

Obstructive Parotitis Secondary to an Acute Masseteric Bend

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Key Words

Parotitis · Acute masseteric bend · Masseteric hypertrophy

Abstract

Aims: To investigate 3 cases of chronic parotitis secondary to an acute bend in Stensen’s duct caused by an enlargement of the masseteric space. **Results:** Three female patients presented with symptoms consistent with obstructive parotitis including glandular swelling and tenderness during meals. A 10-year-old patient had unilateral facial swelling with enlargement of the masseter muscle and mandible later diagnosed as fibrous dysplasia. Salivary endoscopy showed an acute bend in Stensen’s duct secondary to a mass effect. The patient’s parotid swelling resolved following debulking of the mandibular mass and sialendoscopy with irrigation. Two adult patients with bilateral parotid involvement presented with bilateral masseteric hypertrophy and dental wear facets consistent with bruxism. Salivary endoscopy revealed bilateral kinking of Stensen’s duct with jaw closure. Both patients improved symptomatically following nightly bite guard use and ultrasound-guided Botox injections of the masseter muscle and parotid. **Conclusion:** Obstructive parotitis is rarely caused by an acute masseteric bend. Diagnosis of a kinking Stensen’s duct is aided with salivary endoscopy and imaging to determine the precipitating pathology. In the case of masseteric hypertrophy, symptomatic improvement can be achieved with Botox-induced atrophy of masseteric hypertrophy or with surgical reduction for associated fibrous dysplasia.

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Introduction

Chronic sialadenitis from obstruction of the salivary gland is a relatively common disorder with an estimated lifetime prevalence of 2% of the population [1]. Salivary gland obstruction often results in pain and swelling, primarily while eating, in the cheek area or under the jawline. Foul-tasting drainage can be produced into the mouth. If untreated, limited salivary flow due to obstruction can progress to a significant infection with high fevers, pain and a neck abscess [1].

The causes of chronic sialadenitis include salivary stones, scar tissue, allergic disorders, dehydration, medication side effects, autoimmune diseases and tumors. During the workup, ultrasound and computed tomography (CT) scanning are the imaging studies most helpful for identifying salivary stones, ductal blockage and dilation, or masses. Salivary endoscopy (sialendoscopy) can also assist in the diagnosis and treatment of these pathologies. In this technique, a semirigid, ultrathin endoscope is inserted into the gland’s natural ostium in the oral cavity allowing visualization of the obstructing lesion. Stones and scar tissue can be removed to alleviate the blockage, or the gland can be irrigated with a saline solution and steroids to provide symptomatic control. Over the last decade, this technique has been developed and successfully applied in Europe, but has only recently seen increasing use in the USA [2].

This report investigates 3 recent cases of chronic sialadenitis that presented to the tertiary salivary clinic at the

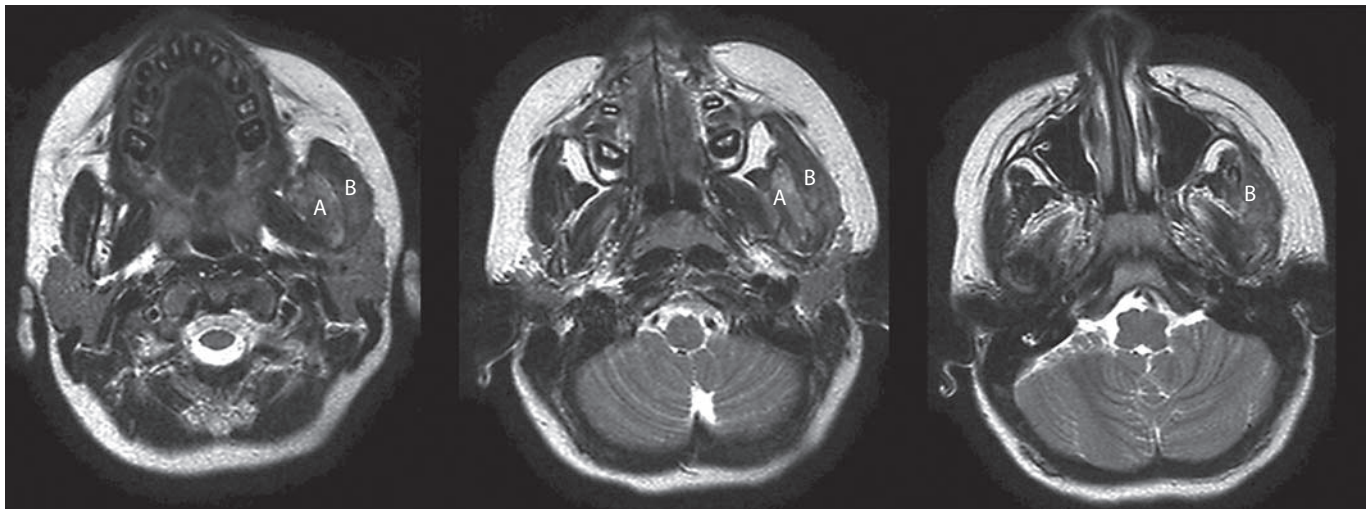


Fig. 1. MRI of patient 1 showing bony expansion (A) of the mandibular ramus and the masseteric hypertrophy (B).

Medical University of South Carolina. With the aid of sialendoscopy, the cause of these patients' obstructions was diagnosed as an acute bend in Stensen's duct caused by enlargement of the masseteric space.

Case Reports

Case 1

An otherwise healthy 10-year-old girl was referred to our clinic with a left-sided facial swelling with associated tenderness. Antibiotic trials had failed to reduce the swelling and tenderness. Extraoral examination showed left-sided facial fullness with a moderately enlarged parotid. Clear saliva was produced from the left Stensen's duct.

Further testing with imaging and salivary endoscopy was performed. CT scan and magnetic resonance imaging (MRI; fig. 1) showed bony expansion of the mandibular ramus (fig. 1, point A) and masseter muscle enlargement (fig. 1, point B). Salivary endoscopy depicted an acute kinking of the left Stensen's duct around the masseter muscle. The endoscope was unable to be completely passed to the parotid hilum, and irrigation of the gland released thick, mucoid debris from the duct.

The patient underwent transoral debridement of the expanded mandibular ramus with debulking of the periosteum and surrounding masseter muscle. The final pathology was consistent with fibrous dysplasia of the mandible. Therefore, the expansion of the masseteric space by the fibrous dysplasia had resulted in kinking and partial obstruction of the left Stensen's duct. Six months after treatment, the patient has done well without facial swelling or recurrent parotitis.

Case 2

A 49-year-old woman presented with recurrent pain and swelling in the right parotid gland, especially during the first bite of food. The patient's history included a left total parotidectomy to treat left chronic parotitis of unclear etiology. Diffuse, uniform swelling that was tender to palpation was present over the right parotid gland. Intraorally, the patient's right Stensen's duct produced clear saliva, and the lower teeth facets were significantly worn exposing dentin (fig. 2). Laboratory results showed negative autoimmune and Sjögren's serologies.

Ultrasound illustrated a dilation of the main right Stensen's duct as it passed over the masseter muscle. Using sialendoscopy an acute bend with proximal dilation in the right main Stensen's duct was visualized as the endoscope was passed around the masseter muscle. A 0.8-mm endoscope was able to pass into the tertiary ducts, but a larger 1.1-mm endoscope could not make it past the masseteric bend. The patient was diagnosed as having masseteric hypertrophy with kinking of Stensen's duct secondary to chronic bruxism.

The patient was treated with a nightly bite guard appliance and ultrasound-guided botulinum toxin A injections. One dose of 25 units was injected in the right masseter muscle, and 3 doses of 25 units were injected in the right parotid gland. The patient noticed reduced pain, swelling and inflammation for 3–4 months but required 1 more treatment with Botox injections. The patient has done well after 12 months of follow-up with only intermittent symptoms of gland obstruction.

Case 3

A 51-year-old woman was referred to our clinic with a 20-year history of recurrent bilateral swelling of the parotid glands primarily during eating. The patient also noted an occasional, intraoral, foul-tasting mucous discharge. On examination, parotid glands and masseter muscles were mildly enlarged and tender bilaterally. Intraorally, clear saliva was produced from both Stensen's ducts. The front teeth showed dental wear facets with dentin ex-



Fig. 2. Patient 2 presenting with worn teeth facets and dentin exposure.

posure and loss of canine heights. Sjögren's serologies were negative.

No stones or masses were found on CT scan, and ultrasound showed a dilation of Stensen's duct bilaterally (fig. 3). During salivary endoscopy, only a 0.8-mm endoscope could pass through the kinking in both Stensen's ducts as they passed over the masseter muscles.

Once the diagnosis of masseter muscle hypertrophy (MMH) with kinking of Stensen's duct secondary to chronic bruxism had been established, the patient was treated with ultrasound-guided botulinum toxin A injections of the bilateral masseter muscles and parotid glands, and a nightly bite guard appliance was prescribed. During 16 months of follow-up, the patient has undergone one additional masseteric Botox injection and has had reduced pain, swelling and inflammation.

Discussion

Numerous pathological enlargements or masses can be found in the masseteric space, also known as the masticator space. These lesions can be extremely varied, including vascular malformations, muscle denervation, autoimmune syndromes, juvenile recurrent parotitis, branchial cleft cysts, MMH, bacterial or viral infections, fibrous dysplasia, radiation necrosis, local and metastatic malignant spread, and tumors of the muscle, mandible or salivary glands [3, 4]. Of these, MMH is a fairly rare condition with less than 200 reported cases in the literature [5]. Most commonly it is an asymptomatic enlargement of the masseter muscle that can present unilaterally or bilaterally. There is a higher occurrence between the ages

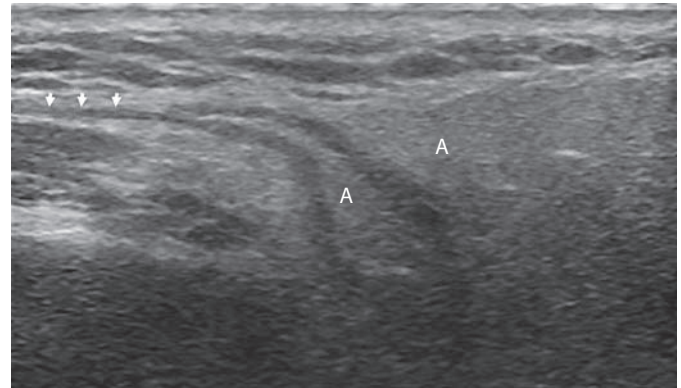


Fig. 3. Ultrasound of the left parotid (A) of patient 3 showing a dilation of Stensen's duct with extension of the dilation into the branches beyond the gland hilum (arrows).

of 10 and 40 years, and a slightly higher rate in men compared to women [6, 7].

The most common etiology of MMH is due to overuse of the jaws due to clenching, bruxism, constant chewing or temporomandibular joint disorder [5, 7]. As a result, MMH has an insidious development over a number of years. Unilateral cases have been diagnosed in patients who clench ipsilaterally or have dental caries contralaterally [8]. Some literature from Asia documents a rare congenital variant of MMH affecting children and adolescents [7, 9].

Most cases of MMH are diagnosed from clinical presentation and imaging after concerns regarding facial asymmetry and the resulting cosmetic defect. Patients often present without functional deficits or symptoms, though they can present with facet wear suggesting bruxism or clenching [8]. CT or MRI imaging studies are useful in order to rule out other masseteric masses and pathology as well as identify MMH [6].

The symptoms of the patients seen in our clinic resulted in a full workup for common causes of sialadenitis and the use of salivary endoscopy for diagnosis. This was a unique opportunity, as this procedure has never been documented for use in the diagnosis or treatment of MMH. With respect to the patients who presented, only a handful of reported MMH cases have been linked to chronic obstructive salivary disease. Some of these cases presented with noted facial swelling or pain, but there was no mention of facial tenderness, salivary discharge, or the other symptoms seen in the patients at our clinic.

The unique presentation of the 2 adult patients with obstructive salivary disease due to MMH can be considered uncommon due to both infrequent diagnosis of MMH and limited presentations with obstructive salivary disease. More investigation would be needed to identify predisposing factors of patients presenting with obstructive parotitis secondary to MMH.

Treatment of MMH is commonly reserved for severe cases or cases with functional symptoms [5]. The standard approach is to use botulinum toxin type A injections of the masseter muscle to allow for muscle denervation and eventual atrophy. Repeat injections may be necessary [7]. Additional Botox injections can be administered to the parotid glands to limit salivary production for patients symptomatic with obstructive parotitis [10]. Simultaneously, the clenching or bruxism that may be a primary cause is often treated with an oral appliance. Some patients may require future treatment with partial resection of the hypertrophied masseter muscle. Long-term follow-up of these patients is often needed.

The presentation of the pediatric patient was also quite uncommon, as pediatric patients account for a small fraction of parotitis cases with the primary sources of parotitis often identified as mumps, juvenile recurrent parotitis or salivary stones [11–13]. There is currently no reported correlation or association between any of these conditions and MMH or fibrous dysplasia. Fibrous dysplasia is a rapid hamartomatous displacement of bone that can occur in pediatric patients. It can have various presentations, not limited to the bones of the head and neck region. Fibrous dysplasia more commonly involves the maxilla or mandible than other head and neck bones and is often visualized on imaging as ground glass, sclerosis or lytic destruction. The enlargement is typically benign, though it can be associated with a mass effect and accompanying symptoms of pain, cosmetic deformity or functional interference with nearby anatomical structures. These symptoms can necessitate treatment with surgical resection and follow-up [14–16].

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