ABSTRACT

Objectives
To describe the clinical, radiographic, and histopathological features of a rare benign disease of the parotid gland: xanthogranulomatous sialadenitis.

Study Design
Case report

Methods
A case report and literature review of xanthogranulomatous sialadenitis of the parotid gland.

Results
A patient presented with a parotid mass and marginal mandibular nerve weakness suspicious for malignancy. Fine needle aspiration was inconclusive. Computed tomography revealed an irregular, spiculated, heterogeneous mass of the right parotid gland. The patient underwent a total parotidectomy with facial nerve preservation. The final pathology revealed xanthogranulomatous sialadenitis. Several weeks after surgery, the patient regained complete facial nerve function.

Conclusions
A clinical and radiographic mimic of malignancy, xanthogranulomatous sialadenitis is a rare, benign condition of the parotid gland. Treatment consists of surgical excision.

Disclosures
Nothing to disclose.

INTRODUCTION

Salivary gland tumors account for 3-6% of head and neck tumors encountered in the United States, with 70-80% of masses being found within the parotid gland. While these tumors are more commonly benign, the physician must maintain a high suspicion for malignancy until proven otherwise. Frequently, the most conclusive means of proof is via surgical resection of the mass by superficial or total parotidectomy.

In this report, we will review a case in which a parotid tumor showed multiple clinical and radiographic features consistent with malignancy. Based on these features as well as an inconclusive fine needle aspiration biopsy, the patient underwent total parotidectomy. Interestingly, final histopathological analysis revealed xanthogranulomatous sialadenitis, a rare, benign cause of sialadenitis that has been previously reported to mimic malignancy. We will report the findings of this case and highlight features of its unusual diagnosis.

CASE PRESENTATION

A 62-year-old female presented to our otolaryngology clinic with a one-month history of a non-painful right parotid mass and right lower facial weakness on smiling. The patient's past medical history was significant for hypertension, hyperlipidemia, and a mild stroke but she denied previous head or neck masses or facial paralysis. Her past surgical history included tonsillectomy, hysterectomy, and oophorectomy. The patient denied any history of cigarette or alcohol use. On initial physical examination, a right parotid mass of approximately 2 cm diameter was palpated. The mass was nontender and no cervical lymphadenopathy was appreciated. Cranial nerves II through XII were grossly intact with the exception of the right marginal mandibular branch of cranial nerve VII, which was weak on full smile. There were no scars or skin lesions in the head and neck region. The remainder of the physical examination was unremarkable.

Fine needle aspiration of the mass was inconclusive. A computed tomography (CT) scan of the neck with and without contrast was ordered and showed a 1.7 x 1.2 x 1.0 cm poorly marginated mass within the lower middle aspect of the right parotid gland. The mass enhanced with contrast, showed a spiculated contour, and distorted the internal architecture of the right parotid gland. Central areas of low density were also seen within the mass. Adjacent soft tissue stranding was present but there was no extracapsular extension of the mass. Two additional sub-centimeter masses that enhanced with contrast were also seen within the gland.

With consideration of the patient's marginal mandibular nerve involvement and aggressive radiographic features, our index of suspicion for a parotid malignancy was high. The patient was apprised of surgical versus observational management options and elected to undergo total right parotidectomy.

RESULTS

The patient underwent right total parotidectomy. The resected mass was sent for frozen and permanent pathological analysis. Frozen analysis returned that the mass was consistent with an infected lymphoepithelial cyst and chronic sialadenitis. At this time, the plastic surgery team was consulted and closed the wound primarily in three layers. There were no intraoperative complications and the patient was sent to recovery in stable condition.

Final histopathological results were consistent with xanthogranulomatous sialadenitis. Right marginal mandibular nerve weakness resolved by one-month follow-up. There were no post-operative complications and no evidence of recurrence at nine months.

DISCUSSION

A xanthogranulomatous tissue reaction occurs uncommonly but has been described in many areas of the body, most classically in the kidney, gallbladder, and appendix. Very few reports have been published about its diagnosis within the salivary glands, making it a rare but important entity in the head and neck region. It has been shown to mimic malignancy in other published cases in the parotid gland as well as in the kidney, but its benign diagnosis obviates the need for adjuvant therapy that would typically occur with a malignant lesion.

On histologic analysis, xanthogranulomatous sialadenitis is comprised of numerous foamy, lipid-laden macrophages aligned in sheets with surrounding neutrophils, lymphocytes, plasma cells, and foreign body giant cells (Figure 3). Areas of palisading histiocytes and spindled-shaped cells are commonly found at the periphery. There is usually a variable degree of tissue fibrosis. On gross pathology, the tissue has been described as yellow and friable centrally with a more grey, fibrotic appearance in the periphery. A specific mechanism for the development of a xanthogranulomatous tissue reaction has not been fully elucidated but one study has suggested its possible relation to ductal outflow obstruction.

While the imaging studies were consistent with a more aggressive tumor, the patient's new-onset facial nerve weakness further concerned us about malignancy. The age of our patient also increased our suspicion, as malignant parotid tumors are more commonly seen in patients over 45 years of age. With the available preoperative diagnostic information, we hold that total parotidectomy was the safest course of action for this patient. Other reported cases of xanthogranulomatous sialadenitis were likewise not diagnosed until after surgical intervention, most likely because of similarly concerning features that prompted more aggressive management.

CONCLUSIONS

We conclude that surgical management was indicated in this case secondary to clinical and radiographic features of the mass that were suspicious for malignancy. Though xanthogranulomatous sialadenitis is a rare entity, the astute clinician should include this benign condition in the differential diagnosis of parotid tumors.

REFERENCES