Sialadenitis with Associated Auriculotemporal Symptoms and Syncope: A Case Report and Review of the Literature

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ABSTRACT

Objectives: To describe a rare but severe case of obstructive sialadenitis with associated auriculotemporal symptoms, hypotension, and syncope.

Study Design: A case report and literature review are presented.

Methods: We report the first case of obstructive sialadenitis manifesting with auriculotemporal symptoms of flushing and sweating, progressing to hypertension and syncope. We review the literature on related symptomatology associated with salivary gland pathology.

Results: A 54-year-old man presented with a year-long history of right-sided facial swelling and pain associated with meals. He noticed recurrent episodes of concomitant facial flushing, sweating, and lightheadedness progressing to eventual syncope. While hospitalized, symptoms were reproducible with periprandial parotid gland swelling and pain, followed by sudden and documented hypotension. Abatement of symptoms was noted with parotid massage. A full panel of lab workup failed to yield an inflammatory, autoimmune, infective, or metabolic etiology for his symptoms. A CT neck demonstrated a 8mm sialolith in Stenson’s duct, posterior to the masseter muscle consistent with obstructive sialadenitis. The patient underwent sialendoscopy, transfacial parotidotomy, and saliodochotomy for removal of the stone. He remains symptom-free at one year follow-up.

Conclusions: We present the first known case of obstructive sialadenitis with associated auriculotemporal symptoms, hypotension, and syncope. Although the literature reports on cases of salivary gland neoplasms presenting with heightened carotid sinus sensitivity and associated hypotension, there are no other reports associating this constellation of symptoms with obstructive sialadenitis. Furthermore, auriculotemporal syndrome is associated with trauma or surgery in the region, but has not previously been associated with obstructive sialadenitis. Other potential mechanisms such as glossopharyngeal neuralgia or heightened autonomic response due to mechanosensory function of periparotid issue are presented. Otolaryngologists should be familiar with this rare but unusual presentation of obstructive sialadenitis and the various treatment modalities that are available.

INTRODUCTION

Obstructive sialadenitis may be attributed to intraductal calculi (sialolithiasis), mucus plugs, autoimmune disease, external beam radiation, radioactive iodine, or other conditions which produce a persistent stasis of saliva or salivary duct stenosis. Symptoms of obstructive sialadenitis typically manifest with periprandial salivary gland swelling and tenderness of the involved gland, which abates shortly after eating. If mechanical obstruction of the salivary duct persists, the gland may become secondarily infected with attendant findings such as fever, severe pain of the involved gland, erythema of the overlying skin, and purulent discharge from the duct. Herein, we present the first known case of obstructive sialadenitis presenting with auriculotemporal symptoms and syncope.

CASE PRESENTATION

A 54-year-old gentleman with a history of hypertension and hyperlipidemia was referred to our clinic for evaluation of recurrent right-sided facial swelling, pain, acute facial flushing, diaphoresis, lightheadedness, and syncope. The patient reported that these episodes had occurred intermittently over the course of a few years and that symptoms exclusively presented upon onset of eating. During one particular episode while driving, the patient lost consciousness, crashing his vehicle into a busy intersection without significant injury. He was subsequently admitted to the telemetry unit at an outside hospital where thorough metabolic and cardiac workups were performed for syncope. All of his labs, including a complete metabolic panel and blood counts were within normal limits. An echocardiogram (ECHO) and tracheobronchial esophagogastroduodenoscopy (TTE) revealed no evidence of arrhythmia or cardiac ischemia. Cardiac enzymes were not present and found to be within normal range.

The patient was diagnosed with recurrent obstructive sialadenitis and underwent operative sialendoscopy for evaluation of the stone. During the procedure, the impacted stone was identified in the proximal parotid duct (Figure 2). Attempt at basket retrieval was unsuccessful, thus a transfacial parotidotomy and saliodochotomy was performed with successful retrieval of the stone (Figures 3-5). Postoperatively, the patient was admitted for overnight observation where he was noted to be symptom-free while eating. One year post-operative follow-up revealed complete resolution of symptoms.

DISCUSSION

We present an unusual case of obstructive sialadenitis with associated facial flushing, diaphoresis, lightheadedness, and syncope. While our patient’s symptoms of periprandial facial swelling and pain are typical of this process, we believe this represents the first report of obstructive sialadenitis with associated auriculotemporal symptoms (i.e Frey’s syndrome)—facial flushing and sweating with mastication—and syncope.

In an attempt to elucidate a possible mechanism for our patient’s presentation, we reviewed the literature focusing on an association between parotid pathology and concomitant syncope.

There have been several case reports of parotid neoplasms presenting with vasovagal episodes of syncope. In these cases, it has been postulated that a large, inferiorly-based neoplasm in the tail of parotid region is sufficient to place pressure on the carotid sinus, eliciting heightened vagal tone with subsequent hypertension and bradycardia. It is possible that an obstructed and swollen parotid gland, may place pressure on a high bifurcating carotid system.

Another purported mechanism for this constellation of symptoms may be explained as a variant of glossopharyngeal neuralgia and nerve irritation.

Neuralgia of the glossopharyngeal nerve has been reported previously in the literature and is characterized by intermittent but severe pain in the sensory distribution of the glossopharyngeal nerve. Although most cases of this disease appear to be idiopathic, this presentation has also been reported as secondary to cerebellopontine angle (CPA) tumors, intracranial vascular compression, regional spread of head and neck malignancy, parapharyngeal space abscesses, and cranial base tumors. Despite these associations, syncope appears to be a rare finding in this disease. The most accepted mechanism by which glossopharyngeal neuralgia is thought to elicit syncope is by way of stimulation of CN IX leading to synaptic crossover between the nucleus of the tractus solitarius (CN IX) and the dorsal nucleus of CN X. In this way, irritation of the peripheral portion of CN IX may lead to an efferent vagal response with subsequent bradycardia and inhibition of vasomotor centers.

Lastly, while not clearly elucidated, there remains the possibility of an autonomic reflex arc mediated by stretch or mechanoreceptors within the parotid duct itself. Recently, histologic features consistent with extrinsic injury to parotid ducts have been described in the juxtaparotial organ (of Chievitz) near the angle of the mandible, lending evidence to support this organ’s role in mechanosensory function in the region.6-8It remains unclear whether this pathway would be implicated in our patient’s presentation.

CONCLUSIONS

We present the first known case of recurrent syncope and auriculotemporal symptoms induced by acute parotid obstruction. Proposed mechanisms include the following:

- **External compression of carotid sinus by gland swelling/inflammation**
- **Glossopharyngeal-vagal reflex arc as mediated by a variation of glossopharyngeal neuralgia**
- **Autonomic reflex arc mediated by mechanosensory receptors in the periparotid tissue**

Otolaryngologists should be aware of the various available treatment modalities for sialolithiasis of the parotid duct. Namely, parotid stones posterior to the masseter may require transfacial approach for management.10

REFERENCES