

SHORT REPORT

Fatal instability following "odontoid sparing" transoral decompression of a periodontoid pseudotumour

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Pseudotumour of the craniovertebral junction is an uncommon cause of high cervical myelopathy in the elderly. The anterior transoral approach is the preferred means of accessing these lesions. An "odontoid sparing" transoral approach, in which there is only minimal bone removal, is thought to preserve stability of the craniovertebral junction, obviating the need for posterior stabilisation. This report is of an 82 year old man who developed fatal atlanto-axial instability following an odontoid sparing transoral resection of a pseudotumour. This complication has not previously been described and its occurrence has important implications for the surgical management of this condition.

Anteriorly placed extradural mass lesions at the craniovertebral junction causing high cervical myelopathy are uncommon.¹ Neoplasms are the most frequently encountered masses, although non-neoplastic lesions such as rheumatoid pannus, calcium pyrophosphate dihydrate, gout, and hypertrophic non-union of odontoid fracture have also been described.²⁻⁵ Peri-odontoid pseudotumour is another non-neoplastic mass lesion, usually encountered in elderly patients with degenerative disease of the cervical spine.²

The transoral approach is the established means of accessing anteriorly placed midline extradural pathology and may need to be accompanied by some form of posterior fusion to prevent any subsequent craniovertebral instability.^{6,7} An odontoid sparing transoral approach for the removal of periodontoid pseudotumours has been described, which was not thought to destabilise the craniovertebral junction, thus obviating the need for postoperative immobilisation and subsequent posterior fixation.² We present a case of a periodontoid pseudotumour in which the patient developed fatal atlantoaxial instability following an odontoid sparing transoral resection without posterior fixation. We discuss the biomechanical reasons for this devastating complication together with the implications for the surgical management of this condition.

CASE REPORT

An 82 year old man was admitted to hospital with a six month history of a severe, progressive, high cervical myelopathy. Respiratory function was subjectively unaffected. There was no history of rheumatoid arthritis or trauma. The patient had a history of hypertension, which was controlled on drug treatment, but was otherwise fit and well, with no previous history of angina, myocardial infarction, or other major medical problems.

On examination he had a spastic quadriplegia, with increased tone and reflexes in all four limbs, positive Hoffmann's signs bilaterally, and extensor plantar responses. There was wasting of the small muscles of both hands. Dorsal column sensation was impaired in all four limbs.

Magnetic resonance imaging (MRI) (fig 1A) showed a smooth, midline, ventral, extradural soft tissue mass, the bulk of which extended posterior to the odontoid process causing cord compression and myelomalacia. Plain radiographs confirmed extensive subaxial spondylotic change but flexion and extension films showed no evidence of instability. Routine preoperative investigations including plasma urea and electrolytes, full blood count, and ECG were normal.

A standard odontoid sparing transoral approach to the mass was used (fig 1B).² This created a "window" through which the pseudotumour, occupying the retro-odontoid space, could be seen and removed by a combination of suction and rongeurs until dural pulsation was obtained. The typical amorphous, xanthomatous pseudotumour material could not be clearly defined as separate from the transverse ligament. This is important because in resecting the lesion—which was not exophytic but rather was within the transverse ligament—inadvertent damage to this stabilising structure may have occurred. The wound was then irrigated and closed in a standard fashion.⁶ There were no intraoperative complications and no orthosis was used.

Histopathological examination of the mass showed degenerate ligament and fibrocartilage, with an associated fibrinous exudate, but no inflammatory infiltrate. No calcium pyrophosphate dihydrate or uric acid crystals were seen. The appearances were consistent with a diagnosis of pseudotumour.

Postoperatively the patient reported an immediate improvement in his neurological symptoms. Postoperative flexion-extension films of the cervical spine were not performed because at the time the procedure was not regarded as potentially destabilising.² On the second postoperative day he was found collapsed, having suffered a cardiorespiratory arrest. Cardiopulmonary resuscitation restored a cardiac rhythm and blood pressure but there was no respiratory effort. He was ventilated and transferred to the intensive therapy unit. An EEG was consistent with extensive anoxic brain damage. The patient failed to improve and died on the fifth postoperative day.

At a coroner's necropsy examination there was no cord compression in the neutral position and both the odontoid, which remained attached to the body of C2, and the anterior arch of C1 remained intact. However, on digital examination of the foramen magnum with the head flexed the odontoid did compromise the spinal cord. This ligamentous atlanto-axial instability had resulted in severe contusion of the spinal cord at this level. The macroscopic appearances of the contusions were typical and further microscopic analysis was therefore not performed. There was no suggestion of any other possible cause of death; in particular, there was no evidence of a pulmonary embolism, deep vein thrombosis, stroke, or myocardial infarction.

DISCUSSION

Periodontoid pseudotumours were first described as a separate pathological entity in 1991 by Crookard *et al.*² They

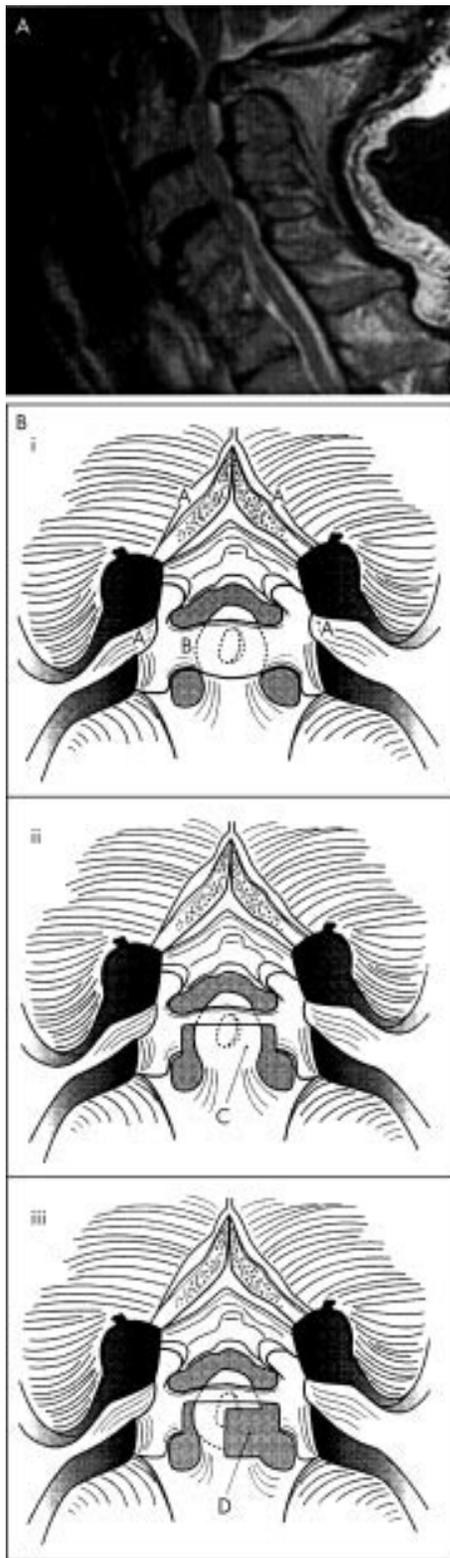


Figure 1 (A) Sagittal T₂ weighted magnetic resonance image in the mid-sagittal plane, showing the extradural, posteriorly convex retrodental pseudotumour which is causing marked mid-sagittal compression of the spinal cord. (B) Schematic diagram of the craniocervical junction showing the extent of bony resection in an odontoid sparing approach to the retro-odontoid space: (i) retracted pharyngeal mucosa (A) exposing the anterior arch of C1 (B); (ii) the inferior half of the anterior arch of C1 has been drilled away to expose the underlying odontoid process of C2 (C); (iii) one half of the odontoid process has been drilled away, preserving the apex with its associated ligamentous attachments, to expose the retro-odontoid space (D).

in our case may reflect this theory that pseudotumour formation occurs secondary to a damaged transverse ligament, which has an important role in maintaining atlantoaxial stability.⁷⁻¹⁰

The ligamentous structures in this region play a vital role in ensuring stability of the craniocervical junction, preventing subluxation and dislocation, yet preserving functional flexibility.¹⁰⁻¹³ The first line of defence in preventing atlanto-axial dislocation is the transverse ligament, which limits anterior gliding of the atlas in flexion.¹⁰ Although primarily preventing rotatory instability, the alar ligaments—connecting the occipital condyles to the dens—provide a second line of defence in preventing impingement of the odontoid process on the cord.¹¹⁻¹⁴⁻¹⁵ Additional stability is provided to a lesser degree through the tectorial membrane, apical ligament, joint capsules, anterior and posterior atlanto-occipital membranes, and the anterior longitudinal ligament.¹⁵

A standard transoral approach usually involves resection of the anterior arch of the atlas, the odontoid process with its associated apical and alar ligaments, together with the transverse ligament according to the pathology.² Following this type of procedure up to 70% of patients develop craniocervical instability.¹⁶ With an odontoid sparing approach the transverse ligament and tectorial membrane may be (partially) resected, although all the other stabilising structures remain intact, obviating the need for postoperative immobilisation and posterior fusion.² Even if the transverse ligament is divided, some biomechanical studies have shown that the alar ligaments, together with the other remaining ligamentous structures and C1/2 joint capsules, should provide sufficient stabilisation to prevent cord compromise.⁸ The alar ligaments are relatively strong, and in dynamic tests reported by Dvorak *et al*¹¹ can bear a median axial load of 250 N, approaching that of the transverse ligament, which tolerates a median load of 330 N. Furthermore, these investigators felt that the strength of the alar (but not the transverse) ligaments may have been underestimated.¹¹ What is controversial, however, is the degree of excursion permitted by the apical alar ligamentous complex before rupture, following division of the transverse ligament. Fielding *et al* suggest that the elasticity of the alar ligament may be sufficiently great to cause potential cord compromise.¹⁰ More recent histological and biomechanical studies, however, have demonstrated the inelastic nature of this ligament and concluded that the alar ligaments can act as a “second line of defence.”¹¹⁻¹⁷ In reported cases of isolated traumatic rupture of the transverse ligament, instability may develop,¹⁰⁻¹⁸ and some investigators strongly maintain that transverse ligament integrity is essential for craniocervical stability, although these conclusions are based on observations in conditions in which remaining bone or ligamentous stabilising structures may also be compromised.⁷⁻⁹

This case shows that instability can occur after resection of a pseudotumour using this odontoid sparing approach. The reason may be related to the underlying pathological basis of pseudotumour formation. Fielding *et al* found considerable age independent variability in the strength of the normal transverse ligament when subjected to flexion loading.¹⁰

described a mass consisting of amorphous yellowish material, histological examination of which showed degenerate ligament, fibrocartilage, and fibrin but no inflammatory cell infiltrate. It was proposed that these periodontoid pseudotumours arose following a partial tear in the transverse or posterior longitudinal ligaments, with subsequent aberrant attempts at repair resulting in a vicious cycle leading to a progressively enlarging mass. The development of postoperative instability

Twenty per cent of the ligaments in their series were unaccountably weak, rupturing after a force only 25% of that required for the remainder. It may be that it is this group of patients with inherently weak ligaments who are predisposed to pseudotumour development, owing to an increased propensity to ligamentous injury following relatively minor trauma. It is also possible that secondary stabilising ligaments are similarly affected and are therefore unable to provide sufficient structural stability following surgical division of the transverse ligament.

An alternative explanation for the development of instability is that a long forgotten whiplash injury may have caused a partial transverse ligament tear, which after many years of attempted healing results in pseudotumour formation. The mechanism of whiplash injury, particularly if occurring with the head in maximum rotation, could also result in stretching or rupture of the alar ligaments,¹⁴⁻¹⁷ which may not then be able to provide adequate secondary stability following pseudotumour removal.

We believe that this case shows that pseudotumours arise from weakened ligaments, and the occurrence of postoperative instability was to some extent an expression of the natural progression of this degenerating process. The surgical implications are that even following odontoid sparing resection, stability in these cases cannot be presumed, although the risk of instability is substantially reduced. Patients should be immobilised in an orthosis postoperatively and undergo early and delayed flexion/extension plain radiographs of the cervical spine, followed by posterior fusion if instability is demonstrated.

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