

## C1-C2 Joint Distraction and Fusion: A Robust Surgical Technique for Atlantoaxial Dislocation with Basilar Invagination

Tejesh Shavi\*, Sathwik Shetty, Kanjithanda Mandanna Bopanna, Praveen Ganigi and Khurram Khan

Apollo Hospitals Chennai Grems Road, Chennai, Tamil Nadu, India

### \*Correspondence to:

Tejesh Shavi

Apollo Hospitals Chennai Grems Road,  
Chennai, Tamil Nadu, India.

E-mail: [tejesh.shavi13@gmail.com](mailto:tejesh.shavi13@gmail.com)

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### Abstract

**Background:** Atlantoaxial dislocation (AAD) and Basilar invagination (BI) are complex craniospinal alignment pathologies with significant clinical morbidity at presentation and a potential for life threatening complications. To realign the craniospinal complex optimally various strategies have been employed over the years ranging from, transoral odontoidectomy to sublaminar wire fixation and in situ screw fixation with significant morbidity and sub-optimal outcomes. This study focuses on our experience with the current treatment strategy of C1-C2 joint distraction and fusion, in managing this complex pathology.

**Purpose:** To study the clinical and radiological outcome in patients with atlantoaxial instability undergoing posterior C1 lateral mass and C2 joint distraction and pedicle screw rod fixation.

**Study design:** Prospective clinical study with 30 patient samples.

**Outcome measures:** Clinical outcomes-Modified Japanese orthopaedic association (mJOA) score, Nurick's grading for myelopathy and visual analogue scale (VAS) for pain. Radiological outcomes-Bony fusion and alignment was analyzed by X-ray and computed tomography (CT) craniocervical junction (CVJ).

**Methods:** All adult and paediatric patients who were admitted to Manipal Hospital, Bengaluru (Karnataka, India) between 10-78 years of age with atlantoaxial instability and underwent posterior C1-C2 joint distraction and fixation from 2016 to 2021, comprised the study group. All patients were followed up immediately post operatively and at intervals of 3 months 6 months and 1 year. Clinical follow-up was done using the mJOA score, Nurick's grading for myelopathy and VAS for pain. Radiological documentation of bony fusion and alignment was done by X-ray and CT CVJ.

**Results:** 30 patients underwent the procedure during the study period. All our patients had good clinical outcome with a statistically significant ( $p < 0.01$ ) improvement in spasticity on both the mJOA and Nurick's grading. The reduction in neck pain on the VAS was also found to be statistically significant. Correction of craniocervical alignment was noted with, the odontoid process receded by a mean of 4.4 mm, 3.7 mm and 4.5 mm from the Chamberlain's line, Mc Rae's line and the Mc Gregor's line respectively with a reduction in Atlanto-dental interval (ADI) by a mean of 4.37 mm. The change in the post-operative craniometric measurements clearly indicated a significant re-alignment of the craniocervical junction towards baseline. None of the patients developed neurological worsening post-surgery, while 2 patients had superficial wound infection and another had an occipital pressure sore, all of which were managed conservatively.

**Conclusion:** C1-C2 Joint distraction-reduction-realignment is a safe and effective option to treat AAD and BI. Although this procedure has a steep learning curve, it is biomechanically stable and provides good circumferential decompression of the neural elements by a purely posterior approach.

## Keywords

Atlantoaxial dislocation, Basilar invagination, Craniovertebral junction

## Introduction

The CVJ is a complex transition zone between the skull and the upper cervical spine incorporating the brainstem and spinal cord respectively [1]. Thus, any bony abnormality at the CVJ involves not only the bony structures but also the nervous system encompassed by it [2, 3].

The atlantoaxial joint (C1-C2 joint) is characterized by a high degree of mobility and little intrinsic stability [4]. Atlantoaxial instability can occur when the ligaments and joints are damaged by trauma, inflammation, neoplasm or due to congenital defects [5]. Abnormalities that affect this complex can result in cervicomedullary neural compression, vascular compromise of the vertebrobasilar system and abnormal CSF dynamics.

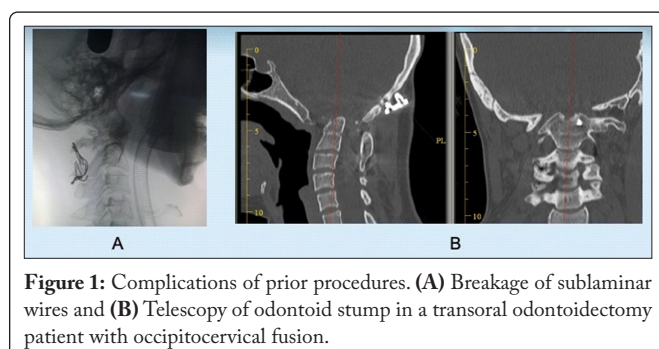
Historically, multiple techniques have been described for decompression and stabilization at the CVJ. These techniques have ranged from a posterior cable (sublaminar wires) and bone graft to transoral odontoidectomy with occipito cervical fusion. These procedures had significant limitations including wound breakdown (transoral procedures), high implant failure rates (sublaminar wires), restenosis and neural compression due to telescoping of the odontoid stump following transoral odontoidectomy and occipitocervical fusion (Figure 1).

C1-C2 joint distraction and fusion was first described by Goel et al. [6-8] (Neurosurgeon from Mumbai, India). He provided a fresh perspective and shifted the focus of the pathology from the medial C1-C2 joint to the lateral C1-C2 joints. Harms and Melcher [9] subsequently reported a modification of the Goel technique. The Harms-Goel technique has largely become the standard of treatment for atlantoaxial instability, and it involves manipulating and distracting the lateral C1-C2 joints using joint spacers followed by stabilization using C1 lateral mass screws and C2 pedicle screws connected by rods.

This prospective study looks at our patients about the clinical and radiological outcome in patients with complex craniovertebral junction anomalies.

## Material and Methods

This is a prospective study carried out in the Department of Neurosurgery, Manipal Hospital, Bengaluru. The aim was



**Figure 1:** Complications of prior procedures. (A) Breakage of sublaminar wires and (B) Telescoping of odontoid stump in a transoral odontoidectomy patient with occipitocervical fusion.

to study the clinical and radiological outcome in patients with atlantoaxial instability undergoing posterior C1 lateral mass and C2 joint distraction and pedicle screw rod fixation and fusion. All adult and paediatric patients who were admitted to Manipal Hospital, Bengaluru between 10-78 years of age with atlantoaxial instability and underwent posterior C1 lateral mass and C2 pedicle screws fixation during the study period (2016-2021) formed part of the study group. Patients with atlantoaxial instability arising from trauma, rheumatoid arthritis and congenital AAD were included in the study. Radiological evidence of ADI of >3 mm was considered mandatory for inclusion in the study. Patients with concomitant thoracic kyphosis and active pyogenic infection were excluded.

A detailed history of symptoms, demographic analysis, clinical and radiological assessment were documented in all patients operated for atlantoaxial (C1-C2) stabilization. A detailed systemic and neurological examination of all the patients was performed and recorded. Imaging included X-ray CVJ (neutral, flexion & extension views), CT CVJ with 2 and 3D reconstruction, CT Angiogram and MRI CVJ. The anatomic characteristics of the craniovertebral junction, presence of ventral or dorsal compression and atlantoaxial stability were assessed and recorded. Following clinical optimization, patients were operated under GA in prone position. The head was placed on horseshoe and cervical traction applied with Gardner-Wells tongs. A midline skin incision made frominion to C4, and underlying tissues separated subperiosteally. The dense venous plexus between C1 and C2-which can pose a significant challenge due to potential torrential bleeding-must be isolated, coagulated and cut. The C2 ganglion is mobilised or cut bilaterally to expose the lateral C1-C2 joints. The C1-C2 articular surfaces are prepped under magnification. C1 lateral mass and C2 pedicle or pars screws placed under fluoroscopic guidance taking care to avoid the vertebral artery and the spinal canal. The C1 and C2 joints were manipulated using controlled distraction, and this distraction was maintained with spacers. After optimal re-alignment was confirmed on cross table fluoroscopy, the rods were placed and torqued, and hemostasis was confirmed. Each layer of exposure was individually closed. All post-operative events including neurological change, infections, pain (VAS) were recorded. All patients were followed up immediately post operatively and at intervals of 3 months 6 months and 1 year. Follow up was done clinically and documented using the mJOA score, Nurick's grading for myelopathy, VAS for pain and radiological documentation of bony fusion and alignment by X ray CVJ, CT CVJ and MRI CVJ.

**Statistical analysis:** Data was entered into the Microsoft excel data sheet and was analysed using SPSS 22 version software. Categorical data was represented in the form of Frequencies and proportions. Chi-square test or Fischer's exact test (for 2x2 tables only) was used as test of significance for qualitative data. McNemar's test was used as test of significance for paired qualitative data. Continuous data was represented as mean and standard deviation. Paired t test is the test of significance for paired data such as pre op vs postop for quantitative data. Graphical representation of data: MS Excel and MS word were used to obtain various types of graphs such as bar diagram, Pie diagram.

p value (probability that the result is true) of < 0.05 was considered as statistically significant after assuming all the rules of statistical tests.

Statistical software like MS Excel, SPSS version 22 (IBM SPSS Statistics, Somers NY, USA) was used to analyse data.

## Results

30 patients who underwent C1-C2 joint distraction and fusion formed the study group. 66.7% were males. Traumatic cases accounted for 56.67% (17/30) of patients, with congenital causes (23.3%), rheumatoid arthritis related (13.3%) and tuberculosis related (6.6%) accounting for other pathologies in our study group. 6 patients had fixed AAD, and 24 patients had reducible AAD. The duration of symptoms ranged from 24 h to 60 months. Patients were followed up for a year. Overall, the patients had a good clinical outcome as illustrated in table 1. None of the patients developed fresh neurological deficits post-surgery. There were no implant failures. 2 patients with superficial wound infections were treated with appropriate antibiotics. 1 patient developed an occipital pressure sore which was managed conservatively.

### Clinical outcome

#### Myelopathy

Myelopathy was assessed based on the mJOA score and Nurick's grading. Pre-operatively, 16.7%, 46.7% and 26.7% of the patients had mild, moderate, and severe myelopathy respectively as per the mJOA score. 3 patients (10%) did not have myelopathy at presentation. Post-op, there were a significant number of patients (25-83.3 %) in which improvement in

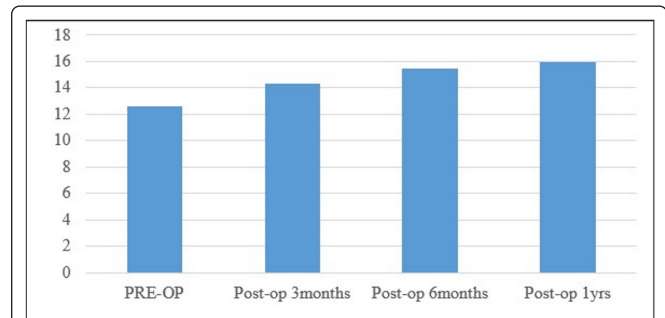


Figure 2: Pre-op and post-op mJOA score (mean).

Table 2: Comparison of mJOA score pre-op and post-op.

	Mean	St. Deviation	P value
Pre-op	12.63	4.173	< 0.01
3 months post-op	14.27	3.493	
	Mean	St. Deviation	P value
3 months post-op	14.27	3.493	< 0.01
6 months post-op	15.47	3.037	
	Mean	St. Deviation	P value
6 months post-op	15.47	3.037	< 0.01
1-year post-op	15.93	2.572	

myelopathy was noted as compared to the pre-operative status. There was no change in the state of myelopathy in 4 patients (13.3 %) and slight improvement in 1 patient (3.3 %).

In mJOA score there was statistically significant difference between pre-op and 3 months post-op score with sustained improvement at 1 year follow-up (Figure 2 and table 2).

In Nurick's grading there was statistically significant difference between pre-operative and post-operative Nurick grading at 3 months, 6 months and 1 year with a significant p value of < 0.01 (Figure 3 and table 3).

#### Pain

There was statistically significant difference between pre-op and post op 3 months 6 months and 1-year visual analog scale with a significant p value < 0.01 (Figure 4).

Table 1: Comparison of clinical signs and symptoms in patients pre and post-operatively.

Clinical features	Number of patients	
	Pre-op	Post-op
Neck pain	25	Subsided in all patients
Torticollis	4	Near complete correction
Respiratory insufficiency	3	Improved in all patients
Lower cranial nerve dysfunction	3	Improved in 2 patients
Grade of myelopathy	3	Normal
Normal	5	Improved
Mild	14	Improved in 13 patients
Moderate	8	Improved in 6 patients
Severe		No change in 3 patients
Bladder involvement	8	Complete improvement in 6 patients
Paraesthesia	15	Improved in 9 patients
Upper limb paraesthesia	13	Improved in 12 patients
Lower limb paraesthesia		Mild sensory disturbances persisted in 6 patients and 1 patient continued to have severe sensory loss

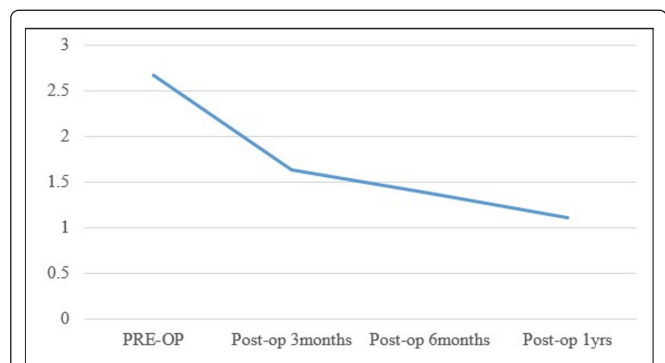
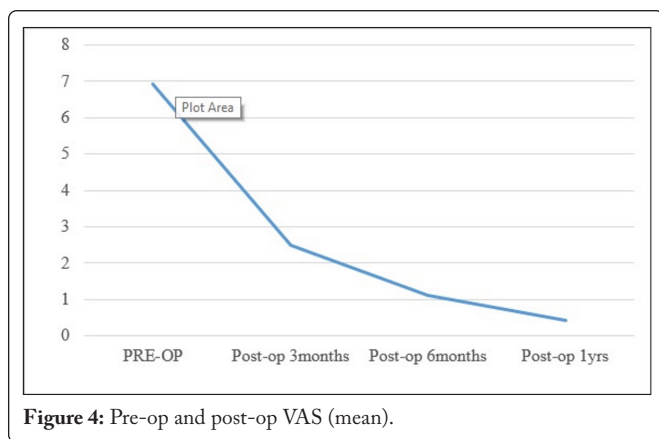


Figure 3: Pre-op and post-op Nurick's grade (mean).

**Table 3:** Comparison of Nurick grading pre-operatively and post-operatively.

	Mean	St. Deviation	P value
Pre-op	2.67	1.295	< 0.01
3 months post-op	1.63	1.351	
	Mean	St. Deviation	P value
3 months post-op	1.63	1.351	< 0.01
6 months post-op	1.37	1.377	
	Mean	St. Deviation	P value
6 months post-op	1.37	1.377	< 0.01
1-year post-op	1.10	1.398	



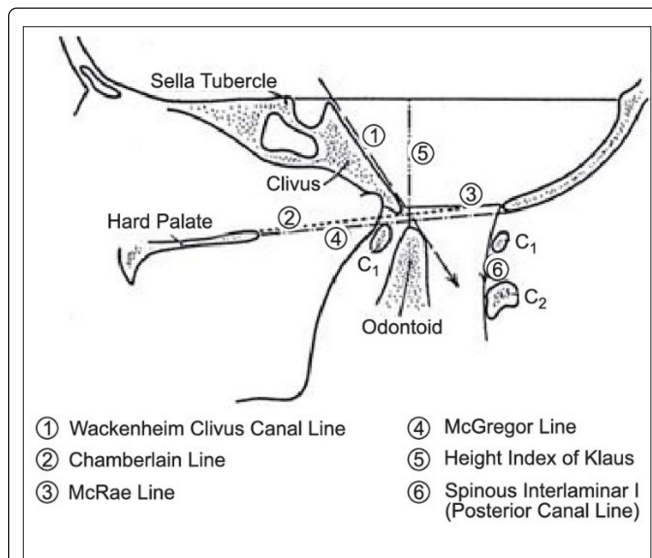
**Figure 4:** Pre-op and post-op VAS (mean).

### Craniometric measurements

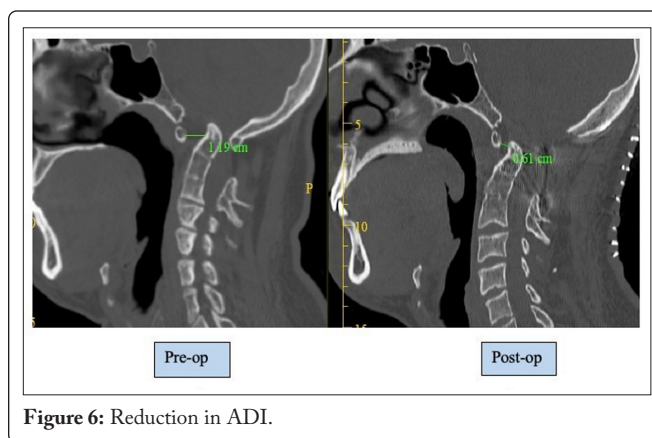
The odontoid process receded by a mean of 4.4 mm from the Chamberlain's line, 3.7 mm from McRae's line and 4.5 mm from the McGregor's line. ADI reduced by a mean of 4.37 mm. There was a statistically significant difference ( $p$  value < 0.01) between pre-op and post-op craniometric measurements with respect to ADI, Chamberlain's line, Wackenheim clivus canal line, McRae's line and McGregor's line (Figure 5 and table 4). Representative CT images of a patient have been illustrated in figure 6, figure 7 and figure 8.

### Discussion

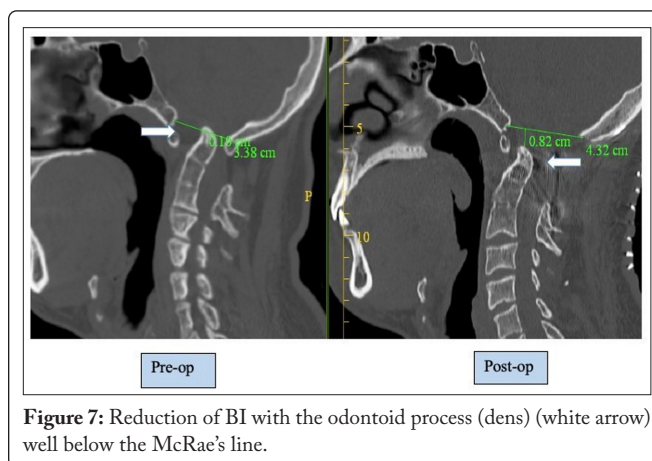
Atlantoaxial instability is a serious life-threatening condition which can occur due to a bony or ligamentous disruption at the atlantoaxial joint and can be either due to a congenital anomaly, trauma, or an inflammatory pathology such as rheumatoid arthritis and tuberculosis [10]. The presentation varies from progressive myelopathy, radiculopathy, lower cranial nerve dysfunction, or deformities of the craniocervical region. Any disruption of the atlantoaxial joint needs surgical attention with the aim of atlantoaxial fusion surgery to restore normal alignment, to ensure adequate decompression, and to achieve structural stability. The atlantoaxial complex, bearing the weight of the head and the atlantoaxial joint, generates most of the rotation of the head and neck [11]. Because of the high multiplanar mobility of the occiput-C1-C2 motion segment, fusion rates at this level are challenging with substantially lower than those achieved at the sub axial spine. The aim of treatment of atlantoaxial instability is to achieve a solid fusion between C1 and C2, virtually eliminating any motion



**Figure 5:** Illustration of lateral craniometric lines.



**Figure 6:** Reduction in ADI.



**Figure 7:** Reduction of BI with the odontoid process (dens) (white arrow) well below the McRae's line.

between them. This is expected to relieve the neck pain and avoid the risk of further neurological deficit at the expense of variably restricted neck rotation.

Myelopathy was the most common presenting feature in our patients. Following surgery, the improvement in myelopathy was ongoing with maximal improvement noted by 6 months post-operatively. The extent to which myelopathy improves post-surgery depends on the duration of the symptom and the presence of structural neural damage as seen in the spinal cord on MRI. On pre-operative MRI performed prior

**Table 4:** Pre-operative and post-operative craniometric measurements.

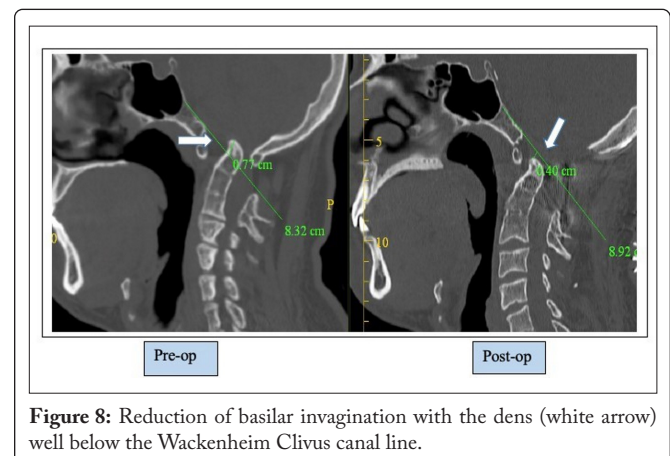
	Age	Sex	ADI		Chamberlain's line		McRae's line		McGregor's line	
			(Pre-op) mm	(Post-op) mm	(Pre-op) mm	(Post-op) mm	(Pre-op) mm	(Post-op) mm	(Pre-op) mm	(Post-op) mm
1	27	M	8.3	1.3	8.0	-3.0	-1.0	-2.0	-7.3	-6.0
2	37	M	5	3	12	1.2	7.3	-2.8	13.2	2.2
3	57	M	1.7	0.8	1.0	-0.6	-3.5	-4.0	1.0	0.6
4	56	F	5.5	2.9	6.0	-2.7	2.5	-7.1	7.0	-1.8
5	40	F	6.5	1.6	7.0	1.0	1.0	-1.0	8.0	1.2
6	51	M	7.2	1.4	6.8	1.5	0.8	-1.0	7.2	1.8
7	61	M	7.0	2.3	-2.3	-3.0	-11.0	-12.0	-1.5	-2.0
8	55	M	6.8	2.8	-10	-13.0	-12.0	-13.0	-9.0	-11.0
9	30	M	1.6	1.5	-6.0	-6.0	-7.8	-7.8	-5.7	-5.7
10	17	M	7.7	1.3	-2.5	-3.5	-4.1	-5.0	-2.1	-3.1
11	44	F	2.8	1.9	10.8	4.9	4.8	-1.2	11.6	5.4
12	13	F	6.0	1.5	-7.0	-7.0	-1.0	-2.0	-6.0	-6.0
13	40	M	6.0	2.0	7.0	4.0	1.0	-1.0	8.0	4.0
14	28	M	6.3	1.3	6.8	5.8	1.2	-2.0	7.8	6.8
15	18	M	9.0	3.0	7.0	4.0	1.0	-1.0	7.5	5.5
16	55	M	6.0	2.8	6.8	3.8	-2.0	-2.0	7.0	4.9
17	14	F	7.3	1.3	-5.0	-2.0	-1.0	-2.0	-6.3	-6.0
18	17	M	8.0	1.4	7.6	2.3	3.8	-1.0	8.0	2.6
19	42	M	5.0	4.0	-6.0	-6.0	-1.0	-2.0	-6.3	-6.0
20	25	M	7.0	2.8	13.5	9.2	4.2	0.0	15.0	10.0
21	39	M	7.2	1.4	7.8	2.5	3.6	-1.0	8.2	2.8
22	58	M	8.9	2.9	13.8	4.7	7.1	-2.8	14.2	5.4
23	20	F	8.4	3.2	10.6	-6.7	7.8	-9.0	9.0	-5.3
24	28	M	8.7	5.8	5.3	-2.5	-1.6	-3.5	6.0	2.0
25	58	F	6.2	3.0	18.2	6.2	9.2	1.1	20.0	7.0
26	25	M	11.9	6.1	6.7	-1.4	1.0	-8.2	7.4	-0.8
27	14	F	10.3	2.3	10.0	2.0	1.0	-1.0	11.0	3.0
28	51	F	6.1	3.1	10.2	5.0	4.8	-2.3	11.0	5.8
29	13	F	6.8	3.1	6.5	-1.6	1.6	-4.2	-7.5	-0.5
30	36	M	11.3	3.7	4.5	3.7	-1.5	-3.9	5.5	4.6

to surgery in all our patients, the following were found: a syrinx in 3.3% of patients, T2 cord signal changes/hyperintensity in 20% of patients, and cord compression in 86.7% of patients. Prior studies [12, 13] noted that T2 signal hyperintensity was associated with poor neurologic outcome in patients with cervical spondylotic myelopathy. We repeated an MRI in patients in which there was no clinical improvement post-surgery and noted a persistent non resolved T1/T2 signal intensity change in this subgroup of patients highlighting the fact that these changes represent chronic compression and irreversible myelopathic changes. In our study, the overall improvement in myelopathy was 86.7%, with no improvement seen in 13.3% of patients at 1 year follow up.

**Technique**

Several types of atlantoaxial fusion techniques have been described over the years from the earliest no-screw fusion (Gallie, Brooks, and Sonntag) to the two screws fusion (trans-articular technique of Magerl and Seemann) and four screws

technique (Goel and Harms) with its variations. The no screw techniques have been used for decades and are technically simple to perform but with high non fusion rates, neurological complications, and biomechanical weakness. The two screws technique (transarticular screws) is associated with a risk of



**Figure 8:** Reduction of basilar invagination with the dens (white arrow) well below the Wackenheim Clivus canal line.

vertebral artery injury higher than other techniques. The four-screw technique (C1 lateral mass and C2 pedicle screw) which was introduced by Goel and Laheri and then modified by Harms and Melcher with the use of polyaxial screws and rods instead of plates has reached a similar popularity being simpler, with much lower risk of vertebral artery injury and higher fusion rate 100% [14-16].

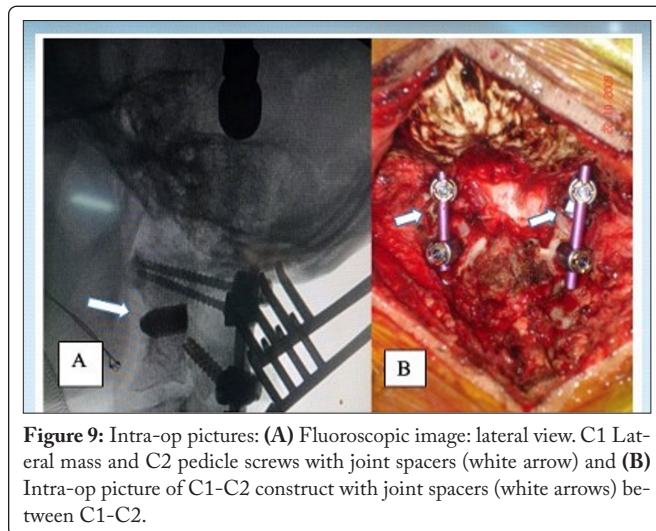
Goel et al. [6-8] technique of C1-C2 joint distraction and fusion using C1 lateral mass and C2 pedicle screw (Figure 8 and figure 9) has obviated the need for anterior decompression (transoral odontoidectomy). This technique shifted the focus of the pathology from the median atlantoaxial joint (between the dens and anterior arch of atlas) and is directed at correcting the subluxation and instability at the lateral C1-C2 joints which is now considered the primary pathology in these patients. It involves manipulating the lateral C1-C2 joint thereby relieving the anterior compression from a posterior approach. This realignment of atlas and axis vertebrae with fixation has a reported 100% fusion rate [6-8]. Multiple subsequent studies [17-20] have shown this procedure to be

reliable with excellent fusion rates. We achieved bony fusion in all (100%) of our patients along with a statistically significant improvement both in neurological and radiological outcomes. The challenge with this technique lies in effectively restoring a normal craniovertebral alignment, controlling bleeding from the large C1-C2 venous plexus, and avoiding injury to the vertebral artery laterally and the spinal cord medially. Further, this technique may need to be individualised based on the peculiar osseous anatomy often encountered intra-operatively especially in congenital AADs. Some patients with hypoplastic posterior elements would require a longer construct involving the occiput and the sub axial cervical spine-Occipitocervical fusion. Figure 9 and figure 10 show the intra-op and post-op images of this technique.

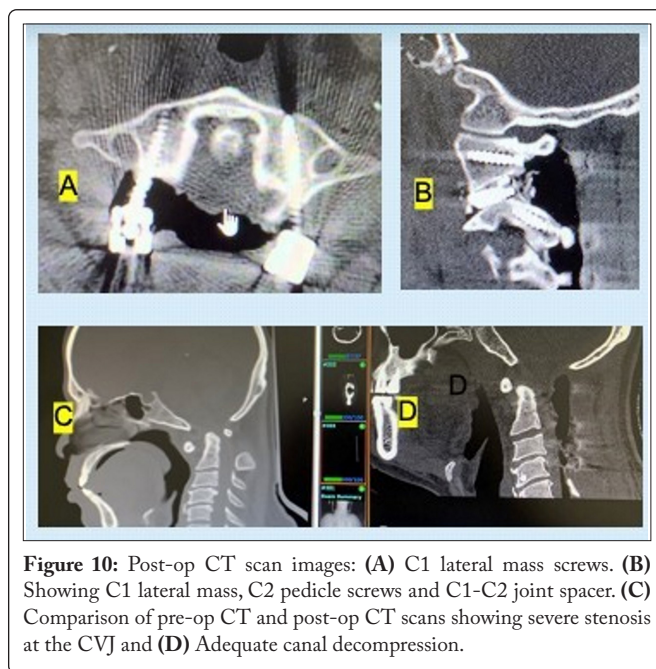
### Modifications

#### DCER technique

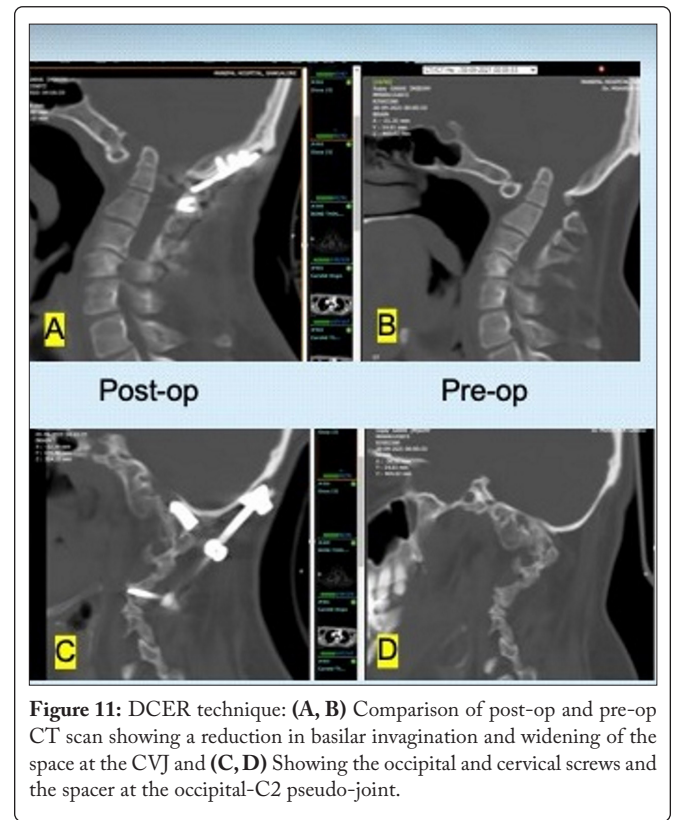
Recently, in patients with occipitalised C1 arch (C1 posterior arch fused to the occiput) presenting with moderate to severe basilar invagination, DCER (Distraction-Compression-Extension-Reduction) technique [21, 22] has been found to be useful in both reducing the basilar invagination and correcting the cervical hyper-lordosis seen in these patients. Although we have not included this subgroup of patients in this study, we have used this



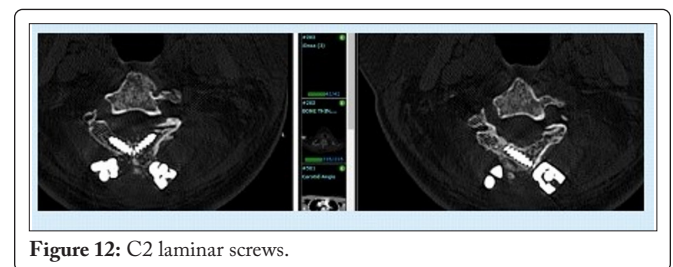
**Figure 9:** Intra-op pictures: (A) Fluoroscopic image: lateral view. C1 Lateral mass and C2 pedicle screws with joint spacers (white arrow) and (B) Intra-op picture of C1-C2 construct with joint spacers (white arrows) between C1-C2.



**Figure 10:** Post-op CT scan images: (A) C1 lateral mass screws. (B) Showing C1 lateral mass, C2 pedicle screws and C1-C2 joint spacer. (C) Comparison of pre-op CT and post-op CT scans showing severe stenosis at the CVJ and (D) Adequate canal decompression.



**Figure 11:** DCER technique: (A, B) Comparison of post-op and pre-op CT scan showing a reduction in basilar invagination and widening of the space at the CVJ and (C, D) Showing the occipital and cervical screws and the spacer at the occipital-C2 pseudo-joint.



**Figure 12:** C2 laminar screws.

technique (Figure 11) successfully and with good neurological outcome in patients with complex CVJ anomalies.

### C2 laminar screws

It's an alternative technique to C2 pedicle screw fixation, especially in patients with pedicle deformity of C2 and in patients with unilateral occlusion of vertebral artery (Figure 12).

## Conclusion

This study reiterates the effectiveness of joint distraction, reduction, and fusion in basilar invagination with both fixed and reducible AAD. Although this procedure has a steep learning curve, with thorough pre-operative planning and meticulous operative technique it provides good anterior decompression of the neural elements by a purely posterior approach. It is biomechanically stable and works towards restoring the normal C1-C2 height. The technique needs to be individualized based on the anatomy, which can be highly variable.

C1-C2 Joint distraction-reduction-realignment, achieved with the help of interlaminar spacers and instrumented fusion, is a safe and effective option to treat AAD and BI.

## Acknowledgements

None.

## Conflict of Interest

None.

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