplasty disc space

**Table 2** Summary of clinical data obtained in 15 patients

Patient no.	Sex/age (year)	Complication	ADI (mm)			CMA		JOA		Follow-up (month)
			Pre	Post	Reduction percentage	Pre	Post	Pre	Post	
1	F/53	None	5	1	80	135°	142°	10	13	26
2	F/24	None	5	0	100	130°	150°	12	14	25
3	F/62	None	4	0	100	117°	138°	12	15	25
4	M/34	None	3	0	100	127°	141°	9	12	24
5	F/36	None	4	1	75	129°	148°	10	12	22
6	F/60	None	4	1	75	130°	151°	12	14	20
7 <sup>b</sup>	F/39	None	6	0	100	127°	150°	9	12	18
8 <sup>b</sup>	F/45	None	4	0	100	131°	145°	12	14	18
9	F/25	Occipital numbness	6	3	50	133°	146°	9	10	16
10	M/36	None	7	2	71	118°	136°	8	11	16
11	M/49	None	6	1	83	124°	141°	13	15	15
12	M/44	None	5	0	100	133°	147°	11	14	14
13	F/46	None	5	1	80	124°	145°	12	15	14
14 <sup>c</sup>	F/28	None	9	4	55	110°	130°	11	11	13
15	F/40	None	4	0	100	132°	143°	10	12	12

JOA Japanese Orthopedic Association scores, ADI atlas-dens interval, CMA cervicomedullary angle

<sup>a</sup>Reduction percentage = (pre-ADI value – post-ADI value) / pre-ADI value

<sup>b</sup>Illustrate cases

<sup>c</sup>Clinical symptoms were stable in 1 patient

distractor, but the size, moment, and angle of the tip of the distractor is specially designed.

In 1994, Goel and Laheri [4] introduced the atlantoaxial fixation technique, which involved individual insertion of screws into the facet of the atlas and the axis. This technique involved the opening of the joint, denuding of articular cartilage, and the introduction of a bone graft within the joint before fixation with a plate/rod and mono-polyaxial screws. A satisfactory odontoid process downward placement could be achieved with this technique through a single posterior approach. From then on, the distraction and restoration of the odontoid process through a single posterior approach have been pursued and proven to be effective by many authors [1, 6].

In 2004, Atul Goel [2] introduced a bilaterally atlantoaxial joint distraction by using an intervertebral

spreader common in anterior cervical disc surgery. The downward placement and restoration of the odontoid process were evaluated by intraoperative radiography. This technique necessitates cervical traction before induction of anesthesia and still requires a wide bilateral exposure of the facets and the section including the large C2 ganglion. The joint capsule was excised, and the articular cartilage was widely removed using a micro drill. Large pieces of cortico-cancellous bone graft, as well as a hydroxyapatite block or mental spacer, were used as a strut graft and packed into the joints. The potential for bleeding from the venous plexus and the safety of the vertebral artery should also be considered during these complicated procedures. Although this technique has proved practical and effective, it was obvious that this procedure was a

**Table 3** Statistical outcomes in 15 patients

Variable	Pre-mean value (range) <sup>a</sup>	Post-mean value (range)			Two-tailed Wilcoxon signed-rank test
		2 weeks <sup>b</sup>	6 month <sup>c</sup>	≥ 12 month <sup>d</sup>	
ADI (mm)	5.1 ± 1.5 (3–9)	0.9 ± 1.2 (0–4)	0.9 ± 1.2 (0–4)	0.9 ± 1.2 (0–4)	<sup>a</sup> VS <sup>b,c,d</sup> <i>P</i> < 0.001
CMA°	126.6 ± 6.9 (110–135)	143.5 ± 5.7 (130–151)	143.5 ± 5.7 (130–151)	143.5 ± 5.7 (130–151)	<sup>b</sup> VS <sup>c,d</sup> <i>P</i> > 0.05
JOA	10.6 ± 1.4 (8–13)	12.7 ± 1.5 (10–15)	12.9 ± 1.6 (10–15)	12.9 ± 1.6 (10–15)	<sup>c</sup> VS <sup>d</sup> <i>P</i> > 0.05

JOA Japanese Orthopedic Association scores, ADI atlas-dens interval, CMA cervicomedullary angle

<sup>a,b,c,d</sup> The mean value of ADI, CMA and JOA at different times

very demanding and anatomically precise technique and should only be performed by experienced surgeons. The modified technique described in this article requires a meticulous microsurgical release of the C1–2 facets. Opening up a narrow space only wide enough to place the 5-mm plates of the distractor into the facets is sufficient. This minimizes the risk of injuring the vertebral artery and avoids resection of the C2 ganglion. In the technique introduced by Atul Goel, the atlantoaxial joint distraction is dependent on the fixed-size spacer. By contrast, our technique makes an adjustable distraction directly between the C1–2 facets.

Another method using a posterior approach was recommended by O'Brien [10] in 2008, for patients with an anomalous course of the vertebral artery and abnormal pedicles precluding safe placement of the C2 pedicle screws. By using the C1 lateral mass and C2 translaminar screws, the distraction was conducted between the C2 translaminar screw head and the rod holder. Tightening the C2 locking nut onto the rod was performed to reduce C1–C2 sublaxations and decompress the spinal cord. Ligament and hyperplastic cartilage joint connections between the C1 posterior arch and the odontoid process, the distraction between the C2 screw and the rod may cause a one directional shift of C1 and C2 during the distraction. This may affect the restoration of the odontoid process. In comparison, using our technique, direct distraction between C1 and C2 can avoid or minimize this one directional shift of C1 and C2, making the restoration more effective.

In 2010, Jian et al. [7] introduced an intraoperative distraction between the occiput and C2 pedicle screws to reduce atlantoaxial dislocation (AAD). This technique has proven to be an effective, simple, fast, and safe method for the treatment of BI with AAD. Some authors have suggested that the distraction of C1–2 through the technique occurred between the occiput and C2 pedicle screws. During distraction, the C1–2 interfacet angle may change, and afterwards the cervical alignment may be affected. The modified direct distraction between the C1–2 facets better distributes the force of the distraction on the surface of the facets and thus may avoid changing the C1–C2 facet angle.

Three types of facet alignment of BI were identified by AtulGoel [3]. Type 1 facet alignment occurs when the facet of the atlas has been dislocated anterior to the facet of the axis on lateral imaging and simulates lumbosacral listhesis. Type 2 facet instability occurs when the facet of the atlas has been dislocated posterior to the facet of the axial. Type 3 facet instability occurs when the facets are in alignment, but instability has been diagnosed on the basis of clinical and radiological evidence. The modified direct interfacet distract technique described above was only practiced on type 1 facet alignment of BI with C1–2 dislocation but might also be effective on type 2 and type 3, the possibility of which should be further explored.

## Conclusion

The aforementioned posterior microsurgical release and direct interfacet distraction of C1–2 can provide a definite and effective downward placement and restoration of the odontoid process in surgical management of CAAD with BI. This modified posterior release and direct distraction technique may shift the surgical management of CAAD towards a less invasive approach. These results from our primary practice are gratifying and give us encouragement to pursue a further detailed study.

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## Compliance with ethical standards

**Ethical approval** All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Declaration of Helsinki and its later amendments or comparable ethical standards. For this type of study, formal consent is not required. This article does not contain any studies with human participants performed by any of the authors.

**Conflict of interest** The authors declare that they have no conflicts of interest.

**Informed consent** Informed consent was obtained from all individual participants included in the study.

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