Atlantoaxial Facet Fixation Using Cervical Facet Cage: Technical Case Report and Review of the Literature

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Case Report

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Abstract

In 1994, the use of interfacet spacer placement was for joint distraction, reduction, and fusion to supplement atlantoaxial or occipitocervical fixation. Here we present a unique case of bilateral atlantoaxial interfacet fixation using cervical facet cages (CFC) in a pediatric patient with basilar invagination. In addition, we review the literature on atlantoaxial facet fixation.

We present a 12-year-old boy with Wiedemann-Steiner syndrome who presented with multiple episodes of sudden neck jerking, described as in response to a sensation of being shocked, and guarding against neck motion, found to have basilar invagination with cervicomedullary compression. He underwent an occiput to C3 fusion with C1-2 CFC fixation. We also conducted a literature review identifying all publications using the keywords: “C1” AND “C2” OR “atlantoaxial” AND “facet spacer” OR “DTRAX.”

The patient demonstrated postoperative radiographic reduction of his basilar invagination from 6.4 mm to 4.1 mm of superior displacement above the McRae line. There was a 4.5 mm decrease in the atlantodental interval secondary to decreased dens retroflexion. His post-operative course was complicated by worsening of his existing dysphagia but was otherwise unremarkable. His neck symptoms completely resolved.

We illustrate the safe use of CFC for atlantoaxial facet distraction, reduction, and instrumented fixation in a pediatric patient with basilar invagination. Review of the literature demonstrates numerous materials can be safely placed as a C1-C2 interfacet spacer including bone grafts, titanium spacers, and anterior cervical discectomy and fusion cages. We argue that CFC may be included in this arsenal even in pediatric patients.

Introduction

In 1994, Goel *et al.* first described atlantoaxial fixation using screws and plates with interfacet spacer placement for joint distraction and load bearing [1]. The Goel technique allows for vertical distraction of the C1-C2 joint and thus reduction of the mobile basilar invagination prior to instrumented fixation. The material placed in the interfacetal space has evolved from bone graft to titanium cages and now includes cervical facet cages (CFC). CFCs are titanium implants used to replace damaged facet joints to restore stability and mobility. In this report, we present the first case to our knowledge of C1-C2 fixation using CFC in a pediatric patient for the treatment of basilar invagination as well as a brief review of the literature.

Case Report

History and Presentation

A 12-year-old boy with a history of Wiedemann-Steiner syndrome complicated by AV block requiring a pacemaker, submucosal cleft palate, choanal stenosis, and micrognathia with midface hypoplasia presented with multiple episodes of neck jerking and guarding against neck motion. Persistent dysphagia was also noted. Seizure workup for these episodes was negative and upon examination the patient had a Lhermitte sign with flexion. Computed tomography (CT) scan of his cervical spine revealed basilar invagination with 6.4 mm of superior displacement of the odontoid process above the McRae line, atlantodental interval (ADI) of 5.5 mm, atlantooccipital assimilation, and C2-C3 fusion (**Figure 1a**). CT myelography demonstrated evidence of compression of the cervicomedullary junction by the retroflexed odontoid process (**Figure 1a**).

Fig 1 A: Preoperative Computed Tomography (CT) Myelogram Imaging of Cervical Spine. Left image shows a sagittal CT myelogram image. Right image shows an axial CT myelogram image highlighting an altitudinal interval of 5.5mm. B: Postoperative CT Imaging of Cervical Spine. Left Image shows a sagittal CT image highlighting derotation of the dens compared to preop. Right image shows an axial CT highlighting an atlantodental interval of just 1.0mm

Surgery

The patient was taken to the operating room for an occiput to C3 fusion with placement of bilateral C1-C2 CFC joint interfacet spacers. Pre-position somatosensory evoked potentials (SSEPs) were normal and reliable. Following exposure, the C2 nerve roots were identified and sacrificed. The C1-2 joint was then drilled and two 10 mm x 5.5 mm x 2.5 mm CFCs were placed parallel to the joint under fluoroscopic guidance. The occipital plate was shaped to the curvature of the occiput and three screws, 8, 2, and 8mm were placed. C2 pars screws (20 mm on the left and 22 mm on the right) were placed under image guidance. Finally, image guidance was used to place a left C3 14 mm pedicle screw and a right C3 12 mm pedicle screw. C1 was not instrumented. Intraoperative CT was used to confirm placement of all hardware before 3.5 mm diameter titanium rods were placed on both sides and secured. All bony surfaces were then decorticated prior to placement of fusion supplements including a mixture of allograft demineralized bone matrix putty.

Outcome

Postoperatively, his neck guarding, and episodic electrical pain resolved, and he remained neurologically at his baseline. The postoperative CT of the cervical spine revealed reduction of his basilar invagination to 4.1 mm of superior displacement of the odontoid process above the McRae line, with a decrease of ADI to 1.0 mm due to decreased dens retroflexion, and stable placement of the hardware (**Figure 1b, 2**). The patient was discharged on post-operative day (POD) 6 with a cervical collar and stable neurologic exam. Unfortunately, he was readmitted with post-operative dysphagia on POD20 and ultimately required percutaneous gastrostomy tube placement for continuous feedings. The patient has followed with otorhinolaryngology for dysphagia management, and he was found to have primarily a food disinterest rather than a mechanical limitation. At three-months post-operation, radiographic imaging confirmed adequate decompression and evidence of bony fusion. At four-months post-operation, the patient was weaned from his cervical collar. He has since resumed oral intake without restrictions.

Fig 2 Postoperative X-ray Imaging of Cervical Spine. A: Lateral view of instrumentation. B: Anterior-posterior (AP) view of instrumentation

Discussion

In this article, we describe the case of a 12-year-old boy with history of Wiedemann-Steiner syndrome with basilar invagination who underwent occiput to C3 fusion with placement of bilateral C1-C2 CFC joint interspacers. Patients often require a combined endoscopic endonasal resection of the odontoid with posterior occipitocervical fusion; however, the indirect spinal canal decompression achieved using C1-C2 CFC joint interspacers with subsequent derotation of the atlantoaxial joint precluded the need for anterior decompression and thereby reduced the risk of additional vascular injury, cerebrospinal fluid leak, dysphagia, and need for tracheostomy.

Results of a literature review of posterior facet fixation of the C1-C2 joint are summarized in **Table 1**. Goel *et al.* first reported the use of iliac bone graft for C1-C2 facet fixation in 1994. From 2004-2008 Dr. Goel's group has reported a high rate of sustained radiographic reduction using hydroxyapatite and titanium spacers with a need for an anterior cervical approach for odontoid resection in only 2 of 64 total patients [2-5]. Other authors within this review utilized tricortical bone allografts [6], anterior cervical discectomy and fusion cages [7, 8], and CFC [9, 10] as used in this case report, with similarly low rates of supplementary anterior approaches. Numerous studies have demonstrated the efficacy of CFCs in treating subaxial cervical radiculopathy and augmenting fusion [11, 12]. Few studies have explored the use of CFCs for atlantoaxial fixation, which was an off-label use of the device at the time of the operation [9, 10].

Table 1 *Summary of literature review.*

Reference	Indication	n	Age (years)	Intraarticular Facet Fixation	Outcomes
Goel et al., 1994 [1]	Atlanto-axial dislocation	30	Adult	Iliac bone graft	Improvement or stabilization of neurologic function; no re-operations or new neurological symptoms reported
Goel et al., 2004 [2]	Basilar invagination	22	8-50	Iliac bone graft (8 cases), Titanium spacers (4 cases)	Sustained distraction and reduction at 6 months; no new neurologic symptoms.
Goel et al., 2005 [3]	Basilar invagination with Syringomyelia	12	14-50	Titanium spacers	Neurological improvement, sustained reduction of distraction and basilar invagination; syringomyelia not assessed.
Goel et al., 2005 [4]	Persistent basilar invagination previously treated by trans-oral decompression	3	22, 17, 18	Titanium spacers	Improved omega angle; clinically improved to be able to walk unassisted.
Goel et al., 2008 [5]	Basilar invagination	11	N/A	Titanium spacers	Neurological improvement; sustained reductions of distraction and basilar invagination at 6-month follow-up.
Yamagata et al., 2020 [6]	Rigid AAS (defined as ADI >5mm) secondary to odontoid dysplasia or os odontoideum	10	5-15 (Mean: 9.6)	Tricortical bone graft used as spacer and fulcrum	Neurosurgical Cervical Spine Scale (NCSS) scores improved in 6 cases and were maintained in 4 cases; all patients achieved increased C1-C2 height, ADI improvement, and bony fusion on post-operative imaging.
Lee et al., 2017 [7]	Rheumatoid arthritis, basilar invagination	1	67	ACDF cages	Improved pain; maintained bony fusion at 3-year follow-up.
Tominaga et al., 2019 [8]	Arthritis, basilar invagination	3	85, 75, 43	Corridor anterior cervical cage	Improved EMS scores, walking ability, and reduced

				(Globus Medical Inc., Audubon, PA)	neck pain; one case of reoperation due to rod breakage and nonunion.
Sommer et al., 2022 [9]	Odontoid fractures	5	Median: 79.6	CFC	Decrease in VAS pain scores; radiographic evidence of fusion on last follow-up.
Sommer et al., 2022 [10]	Trauma (4/9), Neoplasm (1/9), Degenerative changes (4/9)	9	Mean: 68.7+/-16.3	Cervical Facet Cage (CFC)	Significant decrease in VAS pain scores; all patients achieved bony fusion with no evidence of subsidence at last follow-up.
Goel et al., 2005 [13]	Fixed atlanto-axial dislocation	19	12-46 (Mean: 22)	Hydroxyapatite spacers (16 cases), Titanium spacers (3 cases)	13 patients with acceptable reduction (atlantodental interval<4mm); 4 with clinical improvement but no radiographic reduction; 2 patients underwent trans-oral decompression.
Kim et al., 2011 [14]	Basilar invagination	2	45, 69	8mm iliac bone autograft, Autograft iliac bone blocks	Improved Clark Station, Redlund-Johnell Criterium and Ranawat Criterion
Srivastava et al., 2017 [15]	Os odontoideum, Basilar invagination, Basilar invagination, Basilar invagination	4	16, 32, 32, 18	Unspecified spacer or wedge bone graft	Neurological improvement, improved mJOA scores; stable fusions.
Turel et al., 2017 [16]	Traumatic fractures (8), Degenerative stenosis (6), C2 neuralgia (2), C1-2 ligamentous subluxation (2), Os odontoideum (1)	19	Median: 69	Machined cortical allograft (FacetLift, Medtronic, Memphis)	94% successful arthrodesis at 6 months; one patient who did not achieve arthrodesis remained asymptomatic.
Sai Kiran et al., 2018 [17]	Basilar invagination, Os odontoideum	1	44	Unilateral right C1-2 titanium spacer with bone graft	Resolution of pre-operative symptoms; no instability on dynamic radiographs at 10-month follow-up.
Xu et al., 2019 [18]	Alagille syndrome with numerous	1	21	Small cortical bone grafts	Complete resolution of preoperative

	segmentation abnormalities				symptoms; persistent fusion and improved alignment on XR.
Jain et al., 2022 [19]	Odontoid fractures	9	23-52	Unspecified, spacers	Resolution of neck pain; complete fracture alignment in 8/9 patients; 1 patient with with grade 4 listhesis and bone loss; stable fusion at a mean follow-up of 16 months.
Anand et al.	Basilar invagination	1	12	CFC	Reduction of basilar invagination, decreased ADI; temporary gastrostomy tube placement to address dysphagia.

Our report demonstrates the safe utilization of CFC for atlantoaxial fixation in a pediatric patient. The generalizability of this claim is limited by our singular clinical experience. It is our impression that the relatively small size of the graft limits the magnitude of direct reduction potential, but the shape of the graft and angulation of the C1-C2 joint may allow for a significant amount of de-rotation of a retroflexed odontoid process relative to the C1 anterior arch. Ultimately, our current experience suggests that reduction in ADI via derotation may be more clinically significant than the decompression achieved from reducing the degree of invagination.

Conclusion

We highlight the potential safety of using CFCs as an atlantoaxial facet distraction, reduction, and fusion tool in a pediatric patient with basilar invagination. While further studies are necessary to corroborate these findings, we show that CFCs can be a powerful tool for posterior-only approach for atlantoaxial reduction and fixation in pediatric populations.

Declarations

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Competing Interest

The authors have no relevant financial or non-financial interests to disclose.

Consent

The University of Pittsburgh Institutional Review Board (IRB) approved the study "Analyses of Neurosurgery Operative Procedures" on July 27, 2020, under the reference number STUDY20050395. Written informed consent was obtained from the patient's parent for publication of the details of their medical case and any accompanying images.

Author Contribution

Conception & design: All authorsDrafted the manuscript: All authorsCritical evaluation of the manuscript: All authorsApproved the final version to be published: All authorsAgree to be accountable for all aspects of the work in ensuring that questions related to theaccuracy or integrity of any part of the work are appropriately investigated and resolved: Allauthors.

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Figures

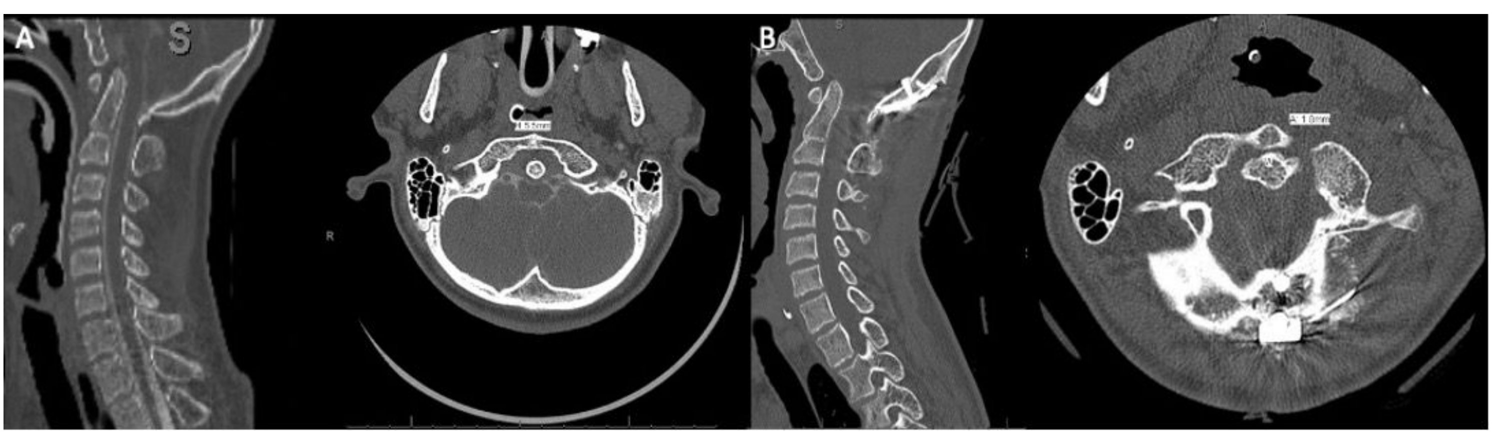

Figure 1

A: Preoperative Computed Tomography (CT) Myelogram Imaging of Cervical Spine. Left image shows a sagittal CT myelogram image. Right image shows an axial CT myelogram image highlighting an altitudinal interval of 5.5mm. B: Postoperative CT Imaging of Cervical Spine. Left Image shows a sagittal CT image highlighting derotation of the dens compared to preop. Right image shows an axial CT highlighting an atlantodental interval of just 1.0mm

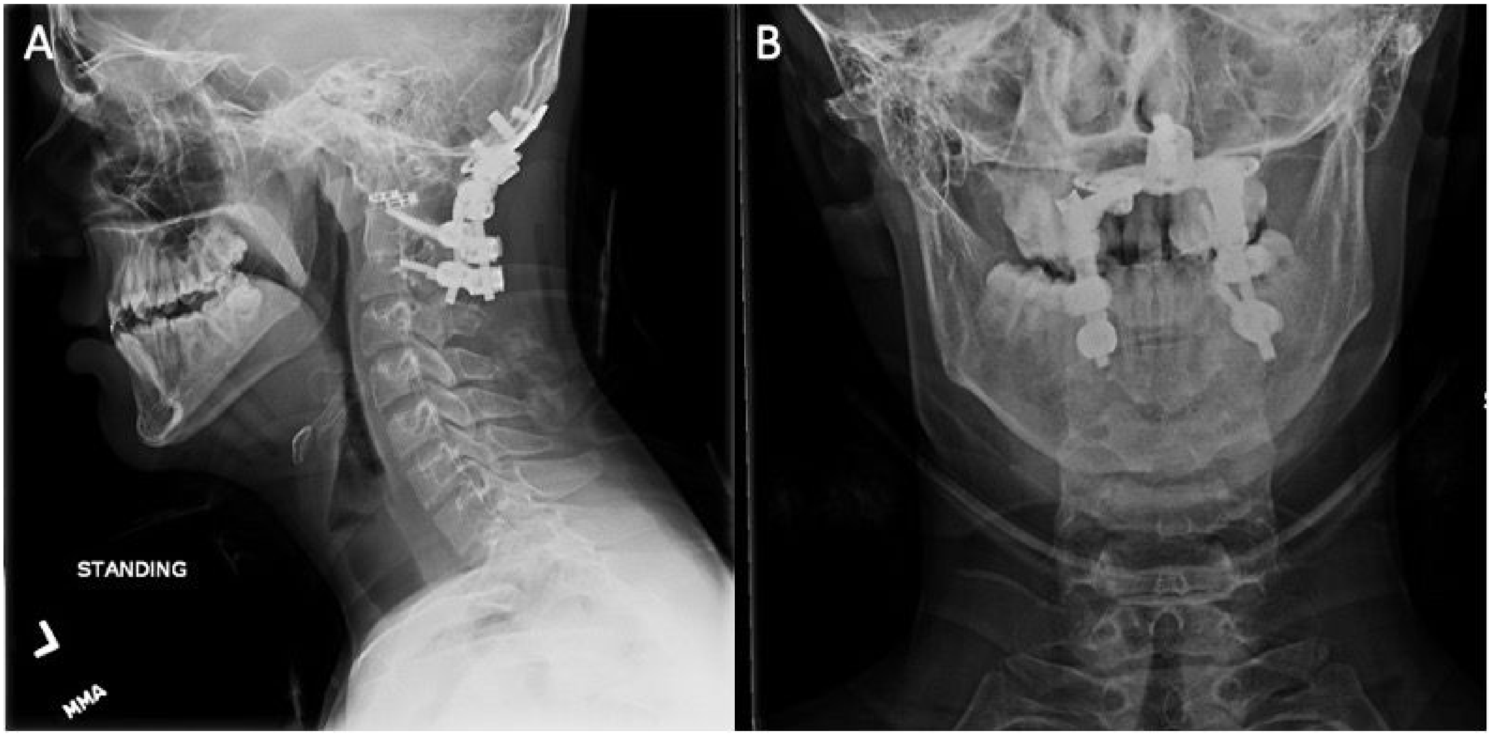

Figure 2

Postoperative X-ray Imaging of Cervical Spine. A: Lateral view of instrumentation. B: Anterior-posterior (AP) view of instrumentation