Periodontoid pseudotumoral lesions

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

| 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
| Author (year)                  | Sample characteristics | AAI               | Associated conditions | Treatment                                                                 | Follow-up duration in months | Outcome                                                                 |
|-------------------------------|------------------------|-------------------|-----------------------|-----------------------------------------------------------------------------|------------------------------|------------------------------------------------------------------------|
| de Souza Batista et al. (2013) | – 5 patients          | None              | Subaxial spondylosis  | – 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 |
|                               | – MA 64.8 yrs.         |                   |                       |                                                              |                              | – 90% had neurological improvement – 80% obtained regression of the lesion extent (one extirpated); 2 patients without available follow-up MRI |
|                               | (55–76)                |                   |                       |                                                              |                              |                                                                        |
|                               | – M:F=3:2              |                   |                       |                                                              |                              |                                                                        |
| Chikuda et al. (2009)         | – 10 patients         | Present in 2 patients | Reduced ROM C2-C7 OALL (60%) | – C1 laminectomy + occipitocervical fusion (5) – C1 and C2 laminectomy + 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 |
|                               | – MA 71 yrs. (58–82)   |                   |                       |                                                              |                              |                                                                        |
|                               | – M:F=3:2              |                   |                       |                                                              |                              |                                                                        |
| Klas et al. (2018)            | – 1 patient           | 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 – 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) |
|                               | – 80 yrs.              |                   |                       |                                                              |                              |                                                                        |
|                               | – Female               |                   |                       |                                                              |                              |                                                                        |
| Kobayashi et al. (2018)       | – 29 patients         | – 88% of patients in fusion group | Subaxial spondylosis (41.4% 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) | – Regression of lesion size in MR performed at 11 months after discharge |
|                               | (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) |                   |                       |                                                              |                              |                                                                        |
| Nakazawa et al. (2019)        | – 1 patient           | 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 |
|                               | – 87 yrs.              |                   |                       |                                                              |                              |                                                                        |
|                               | – Female               |                   |                       |                                                              |                              |                                                                        |
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-inflammatory drugs (in some cases, corticosteroids with or without colchicine),[10] is usually sufficient and is associated with a good prognosis.[11]

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,[13] some authors advocate simple decompression, but others indicate fixation if subaxial spondylosis is present.[11] 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.[19]

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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