An integrated approach to the diagnosis and management of parotid sialadenosis

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INTRODUCTION

Sialosis (sialadenosis) is a chronic, bilateral, diffuse, non-inflammatory, non-neoplastic painless swelling of the major salivary glands that primarily affects the parotid glands, but occasionally involves the submandibular glands and rarely the minor salivary glands (Scully 2008). The current case describes the workup, diagnosis and treatment of parotid sialosis presenting with episodic painful swelling in a patient who does not have the commonly ascribed underlying conditions commonly thought to predispose one to sialosis.

DIAGNOSIS

Sialosis may be idiopathic but may be associated with chronic malnutrition, obesity, diabetes mellitus, alcoholism, liver disease, eating disorders and medication, particularly antihypertensives (Duggan 1957). However, the pathophysiology of the disease is not well understood and there is no well-defined systematic approach to its diagnosis.

The case begins with a wide differential for bilateral parotid swelling including infectious, neoplastic, autoimmune and iatrogenic causes. Serial H&P suggested episodes of acute swelling superimposed on normally bilaterally swollen parotid glands in this patient. Baseline parotid enlargement had been present for at least a couple of years, was diffuse and painless between acute episodes. Empiric antibiotics were used first to address common bacterial causes of parotitis including S. aureus, S. viridans, H. influenzae. When this failed, rheumatology workup was carried out to rule out autoimmune causes. Imaging studies (CT and sialogram) followed to rule out neoplastic and obstructive causes that may not have been detected on physical examination. These studies demonstrated diffuse bilateral parotid enlargement and diminished ductal arborization without any evidence of obstruction as would be seen in sialolith or ductal stenosis. With the findings now favoring sialosis, a biopsy was conducted showing mild acinar hypertrophy.

The pathophysiology of sialadenosis remains unclear but the histological findings in this case are consistent with the proposal that unbalanced autonomic input to the glands or abnormal stimulation (diabetes or bulimia) causes acinar hypertrophy.

CONCLUSIONS

Sialosis or sialadenosis is rare and may present a diagnostic challenge.

The key to diagnosis is a systematic approach beginning with a detailed H&P followed by laboratory tests, imaging and finally, tissue biopsy.

Management options used to address sialadenosis are varied and include observation, trismucinone infusion, botulinum toxin A injection, tympanic neurectomy and parotidectomy

REFERENCES

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Case Report

A 54-year-old male with a two-year history of recurring episodic left parotid swelling presented to the University of Iowa Hospital for evaluation. His first episode of parotid swelling occurred in September 2010 and included aural pain and periorbital edema. This was treated empirically with a course of antibiotics and resolved. One month later, he had a second episode in which the swelling was worse than the first episode. He was again prescribed a course of antibiotics, which alleviated this symptoms. The continued to have recurrent parotid swelling with variable response to antibiotics and was finally seen in the Department of Rheumatology at the University of Iowa Hospital in March of 2012. However, a rheumatology workup was negative for ANA, RF, SSA and SSB making autoimmune etiology highly unlikely. He was subsequently referred to the Department of Otolaryngology for further evaluation.

He did not have history of diabetes, liver disease or alcohol use. Studies, which accompanied the patient at the time of his presentation included CT of the face and neck, which did not show any evidence of sialolithasis, abscess or fluid collection. It did however, demonstrate enlarged, symmetric parotid glands. He was subsequently targeted for left parotid sialogram, which was remarkable for shortening of arborizations in the gland (consistent with sialosis). At that point, the patient was offered treatment options for sialosis including watchful waiting, kenalog infusion and parotidectomy. Due to the painful nature of his parotid swelling and the frequency with which it was occurring, he opted to go forward with Kenalog 10 infusion into his left parotid gland. He remained asymptomatic for only two weeks before the painful swelling returned. At that point, he opted to pursue parotidectomy.

Two months after the Kenalog infusion, the patient underwent an uneventful left parotidectomy. Pathology evaluation of the parotid gland demonstrated abundant parenchymal adipose tissue and mild acinar hypertrophy. This finding combined with the radiological and clinical findings confirmed the diagnosis of sialosis. At follow up 15 months after surgery he had not had a recurrence of any salivary gland swelling. He did have a gustatory sweating, which began approximately six month postoperatively along with some slight numbness around the surgical incision.

Below: high magnification H&E section. Findings suggestive of sialosis.

Above: low magnification H&E section from the left parotid gland demonstrating benign salivary gland tissue with abundant parenchymal adipose tissue and mild acinar hypertrophy (arrow) consistent with sialosis. Below: high magnification H&E section.

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