Occipitocervical fusion for reduction of traumatic periodontoid hypertrophic cicatrix

Case report

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Periodontoid hypertrophic cicatrix resulting from trauma, as demonstrated by magnetic resonance (MR) imaging, is essentially the same as that seen in rheumatoid arthritis. Recent reports suggest that, in rheumatoid arthritis, occipitocervical fusion without transoral decompression of the pannus is adequate for resolution of this anterior lesion. A case of traumatic periodontoid cicatrix is presented in which posterior fusion resulted in reduction of the anterior mass lesion, clearly demonstrated by MR imaging. The etiology of periodontoid hypertrophic scarring, both traumatic and rheumatoid, is discussed in light of MR findings, and treatment implications are considered.

KEY WORDS - periodontoid cicatrix □9 atlantoaxial dislocation □9 rheumatoid arthritis □9 odontoid fracture

Non-union of odontoid fractures with resultant chronic instability and myelopathy is not uncommon,1,5,11,12,16 particularly in patients older than 40 years of age and with odontoid displacement of more than 4 mm. Anderson-D’Alonzo Type II odontoid fractures (occurring at the base of the odontoid)6 have the highest rate of non-union among fractures at this level;2 Hadley, et al.,5 reported a 26% rate of failure of fusion in these cases, and a 67% failure rate in those with more than 6 mm displacement. This failure of healing in Type II fractures may be due to the disruption of the vascular arcade to the axis and odontoid apex.2 Most authors recommend early operative intervention in such cases, consisting of occipitocervical or C1–2 fusion with wire, bone graft, and/or acrylic cement.1,2,5,11,12,16 Other authors7 have suggested the use of transoral decompression of the odontoid and ligamentous structures, combined with posterior stabilization to remove the neural compressive effects of anterior elements.7,12 Lee and Fairholm7 advocated this combined approach in a case in which they observed exuberant scar tissue between the atlas and the dens. This hypertrophic cicatrix can be confirmed dramatically by magnetic resonance (MR) imaging.18 It is reminiscent of periodontoid pannus found in the atlantoaxial dislocation of rheumatoid arthritis (Fig. 1), which has been managed with a similar operative approach by Crockard, et al.4 The decision to perform fusion alone versus transoral decompression and posterior fusion hinges on the concept that compressive myelopathy is due to mechanical irritation rather than the proliferation of periodontoid soft tissue.

We present a case of a previously undiagnosed Type II odontoid fracture with periodontoid cicatrix compressing the neuraxis, which was treated with a combined transoral decompression and fusion. The transoral decompression only partially removed the mass, but immobilization and a stable fusion resulted in further spontaneous marked reduction of this scar tissue, demonstrated by subsequent MR evaluations.

Case Report

This 56-year-old man presented with a history of progressive gait deterioration. He had undergone a lumbar laminectomy for stenosis and a herniated L5–S1 intervertebral disc 2 years prior to this presentation. At that time he experienced lower-extremity numbness and weakness of a claudicatory nature bilaterally. Paraphysically, the patient described a blow to the vertex which he received 16 years prior to his current presentation. He acknowledged experiencing Lhermitte’s sign at the time of this trauma, and intermittently since that time with Valsalva maneuvers and with sneezing.
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Cervical plain x-ray films revealed an area of bony density around, behind, and superior to the dens. Flexion and extension x-ray films revealed no significant C1-2 dislocation. Computerized tomography (CT) showed diffuse thickening of soft-tissue density and hypertrophy in the area of the dens and the posterior aspect of the C-2 vertebral body. Magnetic resonance imaging showed an epidural mass with soft-tissue intensity at C-2, involving the base of the odontoid, with clear separation of the dens from the body of C-2 and encasement of the dens and anterior arch of C-1. Compromise of the anteroposterior diameter of the spinal canal allowed an 8-mm residual canal space (Fig. 2). Despite a lack of significant dislocation, the appearance of the mass was strikingly similar to that seen in rheumatoid disease (Fig. 1). The MR image showed a marked deformity of the cervicomedullary junction by the periodontoid mass (Fig. 2 left).

Operation. With the patient under somatosensory evoked potential monitoring, a halo apparatus and vest were placed; anesthesia was then induced. After elevation of the soft palate and exposure of the posterior pharynx, a vertical incision was made and the vertebral body of C-2, the anterior arch of C-1, and the dens were visualized. A copious amount of fibrous soft tissue protruded above and below the anterior arch of C-1. With the arch removed, the odontoid process was seen to be fragmented and the base of the odontoid irregular, with fibrous scar tissue encasing the remainder of the dens, which was moveable and significantly separated from the body of C-2. Some of the medial superior aspect of the C-2 vertebra was then drilled away, and a dramatic amount of scar was found impinging on the dura. Fibrotic soft tissue was removed piecemeal with pituitary and Kerrison rongeurs. Pathological analysis confirmed this to be scar tissue. Ultimately, the dura was visualized superior to the posterior portion of the

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FIG. 1. Sagittal magnetic resonance (MR) image (TE 20 msec, TR 500 msec) showing periodontoid pannus with intermediate signal intensity secondary to inflammatory and mechanical instability of rheumatoid arthritis.

Fig. 2. Left: Sagittal T1-weighted magnetic resonance (MR) image showing traumatic periodontoid hypertrophic cicatrix with intermediate signal intensity. Note the similarity to the rheumatoid arthritis pannus shown in Fig. 1. Right: Axial MR image (TE 20 msec, TR 1000 msec) demonstrating compression of the neuraxis by traumatic scar tissue at C-2.
dens and inferior to the inferior portion of the C-2 body; however, adherence of the fibrous tissue to the dura allowed only partial removal.

After this procedure was completed, the patient was placed in the prone position and the suboccipital cranium, posterior arch of C-1, and lamina of C-2 were all exposed and decorticated. These elements were normal in appearance, and there was no overt evidence of subluxation. No wiring or prosthesis of the posterior elements was performed, but the entire area was packed with cancellous bone from an iliac crest graft to effect a long-term fusion. The patient tolerated the procedure well and there was no immediate postoperative neurological change.

Postoperative Course. An MR image 2 weeks postoperatively (Fig. 3) showed modest reduction of the periodontoid cicatrix, but there was significant residual mass. Over the next 3 months, the patient improved progressively in his gait, upper-extremity strength, and manual dexterity. The halo apparatus was removed 10 weeks postoperatively, after lateral cervical flexion and extension x-ray films showed good healing of the graft without evidence of instability. An MR image obtained 9 months postoperatively (Fig. 4) showed significant further reduction of the lesion, with dramatic spontaneous decompression and enlargement of the cervicomedullary neuraxis signal.

Discussion

Periodontoid pannus can cause compression of the cervicomedullary junction, with progressive myelopathy and sudden death. Sherk has stated that 15% to 36% of patients with rheumatoid arthritis experience atlantoaxial instability. In rheumatoid arthritis, this results from synovial inflammatory proliferation and osteoporosis, with destruction of the dens and increased excursion of the atlantoaxial joints. Mikulowski, et al., performed pathological examination of eight atlantoaxial joints in a postmortem study of 11 individuals with rheumatoid arthritis and cord compression; they found periodontoid soft-tissue proliferation in all of them. Magnetic resonance imaging has confirmed the presence of this pannus and its compressive effect on the neuraxis.

Attempts at treatment of the compressive myelopathy of rheumatoid arthritis have been aimed at both

![Fig. 3. Sagittal magnetic resonance images (TE 20 msec, TR 500 msec) obtained 2 weeks after transoral surgery and posterior fusion. Partial decompression of the periodontoid cicatrix and resultant partial widening of the spinal cord are shown.](image)

![Fig. 4. Magnetic resonance (MR) images obtained 9 months postoperatively. Left: Sagittal MR image (TE 20 msec, TR 500 msec) showing resolution of the periodontoid cicatrix, with expansion and normal configuration of the spinal cord. Right: Axial MR image (TE 20 msec, TR 1000 msec) of the C-2 area showing resolution of the periodontoid hypertrophic cicatrix and decompression of the spinal cord, with copious perimedullary subarachnoid space.](image)
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causes of the compression, namely transoral removal of the soft-tissue pannus and posterior stabilization of the dislocation by fusion. Some authors\textsuperscript{4,17} suggest that both posterior fusion and transoral decompression are the best treatment for this lesion. However, fusion fails to address the anterior compressive lesion, and transoral pannus removal has a significant complication rate,\textsuperscript{3,17} including bacterial infection, cerebrospinal fluid leak, and the frequent necessity for tracheostomy. It also fails to deal with the instability of the dislocation.

A recent report by Zygmunt, \textit{et al.}\textsuperscript{20} presented nine patients with rheumatoid arthritis, whose periodontoid pannus was treated by posterior fusion and immobilization only; subsequent spontaneous reduction of the pannus and cord expansion was demonstrated on MR images. This supports the theory that the soft-tissue mass is more the result of reactive fibrosis due to the mechanical stress of the subluxation than an active inflammatory proliferative process. The study by Mikulowski, \textit{et al.}\textsuperscript{10} substantiates this. In their study only two of the eight cases of rheumatoid arthritis with pannus showed active inflammation with rheumatoid granulomas; of the other six cases, four showed only fibrosis and two showed fibrosis with minimal nonspecific inflammation.\textsuperscript{5}

Petterson, \textit{et al.}\textsuperscript{13} analyzed the cervical spine MR images of 23 rheumatoid arthritis patients and found that 14 had periodontoid pannus, all with horizontal atlantoaxial subluxation. They considered that, while the T\textsubscript{2}-weighted image signal intensity was high enough for an edematous inflammatory component, it was too low to be a purely inflammatory process. They found that the largest soft-tissue masses were in patients with the greatest horizontal subluxation, supporting the concept that this is largely fibrous tissue reactive to chronic mechanical irritation. The fact that mechanical factors outweigh inflammation in these compressive lesions is also supported by a report by Thompson and Meyer\textsuperscript{19} on rheumatoid patients with subluxation, in whom C1–2 posterior fusion and halo immobilization successfully arrested myelopathy without transoral decompression.

It appears from the above that the pathogenesis of compressive periodontoid pannus in rheumatoid arthritis is similar to hypertrophic cicatrix found in atlantoaxial dislocation secondary to traumatic odontoid fracture. Sze, \textit{et al.}\textsuperscript{18} presented three patients diagnosed by MR imaging as having “pseudotumor” of the craniovertebral junction, all of whom had either degenerative spinal disease (unrelated to rheumatoid arthritis) or a history of trauma, resulting in atlantoaxial subluxation. The MR images in these cases bore a striking similarity to those in our patient and to those of the rheumatoid arthritis cases presented by Zygmunt, \textit{et al.}\textsuperscript{20} Sze, \textit{et al.}, suggested that it is the mechanical dysfunction and instability of the atlantoaxial joint in these subluxations which promotes the hypertrophic scar tissue in the periodontoid area. Lee and Fairholm\textsuperscript{1} presented three cases of nonreducible traumatic dislocation treated with transoral decompression, posterior fusion, and postoperative immobilization. Their second case demonstrated exuberant fibrous tissue between the atlas and the odontoid. They advocated the combined approach for its ability to remove the anterior mass while allowing reduction and fusion in slight extension, and without force. However, the cases of Zygmunt, \textit{et al.}, demonstrate the propensity of pannus to resolve spontaneously after posterior fusion alone in rheumatoid arthritis. Our case, managed in a similar manner to Lee and Fairholm’s cases, demonstrates that, despite only partial operative transoral decompression as seen on the early postoperative MR image, delayed spontaneous resolution of the mass is also possible in traumatic fibrosis when immobilization of the dislocation has been attained by posterior fusion. Our patient did not demonstrate overt instability on preoperative evaluation; however, his history of trauma and immediate subsequent Lhermitte’s sign, the presence of myelopathy on examination, the preoperative MR image showing a fragmented dens with periodontoid soft tissue, and the operative findings of a mobile, fractured odontoid and pathologically confirmed cicatrix, all are compatible with a traumatic etiology for this problem. We feel that the lack of overt preoperative x-ray instability is explained by the development of the large periodontoid cicatrix, preventing movement of the C1–2 junction. This case was not managed by posterior fusion alone; but the early postoperative MR image demonstrates that only a very limited portion of the cicatrix was removed by the anterior approach. The late postoperative MR image clearly indicates that the mechanical immobilization attained by posterior fusion, rather than the anterior decompression, was the cause of the disappearance of the scar. We look forward to further reports of resolution of posttraumatic cicatrix in patients managed by posterior fusion alone.

\textbf{Conclusions}

Magnetic resonance imaging graphically and dramatically demonstrates the periodontoid hypertrophic fibrous tissue associated with rheumatoid arthritis and traumatic atlantoaxial instability. We consider that the etiology of this material is the same in both conditions and results primarily from mechanical rather than inflammatory processes. The postoperative serial MR imaging in our case demonstrated that posterior stabilization more than transoral decompression resulted in the spontaneous regression of the periodontoid cicatrix. This is in agreement with recent studies\textsuperscript{20} which suggest the spontaneous regression of periodontoid pannus in rheumatoid arthritis with treatment by posterior stabilization alone. We would encourage further MR evaluation of odontoid fractures to assist in the operative management of these lesions and would assert that MR imaging, rather than CT or myelography, is most useful in identifying the very characteristic appearance of this lesion.
References


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