Endoscopic biopsy sampling of tophaceous gout of the odontoid process

Case report and review of the literature

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The authors present the case of a 71-year-old man who presented with neck pain, a history of gout, and a mass in the dens. Results of transoral endoscopic biopsy sampling demonstrated tophaceous gout. The patient was treated medically and the pain resolved. Tophaceous gout isolated in the dens is extremely rare and should be considered in the differential diagnosis of masses in this region. With the aid of transoral or transnasal endoscopic biopsy sampling, the diagnosis can be reached in a minimally invasive manner. (DOI: 10.3171/SPI-07/07/061)

KEY WORDS  • crystal-associated disease • dens • endoscope • gout • odontoid • tophus

CRYSTAL-associated diseases such as gout and pseudogout are known but rare causes of joint disease in the cervical spine. In 1970 Tkach14 reviewed the epidemiology and presentation of gout in the spine and found that 74 of 100 patients with gouty arthritis complained of significant neck pain. However, the cervical spine represents an uncommon site for tophaceous gout detectable on imaging, although it can cause erosions of the posterior elements, vertebral bodies, and facets.5,13 Among the 13 reported cases of gout involving the cervical spine, there is only one prior case of spinal gout involving C1–2 only, and none limited solely to the odontoid process.1–4,6,8,10–13,15–17 We add an additional case to the literature and emphasize the importance of including gout in the differential diagnosis of masses of the odontoid process, particularly in patients with a history of gout.

Case Report

History and Presentation. This 71-year-old man with a 30-year history of tophaceous gout presented with neck pain. His medical history was significant for tophaceous gout of the appendicular skeleton but no disease in the axial skeleton. He also had a history of rheumatoid arthritis and hypertension. There were no risk factors for cancer or any family history of malignant disease.

Imaging studies obtained included computed tomography, MR imaging with and without contrast, flexion/extension views of the cervical spine, and a nuclear bone scan.
Computed tomography and MR imaging demonstrated a lytic, expansile mass in the odontoid process (Fig. 1), with heterogeneous enhancement with Gd. There was no evidence of compression on the craniocervical junction or cervical spinal cord. A bone scan did not demonstrate radioisotope uptake in the lesion, and there was no evidence of atlantoaxial instability on flexion/extension views.

**Pathological Examination.** To determine the diagnosis, the patient underwent stereotactic endoscopic transoral biopsy sampling of the odontoid lesion. A transnasal biopsy was not possible due to the anatomy of the palate. With the patient in a state of general anesthesia, the soft palate was elevated by suturing it to a red rubber catheter that was passed through the nose. Using a rigid 0° 18-cm, 4-mm endoscope (Karl Storz) guided with frameless stereotaxy (StealthStation, Medtronic-Sofamor Danek), a linear incision was made along the posterior oropharynx, the muscle and soft tissue were retracted, and the dens of C-2 was exposed (Fig. 2). The dens was partially drilled to reveal a flaky white substance that was sent for pathological analysis. Frozen sectioning of the tissue revealed a foreign body granuloma. Frozen sectioning of the tissue revealed a foreign body granuloma. The defect in the oropharynx was closed with fibrin glue (Tisseel, Baxter). Postoperative flexion/extension views of the cervical spine revealed no atlantoaxial instability. Final pathological examination revealed negatively birefringent needle-shaped crystals with fibrous inflammatory tissue and the presence of foreign body granulomas consistent with gout.

**Treatment.** The patient was treated medically for gout with colchicine, allopurinol, and glucocorticoids, and his neck pain improved.

**Discussion**

Tophaceous gout, although more common in the appendicular skeleton, can occur in the spine and lead to significant pain, radiculopathy, and/or myelopathy. Spinal involvement is relatively rare, however. Only 13 cases of cervical gout have been reported in the literature (Table 1). Erosion of the vertebral bodies, disc space narrowing, subluxation, and invasion into the spinal canal and foramina may be observed in such cases. Yen et al. described a case of cervical myelopathy caused by gout-induced cervical spondylosis related to intervertebral disc space disease. The true incidence of gout involving the cervical spine remains unknown and involvement of the dens is extremely rare. There is only one other reported case of odontoid gout in which the patient presented with a Type II odontoid fracture with associated myelopathy and tetraparesis. With the present study, we add a second case to the literature.

The differential diagnosis of cervical gout may include infection, oncological processes (often metastatic disease), and rheumatological disease. Typical imaging findings in spinal gout cases include low-signal intensity on T1- and T2-weighted imaging with some contrast enhancement. However, findings on MR imaging may also include intense homogeneous enhancement. Duprez and colleagues reported such findings in a case of cervical gout visible on...
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imaging that resembled a discovertebral infection. The authors theorized that such enhancement was probably secondary to the presence of vascularized reactive tissue. In addition to infection, other crystal-associated diseases may resemble gout. Indeed, Feydy et al.\(^5\) have reported that hydroxyapatite crystal deposition disease and calcium pyrophosphate dehydrate deposition disease are more common in the spine than gout. In particular, the crowned dens syndrome may resemble gout. In this case, either hydroxyapatite crystal deposition disease or calcium pyrophosphate dehydrate crystals deposit around the odontoid process. Punctate calcified deposits may be visualized on MR imaging, permitting a more definitive diagnosis.\(^5\) Ultimately, direct pathological examination is best for confirming the diagnosis. Degenerative osteoarthritis, also included in the differential diagnosis, may also cause osteophytic formation and cystic changes in the odontoid process. Rheumatoid arthritis can also be confused with crystal-associated diseases especially if atlantoaxial subluxation is present. Although cervical gout is the least common entity in this differential diagnosis, the patient’s clinical history is vital: suspicion for spinal gout is clearly higher in patients with a history of tophaceous gout.

The treatment of cervical tophaceous gout depends on its location and presentation. Medical therapy remains fundamental, while surgical intervention is typically reserved for patients with neurological symptoms related to mass effect or evidence of spinal instability. However, Jacobs et al.\(^6\) reported on a patient with an unstable odontoid fracture secondary to gout and spastic quadriplegia who refused surgery and improved dramatically with medical therapy.

Conclusions

Tophaceous gout of the odontoid process, although rare, is important to consider in a patient with a history of gout who presents with an inflammatory mass in the odontoid process. Endoscopic biopsy sampling\(^7\) and medical management may be adequate unless cervical instability is present.

Disclaimer

None of the authors involved in this study has a financial interest or relationship with any device used.

References


Fig. 2. Endoscopic view of the lesion via the transoral approach demonstrating the limited exposure necessary to obtain an adequate biopsy sample.

Table 1

<table>
<thead>
<tr>
<th>Authors &amp; Year*</th>
<th>Spinal Levels Involved</th>
<th>Treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>Kersley et al., 1950</td>
<td>C1–2, T8–9</td>
<td>traction, collar</td>
</tr>
<tr>
<td>Vinstein &amp; Cockerill, 1972</td>
<td>C3–4</td>
<td>pharmacological, cervical traction</td>
</tr>
<tr>
<td>Magid et al., 1981</td>
<td>C1–2, C3–4</td>
<td>pharmacological</td>
</tr>
<tr>
<td>Sequeira et al., 1981</td>
<td>C5–7</td>
<td>laminectomy &amp; biopsy sampling</td>
</tr>
<tr>
<td>Miller &amp; Percy, 1984</td>
<td>C1–2, C6–7</td>
<td>Philadelphia collar, pharmacological</td>
</tr>
<tr>
<td>Jacobs et al., 1985</td>
<td>C1–2, C6–7</td>
<td>pharmacological</td>
</tr>
<tr>
<td>Alarcon &amp; Reveille, 1987</td>
<td>C6–7</td>
<td>anterior removal of tophi, pharmacological</td>
</tr>
<tr>
<td>van de Laar et al., 1987</td>
<td>occipital–C3</td>
<td>pharmacological</td>
</tr>
<tr>
<td>Sabharwal &amp; Gibson, 1988</td>
<td>C5–7</td>
<td>anterior corpectomy, foraminotomies &amp; fusion</td>
</tr>
<tr>
<td>Duprez et al., 1996</td>
<td>C3–5</td>
<td>anterior microdiscectomies w/ interbody fusion</td>
</tr>
<tr>
<td>Yen et al., 2002</td>
<td>C3–6</td>
<td>anterior decompression/ fusion</td>
</tr>
<tr>
<td>Diaz et al., 2003</td>
<td>C4–5</td>
<td>biopsy sampling, pharmacological</td>
</tr>
</tbody>
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* Each author reported only one case.


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