

Chronic hypertrophic nonunion of the Type II odontoid fracture causing cervical myelopathy: Case report and review of literature

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Abstract

Background: Complications of nonunited Type II odontoid fractures can range from neck pain to progressive neurological deficit from cervical myelopathy. Rarely, the hypertrophic nonunion requires both anterior transoral decompression and posterior decompression with instrumented fusion. We present a case and review literature around this entity.

Case Description: A 68-year-old female presented with rapidly progressive cervical myelopathy (from normal to moderate myelopathy modified Japanese Orthopedic Association [mJOA] 13) over 3 months. Her history was positive for a Type II odontoid fracture managed conservatively and lost to follow-up for 25 years. Spinal imaging studies revealed hypertrophic nonunion and craniocervical kyphotic deformity with significant subaxial stenosis and segmental kyphosis. The patient underwent anterior transoral decompression, followed by posterior occipit thoracic decompression and instrumented fusion. At follow-up, the cervical myelopathy has improved to near normalcy (mJOA 17) with no evidence of implant-related complication.

Conclusion: Rarely, nonunion of Type II odontoid fractures may be hypertrophic where both instability and compression cause neurological morbidity. Such cases require anterior transoral decompression, posterior cervical decompression, and instrumented fusions.

Key Words: Hypertrophic pseudoarthrosis, myelopathy, nonunion, odontoid fracture, transoral surgery

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INTRODUCTION

Odontoid fractures are among the most common injuries at the craniocervical junction and account for nearly one in six cervical spine fractures. Various algorithms have been proposed for the management of Type II odontoid fractures, with nonsurgical management including cervical orthosis and halo-vest and surgical management including both anterior and posterior options.^[1] The risk of nonunion is highly variable, likely reflecting the variable population demographics in the various studies. Predictive risk factors of nonunion include significant

fracture displacement or angulation, delayed treatment, and advanced age.^[10]

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The hazards of nonunited odontoid fractures in adults can include mechanical neck pain^[2] as well as progressive craniocervical deformity^[5,8] and neurological deterioration into cervical myelopathy.^[4-9,11,12] Minor delayed trauma may also lead to substantial clinical progression.^[3] The appropriate management once a pseudoarthrosis is identified, is undefined. When surgery is indicated, the role for anterior and posterior approaches further remains controversial. This case illustrates a complex craniocervical deformity leading to progressive cervical myelopathy occurring for 25 years following a nonunited odontoid fracture and in combination with substantial subaxial disease. This required management was by a combined anterior/posterior approach and provided the patient with excellent neurological and structural outcome.

CASE REPORT

We present the case of a 68-year-old female with a past medical history significant for a Type II odontoid fracture sustained 25 years previously. This presented originally with neck pain and was detected on spinal radiographs, with conservative management in a soft cervical orthosis implemented. There was no further clinical or radiological follow-up for her condition.

She presented with 6 months of progressive cervical myelopathy that manifests as upper extremity weakness and hand incoordination, extremity paresthesiae, and gait instability. Her modified Japanese Orthopedic Association score (mJOA) was 13. On physical examination, she exhibited increased tone and diffused hyperreflexia, unsteady broad-based and hesitant gait, and bilateral Hoffman and Babinski signs. Computed tomography (CT) [Figure 1], demonstrates a chronic odontoid pseudoarthrosis with anterior subluxation, a significant posterior osteophyte narrowing the spinal canal (arrow), and segmental kyphosis.

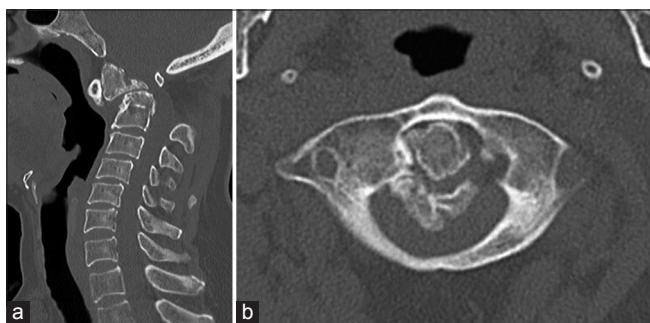


Figure 1: A 68-year-old female with remote odontoid fracture presents 25 years later with acutely progressive myelopathy. Computed tomography (sagittal, a) demonstrates a chronic odontoid pseudoarthrosis with anterior subluxation, significant osteophyte (arrow), and segmental kyphosis. Axial section (b) demonstrates the severity of canal compromise arising from the combination of deformity and heterotopic osseous formation

and significant segmental kyphosis overlying the pseudoarthrosis. Magnetic resonance imaging [Figure 2] reveals spinal cord compression ventrally with tension over the posterior osteophyte and the remainder of the C2 body. Further, significant subaxial spinal spondylotic disease is evident.

At surgery, following awake preoperative halo traction revealing the deformity to be mobile, permitting kyphosis reduction [Figure 3], the patient was in the supine position without further attempt to reduce the ventral translation. A two-stage procedure was planned including first a transoral decompression of the odontoid fragment with resection of posterior vertebral body osteophyte to achieve anterior spinal cord decompression. Second, C1 laminectomy was performed along with subaxial decompression to address the remaining spondylotic disease performed with an occipitothoracic fusion.

The patient was discharged home in a halo orthosis. At 4 months follow-up, she neurologically improved to functional independence (mJOA 17), with no evidence of pseudoarthrosis or implant failure on CT scan [Figure 4]. She was weaned from the halo and remains neurologically and structurally stable at 6 months postoperatively.

DISCUSSION

The mechanisms that underlie nonunion of the odontoid are likely multifactorial. Kirankumar *et al.*^[9] have described an algorithm with which to approach patients with odontoid nonunion, with the key decision point being the reducibility of the deformity either on physiological loading or under conditions of halo traction. For reducible atlantoaxial complexes, this algorithm advocates for realignment with either positioning or



Figure 2: Magnetic resonance imaging (sagittal) showed ventral spinal cord compression (arrow) by the posterior osteophyte and the remainder of the C2 body as well as subaxial spinal spondylotic disease

traction followed by posterior surgical stabilization of the segmental instability. For nonreducible atlantoaxial complexes, this algorithm advocates for simultaneous anterior transoral decompression and posterior surgical stabilization. While such an approach is reasonable for the simple atrophic nonunion, it fails in the situation when the pseudoarthrosis develops compressive features.

While nonunion of Type II odontoid fractures is common and the various surgical techniques to address this are within the armamentarium of the complex spinal surgeon, there is further substantial complexity when the nonunion is accompanied by hypertrophy of surrounding structures. The sparse literature surrounding the management of such hypertrophic deformity is summarized in Table 1 with the reports spanning 22 years and including four patients of diverse ages and with variable delays between trauma and neurological deterioration.^[5,8] The challenge when applying any algorithm such as that advocated by Kirankumar *et al.*^[9] to the patient with hypertrophic disease is notably that the neurological safety of fully reducing the deformity must be considered. Consequently, a first-stage transoral decompression is selected after which second-stage posterior surgical

intervention can be implemented. A preoperative trial of low-weight awake halo traction effectively reduced the kyphosis in our patient and provided for easier intraoperative access to the pseudoarthrosis during the transoral decompression. All surgical patients in this review were similarly managed with combined techniques of anterior transoral decompression, posterior cervical decompression, and instrumented fusion that provided for postoperative myelopathy improvement.

CONCLUSION

This case highlights the structural and neurological hazard of the nonunited odontoid fracture in young patients. This patient developed a complex craniocervical deformity with fragment subluxation, kyphosis, and osteophyte formation, with a long-term neurological consequence of myelopathy. Hypertrophic nonunion of Type II odontoid fractures occur rarely as a cause of progressive cervical myelopathy and requires different management inclusive of spinal cord decompression than that advocated for more straightforward atrophic nonunion in which realignment and stabilization may



Figure 3: Preoperative awake halo traction provided for improved cervical spine alignment by reducing the segmental kyphosis



Figure 4: Postoperative X-ray (a) reveals excellent cervical spine alignment following occipitothoracic fusion. Median sagittal computed tomography (b) reveals complete decompression of the odontoid fragment, posterior osteophyte, and residual body

Table 1: Hypertrophic nonunion of the odontoid causing cervical myelopathy

Author, year	n	Clinical details	Delay from fracture	Management
Crockard (1993)	2	30 female 55 male	4 years 5 years	Anterior transoral decompression and posterior atlantoaxial (C1-C2) instrumented fusion Outcome: Improved anterior transoral decompression and posterior atlantoaxial (C1-C2) instrumented fusion Outcome: Improved
Ho (2010)	1	87 male previous head injury, progressive lower extremity weakness	2 years	Nonoperative Outcome: No change
Current (2015)	1	68 female history of MVA, progressive upper extremity loss of coordination and sphincter dysfunction	25 years	Anterior transoral decompression and posterior occipitothoracic (O-T1) decompression and instrumented fusion Outcome: mJOA 13 → 17

mJOA: Modified Japanese Orthopedic Association, MVA: Motor vehicle accident

be the primary surgical objectives. Close follow-up after either initial conservative or surgical management remains important to protect these patients from such delayed complication.

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Conflicts of interest

There are no conflicts of interest.

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