Original Article

Adjacent-segment "central" atlantoaxial instability and C2–C3 instability following lower cervical C3–C6 interbody fusion: Report of three cases

ABSTRACT

Aim: We report adjacent-segment "central" or "axial" atlantoaxial instability and C2–C3 instability as the cause of delayed neurological worsening following multisegmental cervical spinal stabilization.

Materials and Methods: Three male patients aged 34, 56, and 70 years had been operated earlier for cervical spondylosis by multilevel C3–C6 cervical interbody fusion 6–11 years earlier. After an initial improvement for few years, the patients observed relatively rapid clinical deterioration. When admitted, all the three patients were severely quadriparetic and were brought to the hospital on a wheelchair. Central atlantoaxial instability was diagnosed on the basis of our previously published clinical and radiological parameters. C2–C3 instability was essentially diagnosed on the intraoperative observations. The patients underwent atlantoaxial and C2–C3 fixation.

Results: All the three patients had rapid clinical recovery that started in the immediate postoperative period. At an average follow-up of 21 months, the patients walked independently.

Conclusions: Identification and treatment of adjacent-segment central atlantoaxial and C2–C3 instability can lead to gratifying clinical outcome.

Keywords: Adjacent-segment instability, central atlantoaxial instability, cervical interbody fusion

INTRODUCTION

Adjacent-segment spinal degeneration has been frequently identified following single or multilevel lumbar or cervical spinal decompression-fixation surgery for spinal spondylosis.^[1,2] We report our experience of adjacent-segment atlantoaxial and C2–C3 instability following multilevel cervical spinal interbody fusion surgery. Our literature survey did not locate such a report. The surgical results following treatment by atlantoaxial and C2–C3 fixation are reported.

MATERIALS AND METHODS

During March 2017 to June 2018, three male patients, aged 34, 56, and 70 years, who were operated several years earlier for multilevel cervical degeneration by C3–C6 cervical interbody fusion, presented with relatively recent-onset worsening in the clinical neurological function. Table 1 summarizes the

Access this article online				
	Quick Response Code			
Website: www.jcvjs.com				
DOI: 10.4103/jevjs.JCVJS_7_20				

neurological grade on admission. All patients were admitted in a clinical condition that they needed wheelchair mobilization. Apart from Goel Clinical Grading Scale, validated clinical assessment parameters of Japanese Orthopedic Association Score and Visual Analog Scale were used both before and after surgery and at follow-up assessment [Table 1]. All

Atul Goel^{1,2}, Shashi Ranjan¹, Abhidha Shah¹, Survendra Rai¹, Saswat Dandpat¹, Abhinandan Patil¹, Ravikiran Vutha¹

¹Department of Neurosurgery, KEM Hospital and Seth GS Medical College, ²Lilavati Hospital and Research Centre, Mumbai, Maharashtra, India

Address for correspondence: Prof. Atul Goel, Department of Neurosurgery, KEM Hospital and Seth GS Medical College, Parel, Mumbai - 400 012, Maharashtra, India. E-mail: atulqoel62@hotmail.com

Submitted: 15-Jan-20 Accepted: 16-Jan-20 Published: 04-Apr-20

This is an open access journal, and articles are distributed under the terms of the Creative Commons Attribution-NonCommercial-ShareAlike 4.0 License, which allows others to remix, tweak, and build upon the work non-commercially, as long as appropriate credit is given and the new creations are licensed under the identical terms.

For reprints contact: reprints@medknow.com

How to cite this article: Goel A, Ranjan S, Shah A, Rai S, Dandpat S, Patil A, *et al.* Adjacent-segment "central" atlantoaxial instability and C2–C3 instability following lower cervical C3–C6 interbody fusion: Report of three cases. J Craniovert Jun Spine 2020;11:51-4.

© 2020 Journal of Craniovertebral Junction and Spine | Published by Wolters Kluwer - Medknow

patients underwent dynamic (flexion-extension of neck) plain radiographs, computed tomography scan, and magnetic resonance imaging. All the three patients had undergone anterior cervical discectomy with fusion from C3 to C6 levels.

There was no evidence of instability of any segment of cervical spine including atlantoaxial joint when assessed by conventional radiological parameters. Atlantoaxial instability was diagnosed on the basis of alignment of facets of atlas and axis on lateral profile imaging with the head in neutral position and by observations on direct physical manipulation of bones during surgery.^[3] As per the classification, Type 1 instability was when the facet of atlas was dislocated anterior to the facet of axis and Type 2 instability was when the facet of atlas was dislocated posterior to the facet of axis. Type 3 atlantoaxial instability was when the facets of atlas and axis were in alignment and atlantoaxial instability was diagnosed on the basis of direct bone handling and manipulation during surgery. Two patients had Type 2 and one had Type 3 atlantoaxial facetal instability. As the atlantodental interval may not be pathologically altered, there may not be any direct indentation of the dural tube and neural structures, and the subarachnoid spaces may be entirely preserved; Types 2 and 3 atlantoaxial instability was labeled as central atlanto-axial dislocation (CAAD).^[3-5] Accordingly, all the three patients had CAAD. In none of the patients, there was any evidence of neural compression opposite to the odontoid process. C2–C3 instability was diagnosed on the basis of physical observations of joint status and bone manipulations during surgery. Presence of excessive movements, osteophytes in the vicinity, and presence of direct evidence of facetal listhesis were indicators of instability. There was no evidence of C2-C3 instability on imaging, and there was no dural or neural compression opposite to the C2–C3 articulation or disc space.

Surgical treatment

All patients underwent atlantoaxial and subaxial C2-C3 fixation. The technique described in 1994 by the first author was deployed for atlantoaxial fixation.^[6,7] Cervical Gardner-Wells traction was applied after induction of anesthesia. The patients were placed in prone surgical position, and the head end of the operation table was elevated by about 30°. After a midline incision, the craniovertebral junction and the upper cervical spine were exposed. In addition to the clinical and radiological guides, handling and manipulation of the spinous process of the axis bone was done to assess the spinal instability. At surgery, atlantoaxial facetal joint was widely opened, bone graft pieces were packed into the articular cavity, and, subsequently, plate and screw fixation was done. C2-C3 fixation was done by transarticular facetal fixation technique described by Roy-Camille and Saillant in 1972.^[8] [Figure 1] All muscles attached to the spinous process of axis, arch of atlas, and spinous process of C3 vertebra were sharply cut and removed. The host bone of the posterior elements of C1-C3 was decorticated. Bone graft harvested from the iliac crest was placed and stabilized in the region. After completion of surgery and returning the patient to supine position, the traction was removed. The patients were placed in a firm cervical collar, and neck movements were restricted for about 4 weeks. The patients were then asked to carry on their routine activities.

RESULTS

Clinical improvement in all symptoms and major sensory and motor functions was observed in the "immediate" postoperative period. The clinical recovery progressed during the period of follow-up. The follow-up of the patients

Table 1: The pre- and postoperative clinical parameters of the pa

Case	Goel Clinical Grade		JOA score		VAS	
number	Preoperative	Postoperative (12 months)	Preoperative	Postoperative (12 months)	Preoperative	Postoperative (12 months)
1	4	2	11	15	7	1
2	5	2	7	14	9	2
3	4	1	8	16	6	0

JOA - Japanese Orthopedic Association Score; VAS - Visual Analog Scale

Table 2: Patient Satisfaction Index

Parameter	Score 0 (not satisfied)	Score 1 (minimally satisfied)	Score 2 (satisfied)	Score 3 (remarkably satisfied)
Are you happy with the operation?	-	-	-	3
Are you relieved of sensory symptoms?	-	-		3
Can you make your fist/handgrip better?	-	-		3
Can you move your shoulders better?	-	-	1	2
Can you walk better?	-	-	-	3

ranged from 12 to 28 months. The clinical status at follow-up is summarized in Table 1. At a minimum follow-up of 12 months, all patients were able to walk independently. Patient Satisfaction Score was calculated on the basis of questions posed to the patient and to the immediate relative [Table 2]. All the three patients were "highly" satisfied with the outcome of surgery.

DISCUSSION

The general understanding is that neurological symptoms are a result of direct neural compression or deformation. Recent studies have identified that rather than compression, neurological symptoms are secondary to instability-related subtle and repeated micro-injuries to neural structures.^[9,10] There could be instability of the spinal segments even when the bones are in alignment on dynamic imaging.^[5,11] Our recent classification identifies atlantoaxial instability (CAAD) even in the absence of any bone mal-alignment or direct neural or dural compression by odontoid process.

Understanding the clinical entity of CAAD has expanded the horizons of surgical understanding of craniovertebral junction. CAAD has been identified to be the nodal primary or a major point of pathogenesis of commonly treated clinical ailments such as Chiari formation^[12] and basilar invagination^[13] and in cases with cervical myelopathy related to single- or multiple-level cervical spondylosis,^[14,15] ossified posterior longitudinal ligament,^[16] and Hirayama disease.^[17] CAAD is generally a chronic or long-standing instability that leads to subtle, relentlessly progressive, and ultimately disabling clinical symptoms. It was observed that CAAD is "frequently" associated with multisegmental cervical spondylosis, particularly in those patients who are "old" and those presenting with disproportionately severe neurological symptoms. Ignoring the presence of CAAD in cases with cervical spondylotic myelopathy can be an important cause of surgical failure.

Adjacent-segment spinal degeneration is a well-described clinical entity and has been recorded both in the lumbar and cervical spines following single- or multiple-level surgery for spinal degeneration.^[1,2] However, description of high cervical or atlantoaxial instability as an adjacent-segment affection following lower cervical surgical treatment for cervical spondylotic disease has not been described. All our patients had improved following the previous surgery that involved multisegmental interbody cervical fusion. This fact suggests that there was no atlantoaxial instability at the time of first surgery. The exact cause of delayed (average 8.3 years) and severe neurological worsening could not be correlated with a parallel radiological evidence of neurological compression.

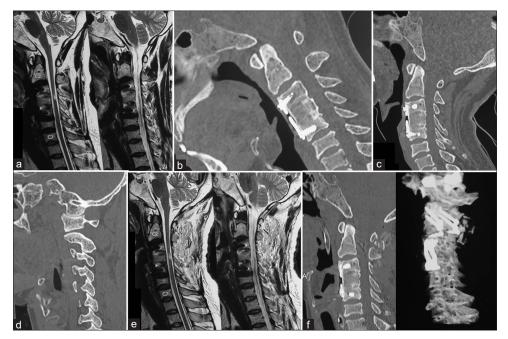


Figure 1: Images of a 34-year-old male patient. The patient had been operated 6 years before presentation. A C3–C4 and C4–C5 discectomy with anterior plating had been successfully performed. (a) T2-weighted magnetic resonance imaging showing the anterior fixation from C3 to C5 level. There is no evidence of compression at C1–C2 or C2–C3 levels. (b) Computed tomography scan with the head in flexion showing C3–C4 and C5 fusion and anterior plate. There is no evidence of C1–C2 or C2–C3 instability. (c) Computed tomography scan with the head in extension. The odontoid process shows minimal realignment when compared to the image on flexion. (d) Sagittal cut of computed tomography through the facets showing Type 3 atlantoaxial facetal instability. (e) Postoperative T2-weighted magnetic resonance imaging. (f) Reconstruction of computed tomography scan image showing atlantoaxial and C2–C3 fixation implants

In none of our patients, there was any evidence of significant compression of the dural tube or the neural structures opposite to the odontoid process or C2–C3 joint or disc space. Abnormal alteration of atlantodental interval that has been a sole parameter to describe mobile atlantoaxial dislocation for over 75 years was not affected in any case. CAAD was diagnosed on the basis of our recently described classification. C2–C3 instability was diagnosed on direct observation of the joint status and by bone manipulations.

Following atlantoaxial and subaxial C2–C3 stabilization, all patients had remarkable clinical recovery that started in the immediate postoperative phase. The patients progressively improved to the extent that they could walk independently and carry out routine housework without any assistance. The clinical recovery emphasizes the role of instability to produce neurological symptoms without neurological compression.

CONCLUSIONS

Identification and treatment of CAAD and C2–C3 instability as an adjacent-segment disease following cervical spinal fusion can lead to gratifying clinical recovery.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

- Bydon M, Xu R, De la Garza-Ramos R, Macki M, Sciubba DM, Wolinsky JP, *et al.* Adjacent segment disease after anterior cervical discectomy and fusion: Incidence and clinical outcomes of patients requiring anterior versus posterior repeat cervical fusion. Surg Neurol Int 2014;5:S74-8.
- 2. Xia Y, Xu R, Kosztowski TA, Ramhmdani S, Ahmed AK, Lo SL, et al.

Reoperation for proximal adjacent segment pathology in posterior cervical fusion constructs that fuse to C2 vs. C3. Neurosurgery 2019;85:E520-6.

- 3. Goel A. Only fixation for lumbar canal stenosis: Report of an experience with seven cases. J Craniovertebr Junction Spine 2014;5:15-9.
- Goel A. A review of a new clinical entity of 'central atlantoaxial instability': Expanding horizons of craniovertebral junction surgery. Neurospine 2019;16:186-94.
- Goel A, Dhar A, Shah A, Jadhav D, Bakale N, Vaja T, *et al.* Central or axial atlantoaxial dislocation as a cause of cervical myelopathy: A report of outcome of 5 cases treated by atlantoaxial stabilization. World Neurosurg 2019;121:e908-16.
- Goel A, Desai KI, Muzumdar DP. Atlantoaxial fixation using plate and screw method: A report of 160 treated patients. Neurosurgery 2002;51:1351-6.
- 7. Goel A, Laheri V. Plate and screw fixation for atlanto-axial subluxation. Acta Neurochir (Wien) 1994;129:47-53.
- Roy-Camille R, Saillant G. Surgery of the cervical spine. 2. Dislocation. Fracture of the articular processes. Nouv Presse Med 1972;1:2484-5.
- Goel A. Is it necessary to resect osteophytes in degenerative spondylotic myelopathy? J Craniovertebr Junction Spine 2013;4:1-2.
- Goel A. Vertical facetal instability: Is it the point of genesis of spinal spondylotic disease? J Craniovertebr Junction Spine 2017;6:47-8.
- Goel A. Beyond radiological imaging: Direct observation and manual physical evaluation of spinal instability. J Craniovertebr Junction Spine 2017;8:88-90.
- Goel A. Is atlantoaxial instability the cause of Chiari malformation? Outcome analysis of 65 patients treated by atlantoaxial fixation. J Neurosurg Spine 2015;22:116-27.
- Goel A, Sathe P, Shah A. Atlantoaxial fixation for basilar invagination without obvious atlantoaxial instability (Group B Basilar Invagination): outcome analysis of 63 surgically treated cases. World Neurosurg 2017;99:164-70.
- Goel A. Atlantoaxial and subaxial cervical spinal fixation: Can it revolutionize surgical treatment of cervical myelopathy related to ossified posterior longitudinal ligament? J Craniovertebr Junction Spine 2017;8:5-8.
- Goel A. Posterior atlantoaxial 'facetal' instability associated with cervical spondylotic disease. J Craniovertebr Junction Spine 2015;6:51-5.
- Goel A. Is atlantoaxial instability the cause of "high" cervical ossified posterior longitudinal ligament? Analysis on the basis of surgical treatment of seven patients. J Craniovertebr Junction Spine 2016;7:20-5.
- Goel A, Dhar A, Shah A. Multilevel spinal stabilization as a treatment for Hirayama disease: Report of an experience with five cases. World Neurosurg 2017;99:186-91.