Atlantoaxial Instability and Retroodontoid Mass

—Two Case Reports—

Atul GOEL, Umesh PHALKE, Francesco CACCIOLA, and Dattatraya MUZUMDAR

*Department of Neurosurgery, King Edward VII Memorial Hospital and Seth G.S.
Medical College, Parel, Mumbai, India*

Abstract

Two relatively elderly male patients (61 and 78 years) suffered moderately severe trauma to the head 1 and 3 years, respectively, prior to presentation with progressive quadriparesis and neck pain. Investigations revealed retroodontoid ligamentous hypertrophy and subtle mobile atlantoaxial dislocation. Following atlantoaxial fixation, both patients showed remarkable and sustained neurological improvement. These cases provide further evidence that retroodontoid ligamentous hypertrophic mass lesion could be secondary to instability of the atlantoaxial region.

Key words: atlantoaxial dislocation, odontoid process, posterior longitudinal ligament

Introduction

Neural compressive mass lesion posterior to the odontoid process has been variously identified as pseudotumor,²⁴⁾ ligamentous hypertrophy, damaged posterior longitudinal ligament,⁴⁾ synovial cyst,^{6,17,20)} cervical disc herniation,²³⁾ hypertrophic cicatrix,⁴⁾ and elastofibroma.²²⁾ Such lesions are not associated with any generalized arthropathy and have similar clinical, radiological, and histological features. Most patients are treated surgically for these rare lesions under the presumed diagnosis of tumor.^{6,10,14,17,18,22)} We report two cases of retroodontoid mass associated with subtle mobile atlantoaxial dislocation.

Case Reports

Case 1: A 61-year-old male presented with complaints of progressive quadriparesis and neck pain persisting for about 1 year. He had difficulty in performing manipulations with the hands and needed support to walk. He reported a fall from a height of about 6 meters resulting in hyperextension of the neck 1 year before. Except for severe neck pain that lasted for about 1 week, there were no other significant post-trauma sequelae. He had no history of any systemic disease. There was no evidence of rheumatoid arthritis or any other joint-related generalized disease. Neurological examina-

tion found spastic grade 3–4 quadriparesis, but no gross sensory abnormality. Magnetic resonance (MR) imaging showed a retroodontoid process midline lesion indenting into the cervicomedullary cord, appearing as isointense on the T₁-weighted images and hypointense on the T₂-weighted images (Fig. 1). Dynamic radiography revealed subtle mobile and incompletely reducible atlantoaxial dislocation (Fig. 2A, B). The patient was treated with lateral mass atlantoaxial plate and screw fixation as described by us previously.⁵⁾ Satisfactory but incomplete reduction was achieved (Fig. 2C). The patient showed remarkable neurological and symptomatic recovery. The improvement was sustained and at the follow-up examination 14 months after surgery, the patient was almost asymptomatic.

Case 2: A 78-year-old male presented with progressive spastic quadriparesis evolving over the past 6 months. He had been unable to sit or stand without support for about 1 month. He was involved in a car accident 3 years before, in which he suffered moderate head trauma. Clinical and radiological findings were similar to those in Case 1 (Figs. 3 and 4). He was also treated by lateral mass plate and screw fixation. At follow-up examination 2 years after surgery, the patient was completely independent and had participated in a marathon walk.

Discussion

Retroodontoid pannus formation is a common

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Fig. 1 Case 1. A: Sagittal T₁-weighted magnetic resonance (MR) image showing a retroodontoid isointense mass compressing the neural structures. B: Sagittal T₂-weighted MR image showing the hypointense retroodontoid mass. C: Axial T₂-weighted MR image showing the retroodontoid mass.

Fig. 2 Case 1. A: Lateral radiograph of the craniovertebral junction with the neck in the flexion position showing mild atlantoaxial dislocation. B: Lateral radiograph with the head in the extension position showing reduction of the dislocation. C: Postoperative radiograph with the neck in the flexion position showing plate and screw fixation.

Fig. 3 Case 2. A: Sagittal T₁-weighted magnetic resonance (MR) image showing a retroodontoid mass as isointense and hypointense areas compressing the neural structures. B: Sagittal T₂-weighted MR image showing the hypointense retroodontoid mass. C: Axial T₂-weighted MR image showing the mixed intensity retroodontoid mass.

Fig. 4 Case 2. A: Lateral radiograph of the craniovertebral junction with the neck in the flexion position showing atlantoaxial dislocation. B: Lateral radiograph with the head in the extension position showing partial reduction of the dislocation. C: Postoperative radiograph with the neck in the flexion position showing plate and screw fixation.

pathological event in rheumatoid and psoriatic arthritis.^{7,19,26}) Pseudotumors have also been identified in patients with pigmented villonodular synovitis,¹³⁾ spontaneous cervical epidural hematoma,^{2,9)} gouty deposits,¹⁵⁾ and in obese patients and patients on steroid therapy with fat deposition.^{1,8)} Retroodontoid pseudotumor was found in a 74-year-old patient with diffuse idiopathic skeletal hyperostosis¹²⁾ and five cases of retroodontoid mass were found in patients with Forestier's disease.²¹⁾

Retroodontoid process ligamentous mass has been identified in several patients without related systemic illness or generalized joint disease.^{6,10,14,17,23,25)} Most patients were elderly males.^{4,10,11,13,18,23,24)} Some patients had suffered moderately severe head or neck injury.²⁴⁾ The clinical and imaging characteristics in our patients were almost identical to each other and to the reported cases. Therefore, retroodontoid process ligamentous hypertrophy in the elderly is probably a defined and discrete clinical condition. Long-term trauma as a result of atlantoaxial subluxation is suspected to be the underlying cause of the formation of progressively increasing fibrous granulation tissue, with hypertrophy of connective tissue elements as the abnormal response to chronic mechanical stress and friction. The pathogenesis of the retroodontoid process ligamentous mass has not been clearly defined in patients with no atlantoaxial dislocation. The lesions in our cases and in reported cases were isointense on T₁-weighted MR imaging and predominantly hypointense on T₂-weighted MR imaging. These findings are suggestive of the fibrous or the cartilaginous nature of the lesion. Histological examination of the specimen in all reported cases confirmed the presence of degenerated ligaments.

Differentiation of these rare lesions from tumorous mass lesions is important.^{6,10,14,18,20,25)} The presence of the retroodontoid mass lesion and

associated severe anterior indentation of the neural structures in some cases has caused most authors to resort to transoral surgical decompression.^{3,4,13)} Some authors performed laminectomy to approach the anteriorly located extradural mass.^{17,18,23,24)} Patients without atlantoaxial dislocations but with non-reducible traumatic dislocations have also been treated with transoral decompression, posterior fusion, and postoperative immobilization.^{3,16)}

Some degree of atlantoaxial subluxation has been observed in previously reported cases.^{3,10,24,25)} Retroodontoid mass was associated with fracture of the odontoid process¹⁴⁾ and os odontoideum.¹¹⁾ Severe mobile and entirely reducible dislocation has not been reported. Both our patients had marginal but definite mobility of the atlantoaxial region. Why such retroodontoid ligamentous hypertrophy is not associated with all cases of congenital or post-traumatic atlantoaxial dislocation and is limited only to the elderly age group remains unclear. Posterior fixation resulted in immediate postoperative neurological improvement in our and previous patients,^{11,16)} suggesting that most symptoms were due to this instability rather than the retroodontoid mass. Clinical improvement was sustained during the follow-up period, which tends to exclude the possibility of a tumor and suggests that mechanical stress was an important pathogenetic factor in inducing the hypertrophic change and in proliferation of granulation tissue, and that the fixation halted progressive degeneration of the ligaments and also probably resulted in regression. Stainless steel material was used for fixation, and computer-based imaging was not possible in our patients, so whether the retroodontoid mass was reduced following posterior stabilization is unknown. However, radiographical disappearance of the retroodontoid mass has been demonstrated after posterior stabilization surgery.^{11,25)}

The present and previous cases suggest that retroodontoid process ligamentous hypertrophy may be a result of injury to the ligaments. The presence of subtle or manifest atlantoaxial dislocation could result in progression of the ligamentous degenerative mass over time. Stabilization of the atlantoaxial region probably halts the process of ligamentous hypertrophy.

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Address reprint requests to: A. Goel, M.D., Department of Neurosurgery, King Edward VII Memorial Hospital, Parel, Mumbai-400012, India.
e-mail: atulgoel62@hotmail.com