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Treatment of Reducible Atlantoaxial Dislocation and Basilar Invagination Using the Head Frame Reduction Technique and Atlantoaxial Arthrodesis

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Abstract

Study design: Retrospective case series.

Object: To evaluate the outcomes of a head frame reduction and atlantoaxial arthrodesis technique for the treatment of reducible basilar invagination (BI) and atlantoaxial dislocation (AAD).

Methods: Seventy-two reducible BI and AAD cases who were treated with the head frame reduction and atlantoaxial arthrodesis technique from June 2015 to December 2018 were retrospectively analyzed. Radiological measurements including the atlanto-dental interval (ADI), the height of odontoid process above Chamberlain line, Wackenheim line, clivus-canal angle (CCA) and JOA score were evaluated.

Results: There was no death in this series. The follow-up period ranged from 6 to 32 months (mean: 21.2 months). Radiological, complete or 90% reduction was attained and complete decompression was demonstrated in all patients. The CCA increased from 123.22 ± 8.36 preoperatively to 143.05 ± 8.79 postoperatively ($P < 0.01$). There was no patient found postoperative dysphagia. Neurological improvement was observed in all patients, with the JOA scores increasing from 12.53 ± 1.93 preoperatively to 16.13 ± 1.23 postoperatively ($P < 0.01$). Solid bony fusion was demonstrated in 69 patients at follow-up (95.8%).

Conclusion: Head frame reduction technique is a simple and effective treatment which could relief neurologic compression and adjust the CCA in patients with reducible AAD and BI with lower potential risks. Atlantoaxial fixation with short segmental fixation, strong purchase and low shearing force could maintain superior stabilization. The safety and long-term efficacy of such fixation and reduction technique were favorable, which illustrated that it could be a promising treatment algorithm for such kind of disease.

Keywords

basilar invagination, atlantoaxial dislocation, head frame reduction, atlantoaxial arthrodesis, outcome

Introduction

Congenital bony anomalies at the cranio-vertebral junction (CVJ) primarily consist of atlantoaxial dislocation (AAD) and basilar invagination (BI) and often cause compression of the upper spinal cord, which requires surgical intervention.[1,2] Surgery for BI and AAD is complex and challenging, including reduction of the antero-posterior dislocation, distraction of the vertical invagination and correction of the clivus-canal angle (CCA).[3-5] Among all surgical techniques in the treatment of CVJ instability, the Goel's technique, including facet joint release, insertion of intraarticular spacers and posterior atlantoaxial fixation is widely applied and undoubtedly effective.[6] However, the abnormal CVJ bone anatomy, frequent vertebral artery variations and complex venous plexus make this

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technique difficult and challenging.[3,7,8] Exposing and opening the facet joint, drilling the cartilage and distracting the facet joint is extremely technical demanding and risky, posing patients under threats.[3,9-12] Therefore, for the surgical treatment of BI and AAD, a simple and effective technique which could achieve multi-planar realignment of the CVJ while reducing the potential risks is demanded.

Intraoperative manual cervical distraction and extension using head frame followed by posterior occipitocervical fusion has been reported as a simple and effective treatment for pediatric BI associated with Chiari malformation, which proposed a new algorithm for the treatment of such congenital CVJ disease.[13-15] In our institution, the intraoperative head frame reduction technique and atlantoaxial fixation have been used for the treatment of reducible BI and AAD. Based on our experiences, excision of the C2 ganglion, opening and handling the facet joint is unnecessary. This largely reduces the risk of hemorrhage from the venous plexus and the risk of vertebral artery injury. Satisfactory results were achieved, indicating that such fixation and reduction technique could be a promising treatment method for such complex disease. Here, the rationale, detailed process and operative nuances of this technique were introduced and discussed.

Materials and Methods

Patient Population

After the approval of the institutional review board, patients with reducible AAD and BI who were treated with the intraoperative head frame reduction technique under general anesthesia and atlantoaxial fixation from June 2015 to December 2018 at our institution were retrospectively reviewed. Dynamic X-rays were obtained before surgery to judge the reducibility. Patients with severe rigid dislocation which showed no reduction on the dynamic films were excluded, and a modified Goel's technique was performed as previous described.[3,8] In total, 72 patients were treated using this head frame reduction technique. Patient's electronic medical record data was reviewed carefully which include demographics features, clinical presentations, surgery details and perioperative complications.

Surgical Procedure

After induction of general anesthesia, the patient was placed in prone with a Sugita head frame in a neutral position. A posterior midline incision was made to expose the posterior edge of the foramen magnum, the fused C1 lateral mass, C2 lamina, and C2 pars. Exposure of bilateral facets were unnecessary. The venous plexus should be protected carefully to avoid bleeding. The inferior surface of the fused posterior arch was drilled to facilitate C1 screw insertion. The ideal entry point of the C1 polyaxial screw was located at the midpoint of the posterior surface of the C1 facet. After exposing the C1 screw entry points, a burr was made with a 3.0-mm hand drill, and then a 3.5-mm screw (18-24 mm; Vertex, Medtronic Sofamor Danek, Minneapolis, Minnesota) was inserted. Then, C2 pedicle screws were implanted for patients with normal course vertebral artery of C2 or C2-3 transarticular screw fixation technique was used as an alternative for fixation of C2 for patients with the high-riding vertebral artery of C2.[16]

After the C1 and C2 screws were inserted, the intraoperative reduction was achieved with manually reposition of Sugita head frame. First, one surgeon took hold of the head clamp and another assistant released all joints of the head holder. Vertical reduction was then accomplished by manual traction, and horizontal reduction was achieved by extending the head at approximately 20°. Last, the assistant locks the joints and fluoroscopic imaging was performed. All manipulations were based on fluoroscopic demonstration of the basion and the tip of the dens to ensure that proper realignment had been achieved. The maneuver may require 2 to 4 iterations before the final and optimal alignment is confirmed by fluoroscopy (Figure 1). During head repositioning, care should be taken that the head should always maintain in an extending motion and any flexion which may increase spinal compression should be avoided. Neurophysiological monitoring should be used as a safeguard.

Two titanium rods (diameter: 3.5 mm) were bent to curve along the contour of the C1 and C2 vertebrae. Then bilateral rods were linked into the screws and were gradually fastened by tightening the nut. Intraoperative fluoroscopy was used again to check the position of screw and the degree of reduction. The exposed cortical occipital bone and C2 spinous bone were decorticated by using a osteotome. Cancellous bone harvested from the posterior iliac crest was placed over the decorticated suboccipital and atlantoaxial complex. One or 2 cross-linkages were used to strengthen the screw-rod instrument and the wound was closed in layers. Hard cervical collar was worn for 3 months.

Main Outcome Measures

The selected patients were reviewed retrospectively through evaluation of the clinical records before surgery and after follow-up. The radiographic films, thin-slice CT, and magnetic resonance imaging (MRI) were assessed. The anterior atlantodens interval (ADI) was measured to evaluate the horizontal dislocation of C1 over C2. The Chamberlain line and Wackenheim line were measured on sagittal reconstructed CT scans to evaluate the extent of the vertical dislocation. The CCA was measured on sagittal CT scans to evaluate the extent of ventral compression of the spinal cord and medulla by the dislocated dens (Figure 2). Patient neurological status was evaluated by using the Japanese Orthopedic Association (JOA) score before and after surgery.

Statistical Methods

Data was analyzed using SPSS 22.0 software (IBM; Chicago, IL). A paired Student T-test was performed for pre- and post-operative anterior ADI, Chamberlain line, Wackenheim line, CCA, and JOA score. A P value of less than 0.05 was customarily considered significant.

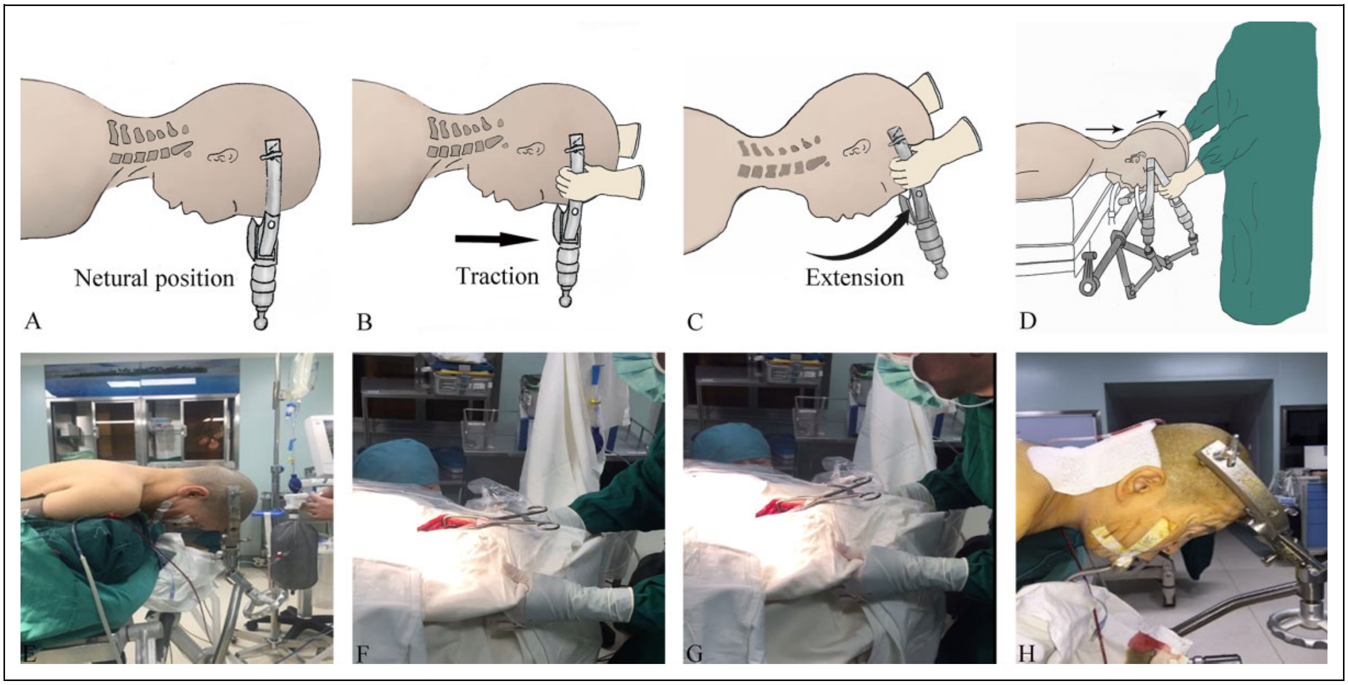

Figure 1. Head frame reduction procedure. (A, E) The head is placed in a Sugita head frame at netural position during exposure. (B, F)Vertical reduction is then accomplished by manual traction. (C, G) Horizontal reduction is achieved by extending the head frame. (D)The illustrations of reduction technique. (H)The position of head frame after surgery.

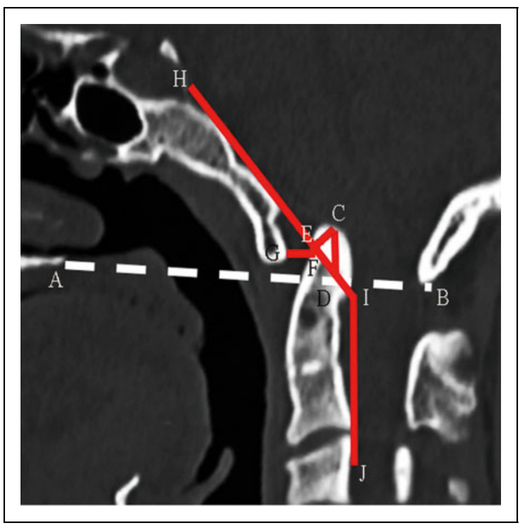

Figure 2. Pre- and postoperative radiologic parameters were measured on mid-sagittal reconstructed CT images. AB: Chamberlain line. C: the top of dens. CD: the height between the top of dens and Chamberlain line. CE: the height between the top of dens and Wackenheim line. FG: the distance of ADI. HI: Wackenheim line. HIJ: clivus-canal angle.

Result

Descriptive Data

A total of 72 patients including 28 male (38.9%) and 44 female (61.1%) patients were recruited in the study. The patient ages ranged from 21 to 65 years (mean age, 42.07 years). The clinical characteristics of the patients are summarized in Table 1. The duration of symptoms ranged from 3 months to 20 years, and the most common concern of the patients was progressive weakness of the extremities (91.7). Paresthesia occurred in 51 patients (70.8%), occiput/neck pain occurred in 57 patients (79.2%), and ataxia occurred in 37 patients (51.4%). Lower cranial nerve dysfunction occurred in 32 patients (44.4%). Occipitalization of the atlas was observed in 66 patients (91.7%), and congenital C2-C3 fusion (Klippel-Feil syndrome) was observed in 26 patients (36.1%). 38 cases (52.8%) had associated Chiari malformation and 29 cases (40.3%) had associated Chiari malformation and syringomyelia. The pre and postoperative radiologic and neurological assessment is shown in Table 2.

Outcome Data

The operative time ranged from 85 to 150 minutes (mean: 79 $\pm$ 55 minutes), and blood loss ranged from 50 to 200 mL (mean: 81 $\pm$ 71 mL). Among the 72 cases, bilateral C1 lateral mass screw and C2 pedicle screw or C2-3 transarticular screw was used. There were 4 patients had post-operative complications and no patient died. Pulmonary infection was found in 3 patients, who recovered by 1-week antibiotic treatment. Deep vein thromboses occurred in 1 patient, who recovered after anticoagulation therapy with heparin. There was no patient found postoperative dysphagia in this series.

Main Results

The follow-up period for all patients ranged from 6 to 32 months (average: 21.2 months). Post-operative reconstructive

Table 1. The Baseline and Clinical Characteristics of All Patients.

Baseline and clinical characteristics	No.
Number / n (male/female)	72 (28/44)
Age / y	42.07 ± 13.9
Duration of symptoms / months	18.79 ± 36.6
BMI / kg/m^2	23.83 ± 3.39
Presenting symptoms / n (%)	
Occiput/neck pain	57 (79.2)
Paresthesia	51 (70.8)
Extremities weakness	66 (91.7)
Ataxia	37 (51.4)
Lower cranial nerve dysfunction	32 (44.4)
Chiari malformation / n (%)	38 (52.8)
Chiari malformation and syringomyelia / n (%)	29 (40.3)
Bony anomalies / n (%)	
Basilar invagination	72 (100)
Occipitalization of the atlas	66 (91.7)
Klippel Feil sydrome	26 (36.1)

Table 2. Pre- and Postoperative Radiologic and Neurological Assessment.

Parameter	Pre	Post	P value (t-test)
ADI (mm)	6.45 ± 2.05	2.48 ± 0.97	<0.001 (14.2)
Chamberlain line(mm)	11.28 ± 2.21	4.15 ± 1.54	<0.001 (12.6)
Wackenheim line(mm)	5.48 ± 2.18	1.07 ± 0.36	<0.001 (17.7)
clivus-canal angle (degree)	123.22 ± 8.36	143.05 ± 8.79	<0.001 (13.3)
JOA score	12.53 ± 1.93	16.13 ± 1.23	<0.001 (13.5)

CT scans and MRI were performed at 1 week and 3 to 6 months after surgery. X-rays, CT scans, and MRI were repeated at 12 months after surgery and annually thereafter (Figure 3). Radiographic evaluation revealed a solid bony fusion in 69 patients (95.8%) at their 12-month follow-up. Three patients (4.2%) of the occiput had no solid bony fusion at their 12-month follow-up. Thus, they underwent a second operation to place cancellous bone grafts, and bony fusion was confirmed 6 months later. Table 2 shows the pre- and postoperative radiologic measurements. The mean postoperative anterior ADI was 2.48 ± 0.97 mm, which was significantly decreased from the preoperative value of 6.45 ± 2.05 mm ($P < 0.01$). Similarly, the Wackenheim line and Chamberlain line both showed significant improvements. The CCA increased from 123.22 ± 8.36 preoperatively to 143.05 ± 8.79 postoperatively ($P < 0.01$). Radiologically, complete or .90% reduction was attained and complete decompression was demonstrated in all patients. Neurological improvement was observed in all patients, with the JOA scores increasing from 12.53 ± 1.93 preoperatively to 16.13 ± 1.23 postoperatively ($P < .01$).

Discussion

The surgical technique for the treatment of congenital AAD and BI has significantly improved in recent decades.[3,4,6,8,11] The current treatment for such disease has shifted from combined anterior and posterior surgery to direct posterior reduction and fixation with manipulating the atlantoaxial joints. Goel et al[6] first reported a direct posterior technique by distraction of the C1-C2 facet joints using spacers under cervical traction followed by C1-C2 fixation, which could lead to both reduction of BI and AAD. Yin et al[3,8] modified this technique and adopted curved rods and C1-C2 screws to draw the C1 backward, push the C2 forward and downward. Chandra et al[2,4] used a distraction, compression and reduction technique with C1-C2 spacer placement and C1-C2/Oc-C2 translaminar screws, which achieved satisfied reduction. Although a significant improvement of neurological status and stabilization has been achieved following these approaches, surgery for CVJ anomalies is still challenging. Exposing and opening the facet joint, drilling the cartilage and distracting the facet joint is extremely technical demanding and risky, especially in patients with vascular and osseous variations.[7,9,12,17,18] Therefore, a technique which could achieve multi-planar realignment of the CVJ while reducing the potential risks is demanded.

In this study, we introduced an effective and simple algorithm for the treatment of such reducible congenital CVJ disease, which include head frame reduction and atlantoaxial fixation. Head frame reduction technique is effective in restoring the CCA compared with the traditional Goel's technique and is also simple in operation. In this technique, traction and extension by reposition of the head frame to achieve reduction of the BI, AAD and CCA were performed. Manual reduction was achieved under general anesthesia. It is because that the tension of neck muscle and ligament was decreased after anesthesia, which enables the intraoperative traction and extension via the head frame to achieve reduction. Besides, there is no need to expose the facet, cut the C2 ganglion, and handle the facet joint based on our experience, which largely reduces the risk of hemorrhage from the venous plexus and the risk of vertebral artery injury. However, it is also worth noting that the in this series severe rigid BI and AAD was excluded. Therefore, in some complex cases, such as patients with osteophyte between the dens and anterior tubercle of C1, satisfied reduction could not be achieved by such reduction technique. In these cases, facet joint release and transoral odontoidectomy followed by posterior instrumented fusion was still a better choice to be recommended.[19]

In treatment of the congenital BI and AAD, the significance of the CCA in patients has more and more attracted spine surgeon's attention due to the recognition of cervical sagittal realignment and the risks of serious post-complications.[20-22] In our clinical practice, however, we found that the Goel's facet joint distraction technique could not restore the upper cervical kyphosis. Patients with occipito-cervical fixation suffer from upper airway obstruction, dyspnea and dysphagia due to the decrease of occipito-cervical angle after surgery has been

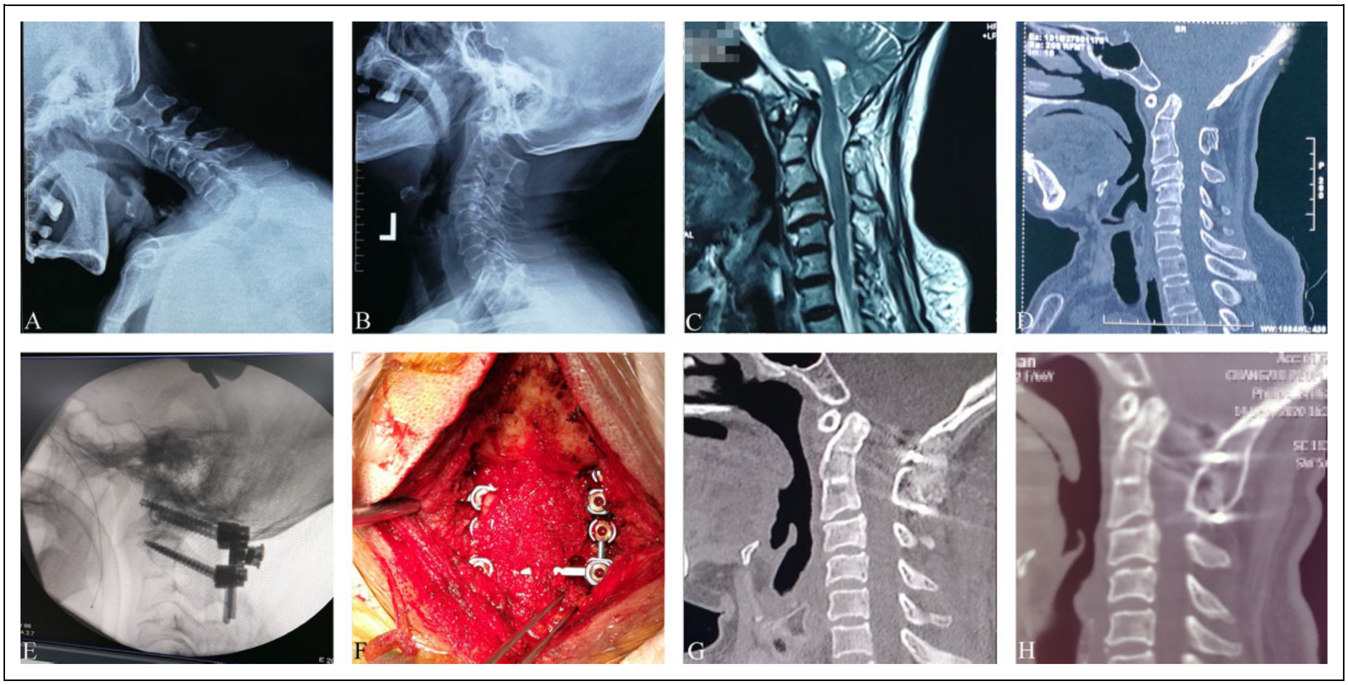

Figure 3. A 64-year-old female presented with weakness of extremities and unsteady gait. (A, B)The preoperative Flexion-extension radiograph showed total reducible atlantoaxial dislocation (AAD). (C, D)The sagittal magnetic resonance imaging (MRI) and reconstructed computed tomography (CT) scanning showed severe atlantoaxial dislocation (AAD), basilar invagination (BI), and occipitalization of the atlas (OA). (E) Fluoroscopic imaging is performed to ensure the proper realignment and the maximal CCA had been achieved. (F) The patient underwent reduction and fixation by the C1–C2 screw-rod technique. (G) The 3-dimensional reconstructed postoperative CT scanning demonstrated the CCA was significantly increased from 136.5 degree to 144.3 degree. (H) Solid bony fusion was also confirmed by the formation of continuous cortical bone between the C0/C1 and C2 lamina on CT scanning at the 24-month follow-up.

reported.[20,23] The possible reason is that the decreased CCA recruit compensatory mechanism to increase subaxial cervical lordosis (C2 through C7), which results in the narrowed space between the esophagus and cervical vertebra eventually leading to the dyspnea and dysphagia. Thus, a proper increase of CCA is necessary for the best outcome of patients during reduction. In our patients, the CCA was restored by extension of the patient's head after anesthesia, which is different from the previous studies. In the post-operative group, the average CCA was significantly increased. Although less than the normal value, the extent of reduction (mean 143.05 $\pm$ 8.79 degree) was enough to achieve clinically satisfied result. Meanwhile, no patients were found postoperative dysphagia in this series.

The mechanical strength provided by internal fixation is crucial for maintaining reduction and facilitating bony fusion.[18] In our study, after maximal reduction was achieved from intraoperative fluoroscopic demonstration, we used atlantoaxial fixation technique for stabilization of BI and AAD. This technique could direct stabilize the atlantoaxial facet with short segmental fixation, strong purchase and low shearing force, showing a superior biomechanical character compared with occipital-cervical fixation. Besides, the C2 vertebra could be pushed forward and downward with force exerted by the screw and rod lever system during rod fixation procedure and further achieve horizontal reduction and vertical reduction. Most cases achieved satisfied reduction with symptoms relieved using this technique.[24,25] Furthermore, to increase bony fusion, 1 or 2 cross-linkages were used to the axial stability, and finally 95.8% fusion rates were observed in our study.[26]

Based on our clinical experience, there are several operative nuances of using such reduction technique. During reduction when the position of the head frame was manually changed, the position of occipital protuberance and the spinous process of the axis should keep straight to guarantee the neutrality position. Throughout the hyperextension manipulation, potential neurological change due to spinal cord compression may happen. Therefore, hyperextension should be carefully done to avoid the severe compression caused by the assimilated infraoccipital margin on the spinal cord during reduction procedures and multimodal neuromonitoring is required during this surgical operation irrespective of severity of dislocation. To ensure satisfactory reduction, real time adjustment and modification is necessary based on the intraoperative fluoroscopic demonstration. Maximal reduction of the CCA and distraction of the basion rostral to the tip of the dens should be confirmed during surgery.

Several limitations should be considered in the present study. First, this was a retrospective survey, and the retrospective nature of this study might impact the overall accuracy. Moreover, no comparison group was included because of the relative paucity of AAD and BI patients. Despite these limitations, our technique achieved favorable outcomes in surgery and have important implications for clinical management as well as future exploration.

Conclusions

Head frame reduction technique is a simple and effective treatment which could relief neurologic compression and adjust the CCA in patients with reducible AAD and BI with lower potential risks. Atlantoaxial fixation with short segmental fixation, strong purchase and low shearing force could maintain superior stabilization. The safety and long-term efficacy of such fixation and reduction technique were favorable, which illustrated that it could be a promising treatment algorithm for such kind of disease.

Authors' Note

Teng Li and Yue-Qi Du contributed equally to this work.

Declaration of Conflicting Interests

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