Is C2-3 fusion an evidence of atlantoaxial instability? An analysis based on surgical treatment of seven patients

Abstract

Objective: The authors analyze the rationale of atlantoaxial fixation in patients presenting with symptoms related to cervical myelopathy and wherein the radiological images depicted C2–3 fusion and presence of single or multiple level neural compression of the subaxial cervical spinal cord attributed to “degenerative” spine.

Materials and Methods: Seven adult males were analyzed who presented with long-standing symptoms of progressive cervical myelopathy and where imaging showed presence of C2–3 fusion, no cord compression related to odontoid process, and evidence of single or multiple level lower cervical cord compression conventionally attributed to spinal degeneration. There was no other bone or soft tissue abnormality at the craniovertebral junction. There was no evidence of atlantoaxial instability when assessed by conventional radiological diagnostic parameters. Atlantoaxial instability was diagnosed on the basis of clinical understanding, atlantoaxial facetal malalignment, and manual assessment of instability by bone handling during surgery. All the seven patients underwent atlantoaxial fixation and no surgical manipulation at lower cervical spinal levels.

Results: At an average follow-up of 34 months, all patients have recovered satisfactorily in their neurological function.

Conclusion: The presence of C2–3 fusion is an indication of atlantoaxial instability and suggests the need for atlantoaxial stabilization. Effects on the subaxial spine and spinal cord are secondary events and may not be surgically addressed.

Keywords: Atlantoaxial instability, C2–3 fusion, spinal degeneration

Introduction

We report cases of seven adult males who presented with symptoms related to cervical myelopathy. On investigation, the patients had C2–3 fusion and radiological evidence suggestive of spinal cord compression related to disk herniation/osteophytes as generally identified in cases with spinal degeneration at a single or multiple lower cervical spinal levels. There was no evidence of reduction of subarachnoid spaces or direct cord compromise/compression or signal alterations of neural tissues at the C1–2 level. There was no other musculoskeletal craniovertebral abnormality. On the basis of recently described novel classification that is based on evaluation of atlantoaxial facetal alignments and other clinical and radiological evidences, “central” atlantoaxial instability was identified and the patients were successfully treated by atlantoaxial fixation.[1,2] No surgical procedure was carried out at the subaxial cervical spine. The rationale of the treatment is discussed.

Materials and Methods

Seven patients who were treated in the neurosurgery departments of the authors between August 2013 and March 2019 comprised the study material. All patients were male and their ages ranged from 18 to 60 years (average...
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40.5 years). These consecutively treated cases have been studied retrospectively. The patients provided written informed consent before surgery, and clinical tests and surgical procedures were conducted according to principles of declaration of Helsinki. As the study is a retrospective analysis of data and previously published surgical techniques were deployed, ethics committee permission was not deemed necessary.

Clinical profile
The clinical and radiological profile of the patients is summarized in Tables 1 and 2. Goel’s clinical grading scale,[3] the Japanese Orthopaedic Association,[4] and visual analog scale[5] scoring systems were used to clinically evaluate the patients both before and after the surgical treatment and at follow-up.

Table 1: Presenting signs and symptoms

| Signs and symptoms       | Number of patients |
|--------------------------|--------------------|
| Stiffness in all 4 limbs | 7                  |
| Weakness                 | 6                  |
| Sensory complaints       | 5                  |
| Neck pain                | 7                  |
| Speech difficulty        | 1                  |
| Urinary complaints       | 1                  |

Table 2: The radiological findings

| Case number | Sex/age | Radiological level of subaxial compression | Type of atlantoaxial facetal instability |
|-------------|---------|--------------------------------------------|-----------------------------------------|
| 1           | Male/48 | C3-6                                       | 3                                       |
| 2           | Male/40 | C5-6                                       | 3                                       |
| 3           | Male/31 | C3-4                                       | 3                                       |
| 4           | Male/18 | C3-4                                       | 3                                       |
| 5           | Male/60 | C3-4, C5-6, C6-7                          | 2                                       |
| 6           | Male/31 | C3-4                                       | 3                                       |
| 7           | Male/56 | C3-4, C4-5, C5-6, C6-7                    | 3                                       |

Radiological analysis
Patients underwent preoperative dynamic plain radiographs and computed tomography (CT) scan with the head in neutral, flexed, and extended positions and magnetic resonance imaging (MRI) of the craniovertebral junction and cervical spine. Imaging showed clear evidences of disk bulge/intervertebral ligamentous buckling or osteophyte related spinal cord compression in the subaxial spinal region [Figures 1 and 2]. The levels of obvious neural compression on MRI are depicted in Table 2. There was C2–3 fusion in all patients. There was no evidence of neural compression opposite the odontoid process. There was no abnormality in the atlantodental interval on dynamic flexion–extension of head imaging. Patients having associated or additional bone anomalies like assimilation of atlas; bifid arches of atlas, basilar invagination, Chiari formation and syringomyelia have not been included in the study.

Atlantoaxial instability was assessed by our recently described alternative classification.[1] Alignment of facets of atlas and axis was observed in lateral profile imaging in neutral head position. Type 1 atlantoaxial instability was when the facet of atlas was dislocated anterior to the facet of axis. Type 2 atlantoaxial facetal 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 instability was diagnosed only on the basis of corroborative clinical and radiological evidences. In both Types 2 and 3 atlantoaxial instability abnormal alteration in the atlantodental interval and dural and neural compression opposite the odontoid process is not the hallmark. These two types of atlantoaxial instability are labeled as “central” or “axial” atlantoaxial instability.[1,2] All patients had central or axial atlantoaxial instability. One patient had Type 2 and

Figure 1: Images of a 31-year-old male patient. (a) T2-weighted magnetic resonance imaging showing C2–3 fusion and signal alteration of cord opposite C3–4 disk space. (b) Computed tomography scan showing C2–3 fusion. (c) Computed tomography scan showing the facets of atlas and axis in alignment and C2–3 facetal fusion. (d) Postoperative computed tomography scan showing atlantoaxial fixation. (e) Postoperative computed tomography scan with the cuts passing through the facets showing the implant.
six patients had Type 3 atlantoaxial instability. Essentially, all patients having C2–3 fusion were considered to have atlantoaxial instability.

**Operative procedure**

Surgery involved atlantoaxial lateral mass plate/rod and screw fixation with the techniques described by the first author in 1994 and 2004.[6,7] The basic surgical steps involved opening of the atlantoaxial articulation, manipulation and distraction of the facets, denuding the articular capsule, packing of bone graft (harvested from iliac crest) within the joint cavity and direct plate/rod and screw fixation of the lateral masses of atlas and axis. Muscle attached to the posterior elements of atlas and axis was sharply cut. Bone graft was additionally placed in the posterior midline after appropriately preparing the host bone by drilling of its outer cortex. No surgical treatment was done in the subaxial spinal region.

The constant observation was that there was clear evidence of atlantoaxial instability on direct manipulation of the bones of the region. In all cases, the articular cartilage was well formed and the joints were seemingly functionally active.

The patients were advised to wear a firm external cervical collar for a period of 3 months after surgery and all neck activities and movements were restricted. After this period, all movements were permitted.

**RESULTS**

The clinical follow-up ranged from 6 to 74 months (average 34 months). Postoperative CT scan was obtained 3 months after surgery and at subsequent follow-up [Figures 1 and 2]. Tables 3-5 show the clinical outcome at the time of the last follow-up. Symptomatic and neurological improvement was observed in the immediate postoperative period and the improvement continued to progress. There were no infection or any other implant-related complications.

**DISCUSSION**

C2–3 fusion is a relatively frequent radiological observation. No specific or focused clinical implication to the presence of C2–3 fusion has been ascribed in the literature. Our earlier studies identified C2–3 fusion in 234 out of 510 cases in association with Group A basilar invagination,[8] in 19 out of 75 cases with Group B basilar invagination,[9,10] in 24 out of 70 cases in association with bifid arches of atlas,[11] and in 20 out of 190 cases in association with os-odontoideum. In all these cases, atlantoaxial fixation was done as treatment and C2–3 fusion was identified to be one of the secondary manifestations and no direct surgical treatment of any kind was done for any subaxial musculoskeletal abnormality even when that lead to neural alteration, deformation, or compression.

Patients presented with symptoms related to cervical myelopathy and had clear evidence of cord compression.

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**Table 3: Distribution as per clinical grading system**

| Grade | Description                                                                 | Preoperative | Postoperative |
|-------|-----------------------------------------------------------------------------|--------------|--------------|
| Grade 1 | Independent and normally functioning                                       | -            | 6            |
| Grade 2 | Walks on own but needs support/help to carry out routine household activities | 1            | 1            |
| Grade 3 | Walks with minimal support and requires help to carry out household activities | 4            | -            |
| Grade 4 | Walks with heavy support and unable to carry out household activities      | 2            | -            |
| Grade 5 | Unable to walk and dependent for all activities                            | -            | -            |
related to single or multiple levels herniated disk/osteophyte. The treatment protocol for such cord compression has been elaborately described and standardized in literature. The association of C2–3 fusion was an additional observation. The issue in question was to identify the significance of the presence of C2–3 fusion. We related the presence of C2–3 fusion to chronic or longstanding atlantoaxial instability that suggested the need for atlantoaxial stabilization. The analysis aims to identify if surgery on lower cervical spine can be avoided in such cases, despite the presence of stark evidences of cord compression.

Our recent publications focus on spinal instability as the primary nodal point of pathogenesis of degenerative or spondylotic spinal changes. The observations stress the need of only fixation as the treatment, without resorting to any kind of decompressive surgical procedure. We earlier reported the association of C2–3 fusion and C3–4 cord compression with atlantoaxial facetal malalignment that was indicative of atlantoaxial instability and suggested the need for atlantoaxial stabilization in addition to C3–4 stabilization. This was despite the fact that there was no evidence of direct compression, cord compromise, or signal alterations on imaging at the C1–2 level.

A spectrum of musculoskeletal and neural alterations has been grouped under the umbrella classification of basilar invagination. Skeletal alterations include assimilation of atlas, C2–3 fusion, os‑odontoideum, bifid arches of atlas, Klippel–Feil abnormality, and platybasia. Neural abnormality includes Chiari formation and syringomyelia. Short neck and torticollis are among the common external manifestation of basilar invagination. It is observed that basilar invagination is a consequence of chronic or longstanding atlantoaxial instability. Subaxial “cervical spondylotic changes” are frequently associated with basilar invagination, atlantoaxial instability, and shortening of neck or chronic neck spasm.

Shortening of the neck is affected more by reduction in the disk space height than by reduction in vertebral body height. Osteophyte formation and radiological evidence of single or multiple level cord compression are frequently identified in cases with basilar invagination, particularly in those patients who have short neck. Long-standing disk space reduction can be associated with bone fusions.

On the basis of our experience with cases having basilar invagination, we identified that all musculoskeletal and neural alterations are not primary abnormalities that are related to embryonic dysgenesis, but are secondary and “protective” natural alterations aimed to stall or delay clinical manifestations related to atlantoaxial instability. It is observed that various features that include basilar invagination, Chiari 1 formation, syringomyelia, bifid arches of atlas, assimilation of atlas, and bone fusions can be identified in isolation and still indicate the presence of atlantoaxial instability. The more important observation was that following atlantoaxial stabilization, there was a potential of reversal of all secondary musculoskeletal and neural alterations. The presenting clinical symptoms reversed as early as in the postoperative period. We had reported increase in the neck length and reversal or torticollis in the “immediate” postoperative phase following surgery that involved atlantoaxial fixation.

The standard and conventional protocol for diagnosing atlantoaxial instability has been on the basis of alterations of atlantodental interval on dynamic flexion–extension images and observation of evidences of cord compression or deformation opposite the odontoid process. Our alternative classification identified that high degree of clinical correlation, surgical experience of handling atlantoaxial facets, and presence of other local anomalies are mandatory to diagnose atlantoaxial instability in Types 2 and 3 atlantoaxial facetal instability. We labeled Types 2 and 3 atlantoaxial facetal instability as “central” or “axial” atlantoaxial instability. This was because in both these situations, atlantoaxial interval may not be altered and direct neural compression by the odontoid process is not the hallmark. Types 2 and 3 atlantoaxial instability is generally associated with longstanding or chronic atlantoaxial dislocation. Central or axial atlantoaxial dislocation has been identified with several commonly treated conditions that include basilar invagination, Chiari formation, “idiopathic” syringomyelia, multilevel cervical spondylotic myelopathy, myelopathy related to ossified posterior longitudinal ligament, and Hirayama disease.

On the basis of understanding that C2–3 fusion is an indication of atlantoaxial instability and that “degenerative”...
changes in the lower cervical spine were secondary events, all patients were treated by atlantoaxial fixation. “Remarkable” clinical improvement was observed in the “immediate” postoperative period and in the follow-up assessments. Despite the fact that the patients improved in their clinical symptoms, no significant reversal of “degenerative” spinal bone, disk, osteophyte, or neural alteration could be seen during the relatively short follow-up period.

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Conflicts of interest
There are no conflicts of interest.

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