

Technical Note

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INTRODUCTION

Treatment of basilar invagination (BI) without Chiari malformation or syringomyelia is offered for ventral compression and cervicomedullary strain, which occur as a consequence of fixed or mobile malaligned facets. Posterior distraction of the atlantoaxial joints as a treatment for BI seems logical in cases without an excessive angular deformity of the odontoid with respect to the atlas. For that reason, that technique has become popular, but it poses an inherent risk of injury to the vertebral arteries, venous plexus, and C2 root, and in some cases it is not possible to achieve reduction of the dislocation. The anterior retropharyngeal approach offers adequate access to both atlantoaxial joints in most cases. The unilateral submandibular route to both

Anterior Retropharyngeal Cage Distraction and Fixation for Basilar Invagination: “The Wedge Technique”

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Objective: Surgery is indicated for basilar invagination (BI) in symptomatic patients. In many patients, symptoms and signs occur due to an upward-migrated and malaligned odontoid with fixed or mobile atlantoaxial instability. Posterior distraction and fixation of the atlantoaxial joints has evolved to become the standard of care, but has some inherent morbidity. In this study, we propose that the unilateral anterior submandibular retropharyngeal approach with customized wedge-shaped titanium cages inserted into both atlantoaxial joints and anterior atlantoaxial fixation with a plate screw construct is a safer and easier option in many cases of BI.

Methods: From February 2014 to February 2019, 52 patients (age range, 15–78 years; 40 males and 12 females) with symptomatic BI with atlantoaxial dislocation and minimal sagittal facetal inclination and only mild Chiari malformation without syringomyelia were offered anterior submandibular retropharyngeal atlantoaxial distraction and fixation surgery.

Results: Neurological improvement occurred in 80% of patients, while the neurological status of 20% remained unchanged. No patients worsened, and no major complications or mortality was observed.

Conclusion: In properly selected cases of symptomatic BI, anterior wedge cage distraction with anterior atlantoaxial fixation is a safe and simple option.

Keywords: Basilar invagination, Atlantoaxial dislocation, Craniovertebral anomaly, Atlantoaxial fixation, Vertebral artery injury, Facetal realignment

joints runs between muscle planes and does not require major disruption of the craniovertebral stabilizing ligaments. There is almost no risk of injury to the vertebral artery or paravertebral venous plexus, and the C2 ganglion is not at any risk. The supine position with mild extension of the head almost always results in reduction of mobile atlantoaxial dislocations, and most irreducible dislocations can be reduced by odontoid and facet manipulation. Customized wedge-shape lordotic titanium cages can be used to distract the axis with the odontoid and to reduce BI. The lateral mass of the atlas and the body of the axis provide adequate bone stock for rigid screw plate fixation without a meaningful risk of vascular injury.

CASES AND OPERATIVE TECHNIQUE

1. Cases

From February 2014 to February 2019, 52 patients (age range, 15–78 years; 40 males and 12 females) with symptomatic BI with mild sagittal inclination (types I and II) of the facets and mild Chiari malformation without syringomyelia were offered anterior distraction fixation surgery. Neck pain, spasticity, grip weakness, loss of balance while walking, and difficulties in urinating were the commonest symptoms.

Institutional Review Board approval was obtained from the ethics committee of Poona Hospital (ECR/327/inst/Mh/2013/RR-16). All patients and their relatives received an explanation about the disease and what solution was being offered, the risks, complications, alternatives, benefits, and the natural history of

the disease if untreated. We always encourage second opinions. Informed consent was obtained from all patients and their immediate relatives in their local language, handwritten by them following Indian law and hospital requirements.

Lateral craniovertebral X-rays in flexion and extension (simulating the supine position on the operating table with a small bolster under the shoulders), magnetic resonance imaging with angiography to study the course of the vertebral arteries, and dynamic computed tomography and angiography with sagittal and coronal reconstructions were performed in all cases. The bone quality of the lateral masses of the atlas and the body of the axis vertebra was analyzed carefully to confirm the presence of adequate bone stock for fixation.

Patients in whom the angle of the mandible on lateral image was near or above the C2–3 disc were selected for anterior surgery (Fig. 1).



Fig. 1. Lateral cervical X-ray in extension showing the anatomical relations of the mandible angle (arrow).

2. Operative Technique

After general anesthesia with fiber-optic assisted intubation, Gardner-Well skull traction was applied in the supine position with mild extension and slight rotation of the head to the left side, and the head was fixed using adhesive tape to the operating table. A right-sided anterior submandibular retropharyngeal approach exposed both atlantoaxial joints, and then the longus coli, anterior longitudinal ligament, and rectus capitis anterior were cleared from the anterior surface of the atlantoaxial complex bilaterally to achieve reduction by extending the head. In irreducible dislocations, the area of soft tissue–abnormal bone fusion between the atlas and the odontoid was excised and the odontoid was impaled with a 3-mm tap after making a drill hole and pulled up to reduce the dislocation. The joints

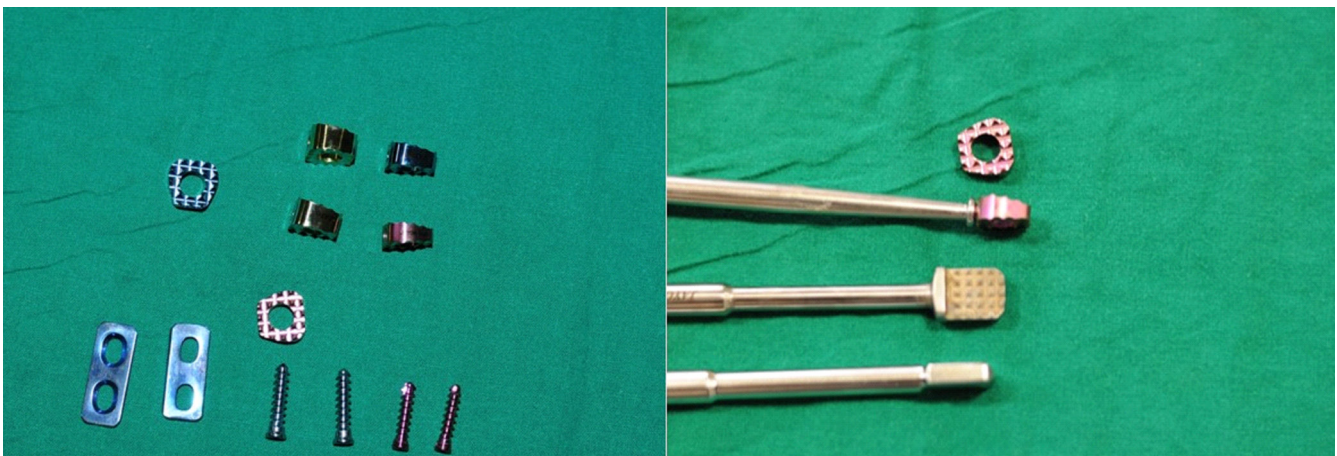


Fig. 2. Customized wedge-shaped titanium cages and plate with screws.

were curetted, and end plates were abraded with a micro-burr until oozing blood was seen. Specially customized titanium wedge-shaped hollow cages (5–7 mm anteriorly, 3–5 mm posteriorly, and 12-mm width [Jayaon Surgical Co., Chennai, India]), which were filled with bone substitute (G. Surgiwear India Ltd., Mumbai, India), were impacted into the joints (Fig. 2). Extension of the head and shape of the cages with intraoperative traction resulted in reduction of the BI and reduction of the atlantoaxial dislocation, as confirmed by intraoperative C-arm imaging. The rotation of the head was corrected to the neutral

position. On either side, a 3.5-mm-wide titanium reconstruction plate was used to fix the lateral mass of the atlas with the body of the axis, using titanium screws (4 mm in width and 18–20 mm in length) bilaterally. The atlas screw entry point was 5 mm above the midpoint of the atlantoaxial joint and was directed 10° upwards laterally. The axis screws were started at the base of the superior facet 5 mm below the midpoint of the joint, and directed to the midline of the body of the axis vertebra.

Postoperative imaging showed adequate reduction of BI, correction of atlantoaxial instability, and rigid fixation of the atlas

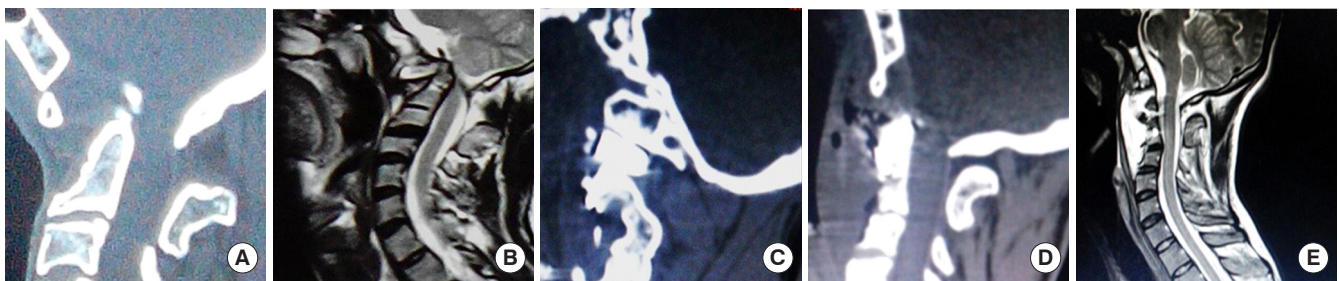


Fig. 3. (A) Preoperative sagittal computed tomography (CT): basilar invagination (BI) with atlantoaxial dislocation (AAD). (B) Preoperative magnetic resonance imaging (MRI): BI with AAD. (C) Postoperative CT: Wedge Titanium Cage distraction of atlantoaxial joint. (D) Postoperative CT: realignment of odontoid. (E) Postoperative MRI: realignment of odontoid.

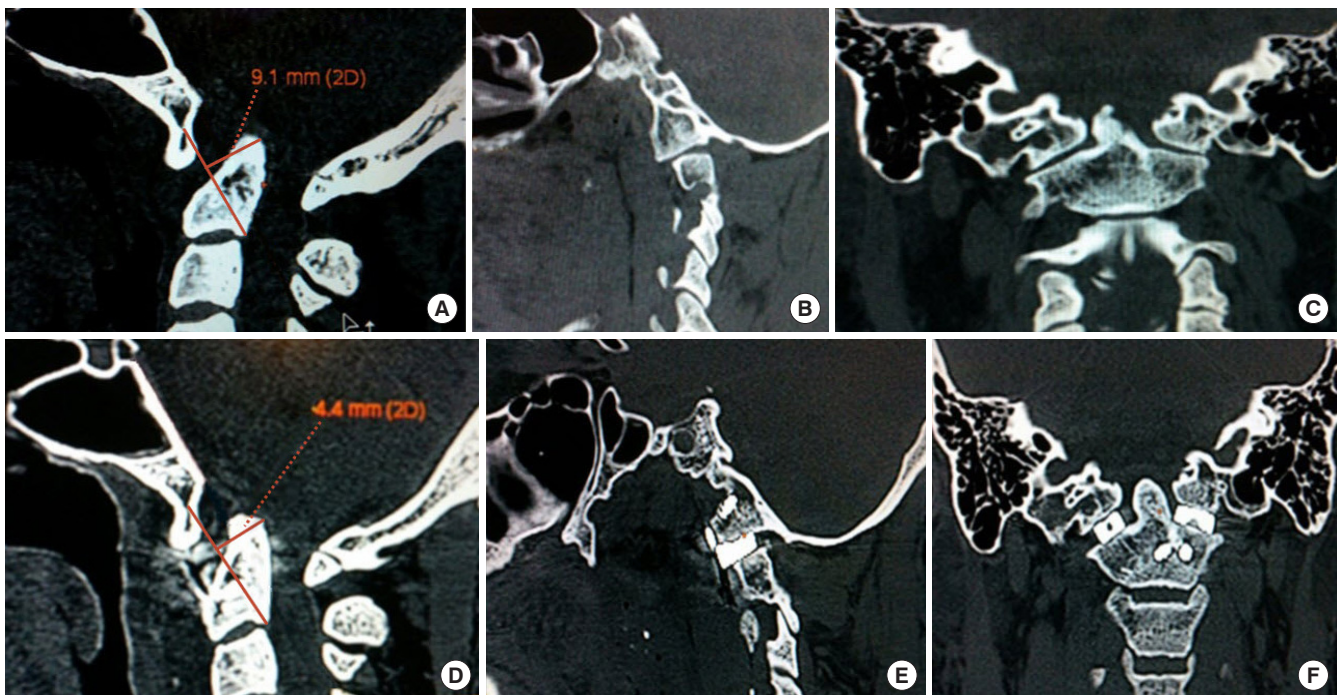


Fig. 4. (A) Preoperative sagittal computed tomography (CT) scan: basilar invagination (BI) with atlantoaxial dislocation and cervicomedullary strain as per Grabb-Oakes index. (B) Preoperative Sagittal CT scan: facet inclination and subluxation. (C) Preoperative coronal CT: atlantoaxial joint inclination. (D) Postoperative sagittal CT: reduction of BI and cervicomedullary strain with favorable Grabb-Oakes index. (E) Postoperative coronal CT with Wedge Titanium Cage for realignment and distraction of atlantoaxial facets. (F) Postoperative coronal CT showing realigned odontoid and distracted atlantoaxial facets.

toaxial joints (Fig. 3).

Oral nutrition was initiated 4 hours after surgery, and ambulation was started 6 hours after surgery with a collar in extension. Patients were discharged from the hospital after 2–5 days, the collar was discontinued after 3 weeks, and follow-up dynamic imaging at 3, 6, and 12 months was advised.

RESULTS

Most mobile dislocations were reduced completely. In the fixed dislocations, the Grabb-Oakes distance decreased by at least 3–6 mm in all cases and was less than 9 mm in all cases (Fig. 4). Neck pain was relieved in 45 of 45 patients, spasticity improved in 30 of 40, grip weakness in 35 of 45, and walking balance in 15 of 30; however, the 10 patients with preoperative urinary disturbances experienced no changes. Three months of postoperative follow-up data were available for 50 patients, 6

months for 40 patients, and 24 months for 26 patients. Neurological improvement occurred in 80% of patients, and the neurological status of 20% remained the same, most likely due the preoperative severity of the cord injury. No patients worsened after surgery. Bone fusion across the joints was seen in 23 patients at 12 months (Fig. 5). Blood transfusion was not required in any case, and the average operating time was 180 minutes after the incision.

In 2 patients, a Grabb-Oakes distance of 8 mm persisted even though the odontoid had migrated downwards; however, the patients experienced symptomatic relief. Mild postoperative neurapraxia of the hypoglossal nerve and marginal mandibular nerve occurred in 6 patients, but resolved completely in 4 weeks. Two patients complained of dysphagia for liquids in the postoperative period for 1 week. Unilateral posterior cage migration without any consequence occurred in 1 patient.

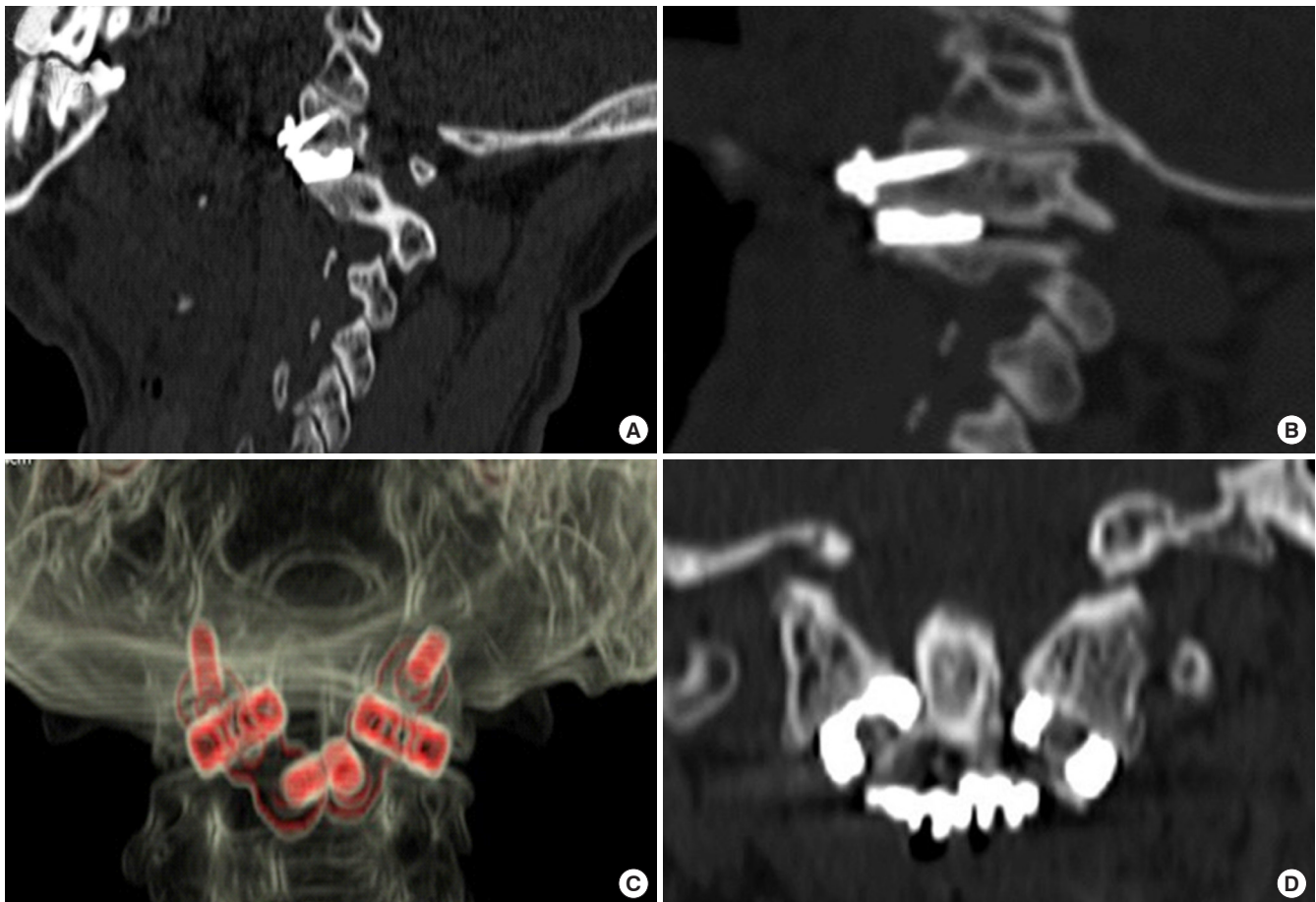


Fig. 5. (A) Postoperative sagittal computed tomography (CT) of facet joint in flexion at 12 months. (B) Postoperative sagittal CT of facet joint in extension at 12 months. (C) Postoperative coronal CT showing implant position at 12 months. (D) Postoperative coronal CT at 12 months showing bone fusion across atlantoaxial joints.

DISCUSSION

BI is a congenital disorder that has been defined as a prolapse of the vertebral column into the spinal cord.¹

Over the past century, various radiological lines have been used to diagnose BI, with commonly used criteria including the Chamberlain, McGregor, and Bull angles, as well as the Fischgold digastric line.² Symptoms and signs in patients with BI occur as the result of the following factors, alone or in combination: compression due to an upward-migrated odontoid process with or without malalignment, crowding at the foramen magnum due to Chiari malformation, and instability of the atlantoaxial joints. The kyphotic clivo-axial angle is increasingly recognized as an index of risk of brainstem deformity and cranio-cervical instability.^{3,4}

Surgical treatment of BI and associated instability has evolved from only a bone overlay to sublaminar wire fixation and, subsequently, screw plate/rod fixation. Facet fixation of the atlas and axis has become the standard of care. Intraoperative bleeding and vertebral artery injury remain the main issues in terms of complications, although techniques including neuronavigation and preoperative 3-dimensional model prints have been proposed to decrease the surgical morbidity.⁵⁻⁷

The classification of this problem by Goel et al.⁸ in 1998 into 2 groups with different therapeutic options was a landmark contribution. In 2004, Goel⁹ proposed distraction of the atlantoaxial joints with bone grafts or spacers with realignment and fixation of the atlantoaxial joints, which revolutionized the treatment of this problem and attracted many researchers to this option. In 2014, Goel¹⁰ proposed that atlantoaxial instability is the root issue of all types of BI, and that associated problems such as Chiari malformation, syringomyelia, neck shortening, torticollis, and Klippel-Feil syndrome are all protective mechanisms to protect the cord at the craniovertebral junction from the perils of atlantoaxial instability. This concept seems controversial embryologically, and the claim that atlantoaxial fixation can reverse these pathologies is difficult to fathom. BI with Chiari malformation and syringomyelia without demonstrable atlantoaxial instability is treated by foramen magnum decompression and duroplasty with good success, as supported by the literature.

Many other researchers have found it impossible to realign the facets in all cases of BI, and complex, elaborate, and difficult procedures such as facet reshaping before atlantoaxial fixation and distraction compression instrumentation with spacers between the occiput and axis (pseudofacets) to achieve this goal

have been described in the literature.^{11,12} Recently, a variable screw placement plate with a pedicle screw construct to enhance manipulation and fixation in posterior atlantoaxial fixation was described by the author.¹³

The anatomy of the facets in extreme inclination in the sagittal and coronal planes does not permit distraction along the vertical axis, and “joint jamming”¹⁴ from behind will result in dragging the misaligned odontoid downwards without changing the clivo-axial angle and the cervicomedullary strain.

The severity of BI and atlantoaxial dislocation correlates with sagittal joint inclination, coronal joint inclination, and cranio-cervical tilt. Yin et al.¹⁵ classified BI based upon facet inclination in the sagittal and coronal planes, and suggested that the insertion of a spacer between the facets in type III is impossible, and that other methods of realigning the craniovertebral junction are equally difficult and dangerous. In such circumstances, they recommend transoral odontoidectomy and posterior fixation, which seems reasonable.¹⁵

Anterior transoral surgery for BI with a special plate has been described recently and needs further evaluation.¹⁶

The anterior extrapharyngeal approach to the atlantoaxial joints has been amply described in the literature.¹⁷ In properly selected cases, the anterior unilateral extrapharyngeal approach offers direct exposure of both atlantoaxial joints with the opportunity to manipulate, reduce, distract, and fix the joints with a plate screw construct. This approach avoids the vertebral arteries, venous plexus, and C2 root ganglion. The anterior approach avoids disruption of the posterior tension band and maintains the ligamentous and muscle attachments of the axis spinous process, which are essential for spinal balance. The surgical procedure is performed with the patient in the supine position with the head in extension, facilitating reduction of atlantoaxial malalignment by excising the soft tissue or bony impediments between the atlas and the odontoid and permitting the odontoid to be impaled and held for realignment (Supplementary video clip 1). The wide exposure of both joint spaces without a space restriction due to proximity of the vertebral artery or spinal cord permits adequate “gardening” by abrading the endplate cartilage with a micro-burr and angled curettes until bleeding starts, which is essential for osteoinduction. A customized lordotic titanium cage with an adequate bone graft can be wedged into the joint to achieve downward migration of the odontoid in the correct axis to eliminate or reduce cervicomedullary strain. A wedge-shaped lordotic cage acts as a simple machine that distracts the joint and pushes the atlas upwards and backwards, thereby reducing the BI, correcting the atlanto-

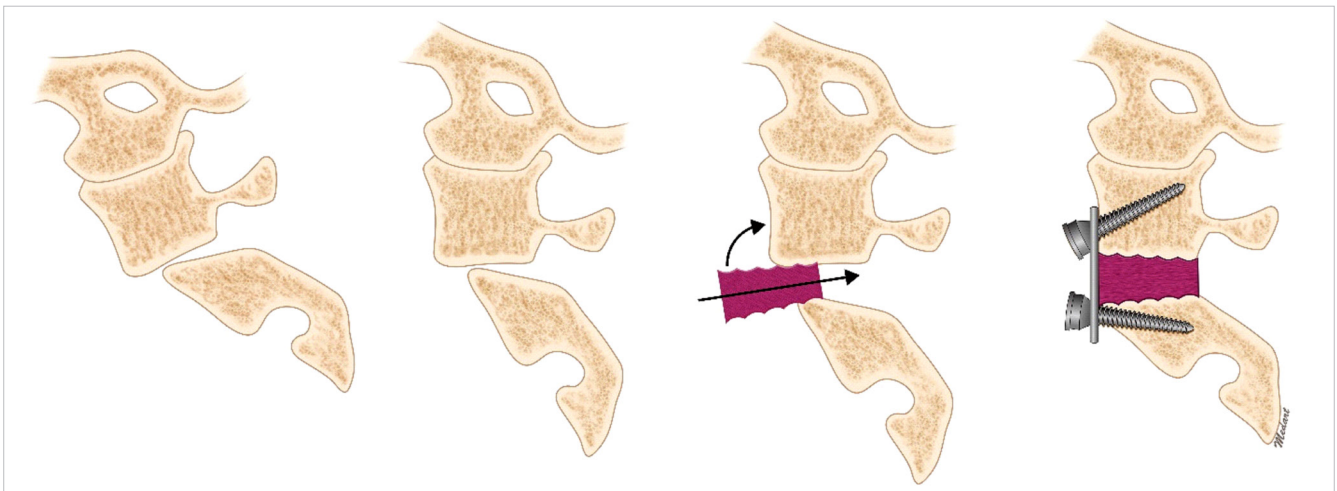


Fig. 6. The “wedge” concept.

axial dislocation, and reducing the cervicomedullary strain (Fig. 6; Supplementary video clip 2). The anterior surface of the atlas and the body of the axis have adequate bone stock to accept 4-mm cortical screws with a plate to allow rigid fixation of the atlantoaxial joints on both sides from an unilateral exposure; this technique was first described in the literature in 2013, and the wedge concept was proposed in 2016.^{18,19}

In 2007, Kothari proposed that aligning the facets of the atlas with those of the axis simulates the positioning of vertebral bodies in lumbosacral listhesis, which probably stimulated the concept of joint spacers for atlantoaxial dislocation, as interbody grafts and cages with pedicle screw fixation were already a popular treatment for reduction and fixation/fusion of lumbosacral listhesis.²⁰

The recent development of lumbar hyperlordotic anterior cages for lumbar listhesis suggests the possibility of powerful sagittal balance correction with a better distribution of vertical loading forces and a higher level of useful bone fusion.²¹ Moreover, the anterior approach to the lumbar spine results in a lower incidence of perioperative and postoperative complications such as infection, nonunion, and hardware failure. Application of this information to the craniocervical region is exciting from a biomechanical perspective and holds great potential.

CONCLUSION

The anterior unilateral submandibular extrapharyngeal approach permits bilateral cage distraction and bilateral anterior atlantoaxial fixation. It is a simple and safe option in many cases of BI.

The surgical treatment of BI needs to be chosen based on the clinical problem, radiological data, and the need to balance safety with the simplicity of the operation. Atlantoaxial fixation cannot treat all the issues related to this clinical entity. Foramen magnum decompression with duroplasty and transoral odontoidectomy may be still required in some cases until more evidence is identified or a better solution evolves.

CONFLICT OF INTEREST

The authors have nothing to disclose.

SUPPLEMENTARY MATERIALS

Supplementary video clips 1 and 2 can be found via <https://doi.org/10.14245/ns.1938172.086>.

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