Xanthogranulomatous sialadenitis: A case report and literature review

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Abstract
Xanthogranulomatous tissue reaction is an uncommon but well-documented process that occurs at many sites in the body. It is most often recognized in the kidney and gallbladder, where its etiology is believed to involve an outflow obstruction. We report the case of a man with a parotid mass that exhibited features consistent with an inflammatory process on fine-needle aspiration biopsy. The mass persisted despite medical management, and the patient subsequently underwent a superficial parotidectomy. Histologic examination of the resected specimen identified a xanthogranulomatous tissue reaction adjacent to a Warthin’s tumor. We compare the features of this case with those of the 2 previously reported cases of xanthogranulomatous sialadenitis, and we discuss its possible etiologies.

Introduction
Xanthogranulomatous tissue reaction is most common in the kidney, where it is believed to develop in the setting of a suppurative infection with renal outflow obstruction. Its involvement in the kidney can occur in a diffuse, focal, or segmental distribution, and sometimes it presents as a mass mimicking a malignancy. Xanthogranulomatous reactions have been described less often at other sites, including the ovary, fallopian tube, endometrium, testicle, appendix, gallbladder, bladder, pituitary gland, colon, prostate, retroperitoneum, and adrenal gland.

In this article, we describe the clinicopathologic findings in a case of xanthogranulomatous sialadenitis that involved the parotid gland in the setting of a Warthin’s tumor, and we compare it with the 2 previously reported cases of xanthogranulomatous sialadenitis (1 case was reported twice).

Case report
A 61-year-old man presented with systemic symptoms of gastric Hodgkin’s lymphoma and a right parotid mass. Following treatment of the lymphoma, we addressed the parotid mass, which had become larger and more painful. The parotid symptoms initially resolved with antibiotic treatment, but they returned after treatment was stopped. A subsequent course of antibiotic therapy was initiated, but the symptoms were refractory to treatment. On fine-needle aspiration biopsy (FNAB), the mass exhibited features consistent with an inflammatory process.

Two months later (1 year after his initial presentation), the patient underwent a superficial parotidectomy. Intraoperatively, a small portion of the mass was sent to pathology for analysis and consultation. Frozen-section analysis identified a proliferation of spindle cells and associated inflammation. Based on these findings, a diagnosis of a reactive/inflammatory process was favored. The entire resected specimen measured 7 × 3 × 3 cm. The mass contained a cystic cavity that was filled with a thick brown fluid. A small portion of tan, finely lobulated salivary gland was identified adjacent to the cavity. The remainder of the tissue was tan-yellow, nodular, and friable.

Histologic analysis identified a xanthogranulomatous tissue reaction characterized by fibroblastic proliferation with mixed inflammatory cells, including abundant foamy macrophages, neutrophils, plasma cells, and lymphocytes (figure). Adjacent to the inflammatory process was a Warthin’s tumor.

Immunohistochemical stains for macrophages (CD68) and epithelial cells (cytokeratin CAM 5.2 and cytokeratin AE1/AE3) were performed on the formalin-fixed, paraffin-embedded tissue according to the streptavidin-biotin-peroxidase method. The results of these stains in the area of fibroblastic proliferation confirmed the presence of abundant macrophages and the absence of epithelial cells. Grocott methenamine-silver and acid-fast stains were negative for fungal and mycobacterial organisms, respectively.

At 2 years of clinical follow-up, the patient’s Hodgkin’s lymphoma was in complete remission and he exhibited no evidence of residual or recurrent xanthogranulomatous sialadenitis or Warthin’s tumor.
Discussion

Xanthogranulomatous reaction in the kidney is a well-documented complication of suppurative renal infection. The mechanism is believed to be associated with renal outflow obstruction in the face of pyogenic infection. It can present as a localized mass lesion or in a diffuse form that obliterates renal parenchyma. In either case, it mimics renal malignancy.

Xanthogranulomatous tissue reaction has been reported at many other sites,1-10 but the cause in these cases has not been well characterized, probably because of the relatively small number of cases that have been reported. Inflammation and obstruction are frequently cited as common factors in the development of the xanthogranulomatous reaction. A review of 40 cases of xanthogranulomatous cholecystitis suggests that the process in the gallbladder stems from an obstruction of the Rokitansky-Aschoff sinuses by inspissated bile followed by subsequent rupture and a xanthogranulomatous reaction.4 Xanthogranulomatous appendicitis is usually an incidental finding in patients with chronic inflammation and obstruction by fecalith.3

In our case, the histologic and immunohistochemical findings were those of a xanthogranulomatous tissue reaction adjacent to a Warthin’s tumor. Our search of the English-language literature revealed only 2 previously reported cases of a xanthogranulomatous reaction involving the parotid gland.11-13

All 3 reported cases of xanthogranulomatous sialadenitis have two significant factors in common (table). First, all were associated with a preexisting parotid neoplasm (a Warthin’s tumor in 2 cases and a pleomorphic adenoma in the other). Second, all 3 patients underwent FNAB prior to surgical excision. In the case reported by Stephen et al, evidence of a Warthin’s tumor was found in the FNAB sample but not in the surgically resected specimen, which contained only the xanthogranulomatous tissue reaction.13 This suggests that the xanthogranulomatous reaction occurred in response to the FNAB. In our case, the FNAB showed features of an inflammatory process only, suggesting that the development of the xanthogranulomatous reaction preceded the biopsy. The predominant finding in

Table. Comparison of the 3 reported cases of xanthogranulomatous sialadenitis

<table>
<thead>
<tr>
<th>Author</th>
<th>Patient’s age/sex</th>
<th>Significant history</th>
<th>Previous procedure</th>
<th>Postop diagnosis</th>
<th>Time between FNAB and excision</th>
</tr>
</thead>
<tbody>
<tr>
<td>Choyce et al,11</td>
<td>60/F</td>
<td>Thyrotoxicosis, atrial fibrillation, removal of right parotid pleomorphic adenoma</td>
<td>FNAB: necrotic tissue and inflammatory debris</td>
<td>XG sialadenitis</td>
<td>NR</td>
</tr>
<tr>
<td>Padfield et al,12</td>
<td>56/M</td>
<td>None</td>
<td>FNAB: Warthin’s tumor</td>
<td>XG sialadenitis</td>
<td>1 mo</td>
</tr>
<tr>
<td>Stephen et al,13</td>
<td>61/M</td>
<td>Hodgkin’s lymphoma</td>
<td>FNAB: cellular debris and acute inflammation</td>
<td>Warthin’s tumor with XG reaction</td>
<td>2 mo</td>
</tr>
</tbody>
</table>

* Present report.
FNAB = fine-needle aspiration biopsy; XG = xanthogranulomatous; NR = not reported.

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