



Case Report

Periodontoid pseudotumoral lesions

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ABSTRACT

Background: Periodontoid pseudotumoral lesions (PPL) are an uncommon cause of cervical pain and myelopathy. In addition, they may be associated with atlantoaxial instability (AAI).

Case Description: Two patients over 60 years of age presented with neck pain alone. Their MR scans showed expansive lesions involving the odontoid process. One patient with AAI required an occipitocervical arthrodesis, while the other patient without instability was managed with an external orthosis (Philadelphia collar). Both of them experienced full resolution of pain and remained neurologically intact an average 36 months later (range 24–48).

Conclusion: Here, we discussed the clinical, MR, and non-surgical (without AAI) versus surgical management (with AAI) for different types of PPL.

Keywords: Atlanto-axial joint, Magnetic resonance imaging, Odontoid process, Skull base, Spinal cord diseases

INTRODUCTION

In this study, we present the clinical, MR appearance, non-surgical, and surgical management for two cases of periodontoid pseudotumoral lesions (PPL), and selectively reviewed the appropriate literature.

CASE DESCRIPTION

Case 1: Non-surgical management of PPL

A 72-year-old male presented with neck pain alone, no neurological deficit, and a cervical MR that showed an expansive lesion posterior to the odontoid process [Figure 1]. As dynamic radiographs did not demonstrate atlantoaxial instability (AAI), conservative treatment (i.e. Philadelphia-type cervical collar) was utilized. Four years later, the patient's neck pain had improved, and he remained neurologically intact [Table 1].

Case 2: Surgical management of PPL

A 61-year-old female developed the rapid onset cervical pain exacerbated with cervical rotation but no neurological deficit.

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Here, the cervical MRI revealed an intense inflammatory reaction involving the atlantoaxial joint (i.e. hyperintense T2

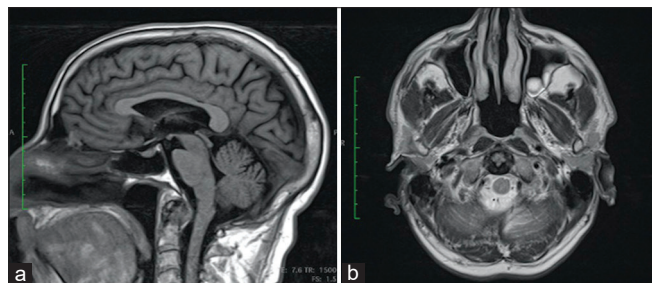


Figure 1: Sagittal (a) and axial (b) sections of magnetic resonance images of the craniocervical junction, presenting an expansive lesion in the posterior aspect of the odontoid process, with low signal at both T1 (a) and T2 (b). There is no evidence of spinal cord compression.

signal in anomalous tissue around the axis dens, with widening of the atlantodental interval) [Figure 2]. Further, the C-reactive protein and erythrocyte sedimentation rates were increased. A diagnosis of AA joint synovitis led to the administration of 60 mg of prednisone daily for 3 weeks. When subsequent dynamic cervical X-rays showed AAI, she underwent an occipitocervical arthrodesis with biopsy of the odontoid process (which later showed crystal deposition). Within a month, her cervical symptoms improved, and at 2 postoperative years, she had neither pain nor any neurological deficits [Table 1].

DISCUSSION

Retro-odontoid pseudotumor (ROP)

ROP tumor is most often attributed to rheumatoid arthritis,^[11] cervical spondyloarthropathy,^[5] primary amyloidosis, and

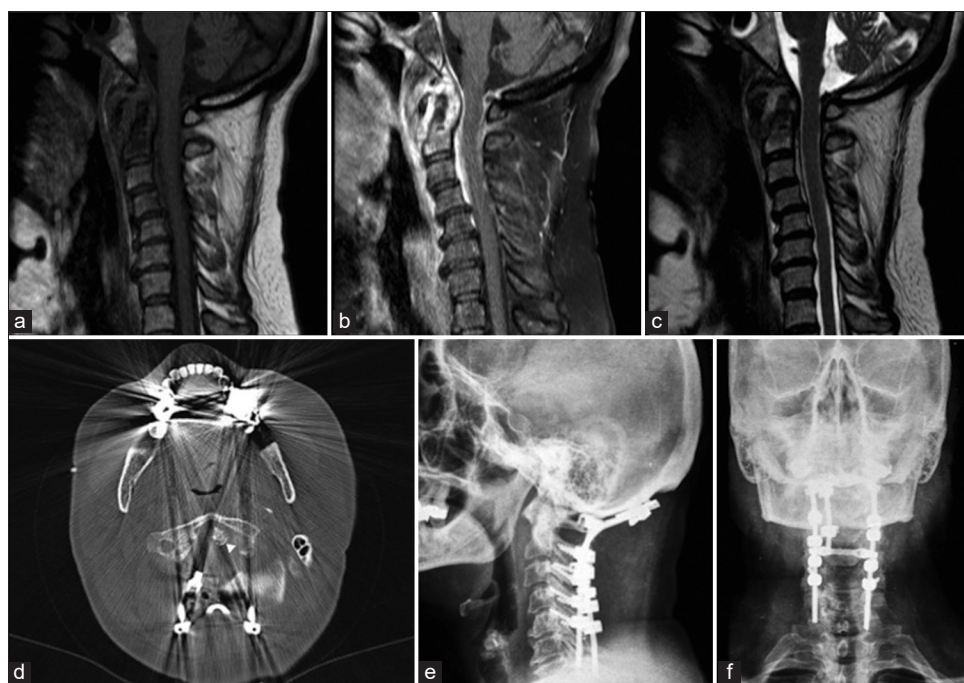


Figure 2: Sagittal sections from preoperative magnetic resonance imaging (a-c) indicating tissue formations that suggest an expansive process at the atlantodental joint, with low signal at T1 (a), intense enhancement after contrast injection (b) and heterogeneous signal at T2 (c). In (d), it is presented an axial section of a computed tomography image of the cervical spine obtained at the immediate postoperative period of occipitocervical arthrodesis; the arrow points to the calcification halo around the axon odontoid process. In (e) and (f), it is presented the final result of the occipitocervical arthrodesis.

Table 1: Summarization of the presented cases.

Case	Age (years), gender	Clinical presentation	Rheumatoid arthritis	Presence of AAI	Treatment	Outcome
1	72, M	Chronic cervical pain No myelopathy	No	No	Philadelphia collar	Good
2	61, F	Intense cervical pain No myelopathy	No	Yes	Occipitocervical arthrodesis and biopsy	Good

AAI: Atlantoaxial instability

Table 2: Review of PPL casuistic in the literature.

Author (year)	Sample characteristics	AAI	Associated conditions	Treatment	Follow-up duration in months	Outcome
Barbagallo <i>et al.</i> (2013) ^[11]	– 5 patients – MA 64.8 yrs. (55–76) – M:F=3:2	None	Subaxial spondylosis (100%)	– C1-C2 fixation (2) – Occipitocervical fixation (3)	32 (22–45)*	– 80% had neurological improvement – Reduction or disappearance of retro-odontoid lesion in all the patients followed
Chikuda <i>et al.</i> (2009) ^[3]	– 10 patients – MA 71 yrs. (58–82) – M:F=3:2	Present in 2 patients	– Reduced ROM C2-C7 – OALL (60%)	– C1 laminectomy + occipitocervical fusion (5) – C1 and C2 laminectomy + occipitocervical fusion (1) – C1 laminectomy + C3-C7 laminoplasty + occipitocervical fusion (1) – C3-C7 laminoplasty + occipitocervical fusion (1) – Direct removal (1) – Partial removal + C1 laminectomy + occipitocervical fusion (1)	30 (12–84)	– 90% had neurological improvement – 80% obtained regression of the lesion extent (one extirpated); 2 patients without available follow-up MRI
Klas <i>et al.</i> (2018) ^[6]	– 1 patient – 80 yrs. – Female	No	– Subaxial spondylosis	– Collar for 12 months (weaning at 8 months)	17	– Neurological improvement – Regression of lesion in MR performed at 7 months after presentation
Kobayashi <i>et al.</i> (2018) ^[7]	– 29 patients (17 in fusion group; 12 in no fusion group) – MA 72.5 yrs. (fusion group)/ 77.5 yrs. (no fusion group) – M:F≈5:1 (fusion group)/ 3:1 (no fusion group)	– 88% of patients in fusion group – None in no fusion group	– Subaxial spondylosis (41.4% of all patients) – OALL (20.7% of all patients)	– Fusion group: C1-C2 fixation without resection of C1 posterior arch (12); C1-C2 fixation with resection of C1 posterior arch (5) – No fusion group: C1 laminectomy (12)	54.3 (fusion group)/49.8 (no fusion group)	– All patients had neurological improvement (without statistical difference between groups) – Regression of lesion: 100% in fusion group versus 42% in no fusion group ($P<0.01$)
Nakazawa <i>et al.</i> (2019) ^[8]	– 1 patient – 87 yrs. – Female	Yes	Subaxial spondylosis	– Philadelphia collar for 12 months, followed by plastic collar for 12 months	≈2	– Neurological improvement – Regression of lesion size in MR performed at 11 months after discharge

(Contd...)

Table 2: (Continued).

Author (year)	Sample characteristics	AAI	Associated conditions	Treatment	Follow-up duration in months	Outcome
Park <i>et al.</i> (2017) ^[9]	– 38 patients – MA 47 yrs. (3–76) – M:F≈1:2	Present in 12 patients	– Os odontoideum (31.6%)	– C1-C2 fixation (33) – Occipitocervical fixation (5)	12.5	– 56.2% had excellent, good or fair recovery rate** after surgery – Reduction of retro-odontoid lesion in all the patients
Tanaka <i>et al.</i> (2010) ^[13]	– 1 patient – 72 yrs. – Male	No	– Subaxial spondylosis	– C1-C2 laminectomy + biopsy + posterior C1-C2 transarticular fixation	6	– Neurological improvement – Regression of lesion size in MRI
Yonezawa <i>et al.</i> (2013) ^[14]	– 11 patients – MA 65.3 yrs. (47–78) – M:F≈1:2	Present in all cases	– Non-detailed data	– C1-C2 fixation with screw and hook system (5) – C1-C2 fixation with transarticular screw (3) – C1 laminectomy + occipitocervical fixation (3)	≈1 (1–5)	– 100% had neurological improvement – Reduction of retro-odontoid lesion in all the patients

PPL: Periodontoid pseudotumoral lesions, AAI: Atlantoaxial instability, MA: Mean age, OALL: Ossification of the anterior longitudinal ligament, ROM: Range of motion. *Corresponds to the follow-up of 4 patients; 1 died 10 days after surgery. **Recovery rate according to Hirabayashi method: 75%<to≤100% (excellent), 50%<to≤75% (good), 25%<to≤50% (fair).

hemodialysis.^[2] Tanaka *et al.*^[13] classified these lesions into three types, those with: (1) atlantoaxial subluxation, (2) subaxial spondylosis, or (3) secondary to herniation of an intervertebral disc.

The first patient we presented had a Tanaka's type 2 ROP lesion without AAI.

Crowned dens syndrome (CDS)

In CDS, calcium pyrophosphate crystal deposition leads to expansion of the soft tissues and local inflammatory reaction (i.e. similar to pseudogout). CT is the most useful method for diagnosis.^[4,10] Clinical treatment, which includes nonsteroidal anti-inflammatories (in some cases, corticosteroids with or without colchicine),^[10] is usually sufficient and is associated with a good prognosis.^[12]

MR imaging of PPL and documentation of three types of lesions

Yonezawa *et al.* classified retro-odontoid lesions in three types based on MR signals: type I (inflammatory, typical of *pannus*) – hyposignal in T1 and hypersignal in T2; type II (fibrous) – hyposignal in T1 and T2; and type III (mixed) – heterogeneous signal in T2.^[14]

Non-surgical and surgical management of PPL

The ideal approach to cases of PPL may be non-surgical or surgical [Table 2].

Since those without instability do not typically warrant fusions,^[3] some authors advocate simple decompression, but others indicate fixation if subaxial spondylosis is present.^[13] In this sense, Kobayashi *et al.* found no statistical differences between the fusion and non-fusion groups regarding neurological outcome, although lesion regression was significantly greater in those undergoing fixation.^[7]

For those with AAI, fusions are usually warranted. Atlantoaxial and occipitocervical fixations are associated with good rates of recovery and reduction or disappearance of PPL.^[1,9]

Here, case 1 of ROP was successfully managed with cervical collar immobilization for an average of 18 months.^[6,8]

CONCLUSION

The best treatment for PPL requires MR evaluation and X-ray assessment of AAI; the latter may warrant fusion.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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