A Case of Disfiguring Bilateral Parotid Sialosis

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ABSTRACT

Objectives: Bilateral non-inflammatory sialosis has been described in the literature to be a rare presentation of a number of systemic pathologies. This paper presents a case report of bilateral parotid sialosis in a diabetic female with pathology and review of the literature.

Study Design: This is a case report of parotid sialosis with a review of the primary literature.

Methods: A comprehensive review of the literature was performed. Articles not available online were obtained in print. The literature was reviewed regarding the incidence of parotid sialosis and associated conditions that have been described.

Results: We present a patient with bilateral parotid sialosis and diabetes mellitus. Histologically, she was found to have benign, diffuse fatty infiltration of the parotid glands bilaterally. Several case reports from the 1950s describe fatty infiltration of the parotid in obese individuals, while more recent reports describe sialosis in association with diabetes mellitus and alcoholism. Many of the recent studies, however, do not provide a review of histology, making it difficult to extrapolate an etiologic relationship with our patient from this data. This report highlights the potential pathologic relationship between diabetes mellitus and fatty infiltration of the parotid gland.

Conclusions: Bilateral parotid gland swelling secondary to fatty infiltration is rarely discussed in the otolaryngologic literature. Generally these patients are worked up in conjunction with primary care physicians and rheumatologists, nonetheless a primary source is not always identified and in reviewing the literature there does not appear to be a clear pathologic process described in the patients. The most common association is diabetes mellitus.

INTRODUCTION

Parotid enlargement is commonly described in multiple settings including infection, inflammation, autoimmune disease, and neoplasia. Furthermore, sialosis is defined as painless, diffuse, non-inflammatory, non-neoplastic enlargement of the major salivary glands (1). It has been described in a number of conditions including as diabetes mellitus, alcoholism, and liver disease (1, 2, 6, 7). The histopathology of the sialosis in these cases, however, varied greatly with regards to the content of the glands, ranging from hypotrophic acini to diffuse fatty infiltration. Here we describe diffuse fatty infiltration in a 44-year-old Caucasian female with a 4-year history of persistent, bilateral, diffuse, non-inflammatory, non-neoplastic swelling of the parotid glands. The patient’s comorbidities of Type 2 diabetes mellitus and dermatomyositis have previously been associated with painless sialosis (1). Studies have shown that in the diabetic population with sialosis, histopathologic evaluation demonstrates abundant adipose infiltration in the stroma (7), which is what we report in the following case.

CASE REPORT

A 44-year-old Caucasian female presented to the ENT clinic in May 2009 with a two-year history of bilateral parotid gland swelling. Symptoms included occasional pain since the fall of 2007, fluctuating size day-to-day, and effortless swallowing symptoms. She reported no change in symptoms with eating. There were no initiating events, no history of bulimia or frequent emesis, and no history of thyroid problems or rheumatoid arthritis. Review of systems was positive for joint pain.

The patient’s past medical history was significant for Type 2 Diabetes, hypertension, hypercholesterolemia, anemia related to uterine fibroids, and a history of dermatomyositis treated with high-dose prednisone in 1989. She denied tobacco, alcohol, and illicit drug use. Medications included metformin/rosiglitazone, glyburide, simvastatin, valsartan, and citalopram.

On physical exam, the patient’s height was 61 inches and weight was 120 pounds, with a BMI of 22.7. The patient demonstrated bilateral symmetric, markedly enlarged parotid and submandibular glands. The oral cavity showed decreased, thick saliva. There was mild proptosis with prominent peribulbar tissue but the remainder of the physical exam, including thyroid, was benign. Initial evaluation with ultrasound showed homogenous enlargement of both parotid glands and submandibular glands with no focal masses within the glands. There was an isolated enlarged lymph node in the left upper neck that appeared to have a normal vascular hilum and echotexture.

Initial laboratory evaluation revealed negative workup for Sjogren’s syndrome and other autoimmune disease, including SS-A, SS-B, RF, and ANA. She had a persistently mildly elevated sedimentation rate. The patient was seen by Rheumatology who had previously followed the patient for dermatomyositis. Despite negative laboratory testing, rheumatology still had a strong suspicion of Sjogren’s and prior to starting immunosuppressive therapy a definitive tissue specimen was requested.

CASE REPORT, CONTINUED

After a needle core biopsy failed to yield a sufficient tissue sample, an excisional biopsy of the right parotid gland tail was performed under local anesthesia. The biopsy showed serous salivary gland tissue, mildly ectatic salivary duct with inspissated material and dystrophic calcification, and no significant inflammation. Post-biopsy follow-up revealed no complications from the procedure.

The patient returned the following year with persistent parotid swelling and recent onset of bilateral axillary tenderness raising a suspicion of an underlying lymphoproliferative disorder. After discussions with rheumatology and the patient, she underwent a right superficial parotidectomy with facial nerve dissection in December 2009. Flow cytometry revealed no evidence of a neoplastic population. The pathologic findings are shown below. The patient did well post-operatively and elected to proceed with a left-sided superficial parotidectomy several months after the initial procedure.

PATHOLOGY

Figure 1. (Hematoxylin and Eosin 10X) There is a marked increase in adipose tissue separating the salivary gland acini.

Figure 2. (Hematoxylin and Eosin 20X) Higher magnification to illustrate the marked increase in fat and the displacement of salivary acini and ducts.

PATIENT PHOTOS

Figure 1. Frontal view status post right superficial parotidectomy

Figure 2. Oblique view showing persistent left-sided enlargement

DISCUSSION

This is the case of a 44-year-old Caucasian female who presented with a four-year history of persistent, bilateral, diffuse, non-inflammatory, non-neoplastic swelling of the parotid glands. Final workup revealed hyperglycemia with diffuse fatty infiltration.

In the 1950s, there were several reports describing parotid gland hypertrophy with diffuse fatty infiltration (2, 3, 4). A cause has not been identified, and obesity was a unifying co-morbidity in many of the patients described, although there was report of a parotid nodule characterized by fatty infiltration in a thin individual (4). The patient we describe in this case likewise had a normal BMI at 22.7, but has diabetes mellitus which has been linked to sialosis in the more recent literature (1).

Scully et al (1) reviewed 35 cases of sialosis with respect to the presentation, symptoms, and other related conditions. Diabetes mellitus was the most commonly identified association, with alcoholism, antihypertensive agents, bulimia, and hypothyroidism comprising the other defined causes. The histology was not reviewed, however, making it difficult to extrapolate an etiologic relationship with our patient from this data. Carda et al (7) studied parotid gland samples from both diabetic and alcoholic patients with sialosis, and found that diabetics had abundant adipose infiltration in the stroma when compared to alcoholics.

It has been hypothesized that malnutrition may play a role in the pathogenesis of sialosis (1, 2). Patients with diabetes mellitus may fall under this category as fat metabolism is modified when compared to non-diabetic patients. In 1978, an animal study in streptozotocin-induced diabetic rats showed that insulin had an effect on salivary gland lipid metabolism, as diabetic rats treated with insulin experienced a normalization of both lipid content and fatty acid profiles after one week of treatment (5). This suggests that diabetic patients who are treated with oral hypoglycemic agents, such as our patient, may not be experiencing the benefits that insulin may provide on normalizing fat content in the major salivary glands.

CONCLUSIONS

Bilateral parotid gland swelling secondary to fatty infiltration is rarely discussed in the otolaryngologic literature. Generally these patients are worked up in conjunction with primary care physicians and rheumatologists, nonetheless a primary source is not always identified and in reviewing the literature there does not appear to be a clear pathologic process described in the patients. The most common association is diabetes mellitus. Superficial parotidectomy can be used to both rule out any malignant process and address the disfiguring lesion.

REFERENCES


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