Salivary Endoscopy for Idiopathic Chronic Sialadenitis

Rishi Vashishta, MD; M. Boyd Gillespie, MD, MSc

**Objectives/Hypothesis:** To describe the findings and therapeutic role of salivary endoscopy for idiopathic chronic sialadenitis.

**Study Design:** Retrospective case series.

**Methods:** The records of 258 patients who underwent salivary endoscopy between November 2008 and May 2012 were reviewed. Included cases presented with recurrent inflammation and swelling of a single major salivary gland, without identifiable etiology on examination and imaging.

**Results:** A total of 51 cases (20%) were identified. These patients had a mean age of 54.4 years (range, 23–75) and 57% were female. Mean duration of symptoms was 22.8 months (range, 1–102), with most cases (75%) involving the parotid gland. The primary imaging modalities used were ultrasound (62%) and computed tomography (31%). The most common findings on imaging included ductal dilation (42%), enlarged lymph nodes (23%), normal glandular imaging (15%), and possible sialoliths (14%). The most common findings on salivary endoscopy included stenosis (57%), strictures (27%), and inflammatory debris (18%). Occult stones were the cause of sialadenitis in only 4 (8%) cases. Outcomes included complete symptom resolution in 31 patients (61%), improved but occasional symptoms in 14 patients (27%), and no improvement in 6 patients (12%) after a mean follow-up time of 20.0 months (range, 4–45). Excision of the involved gland was required in 2 (4%) patients.

**Conclusion:** Salivary endoscopy is a minimally invasive technique that is effective in the management of idiopathic chronic sialadenitis refractory to medical therapy. It provides diagnostic information in most patients and offers a therapeutic intervention with gland preservation.

**Key Words:** Salivary endoscopy, sialendoscopy, salivary gland, obstruction, sialadenitis.

**Level of Evidence:** 4.

**INTRODUCTION**

Chronic sialadenitis is the most common benign disease of the major salivary glands, affecting approximately 1 in 20,000 of the general population. Chronic sialadenitis may result from blockage of the salivary ducts by stones, strictures, fibromucinous debris, anatomic anomalies in the ductal system, inflammatory polyps, or foreign bodies. Any of these factors may lead to impaired physiologic flow of saliva through the duct, resulting in salivary stasis and glandular inflammation. In addition, chronic sialadenitis may result from glandular inflammation and variable obstruction of the ducts from autoimmune, allergic, metabolic, and idiopathic causes. The disorder typically presents with painful swelling of the affected gland, most commonly during meals, and may be further complicated by an overlying bacterial infection causing mucopurulent discharge.

The initial treatment of chronic sialadenitis involves conservative measures designed to stimulate salivary flow, including adequate hydration, sialogogues, warm compresses, and massage of the affected gland. In addition, antibiotics and antiinflammatory medications may be required if a bacterial infection is suspected. However, in cases where conservative management fails, surgical excision of the salivary gland has historically been the mainstay of definitive treatment. Although it may provide permanent symptom resolution, surgical complications following sialadenectomy may result in a significant morbidity such as temporary facial paresis in 25% to 55% of patients, permanent facial nerve injury in 1% to 3% of patients, and Frey syndrome in 8% to 33% of patients undergoing parotidectomy for chronic sialadenitis. In the case of submandibular gland removal, complications include permanent injury to the marginal mandibular nerve in 1% to 8% of patients, hypoglossal nerve in 3% of patients, and lingual nerve in 2% of patients. Other potential complications such as salivary fistulas, sialoceles, hematomas, hypertrophic scars, cutaneous paresthesias, and wound infections may further complicate the postoperative course or impact quality of life following sialadenectomy.
Salivary endoscopy was developed in Europe in the 1990s to provide a minimally invasive approach for the exploration and treatment of sialolithiasis. The technique involves the use of miniature endoscopes and other microinstruments to visualize and treat diseases of the salivary ductal system. As the technology and instrumentation have advanced, salivary endoscopy is quickly emerging as the preferred method for diagnosing and managing salivary gland obstruction, thereby reducing the need for salivectomy and its associated risks.

While there have been a number of studies examining the role of salivary endoscopy for the management of chronic sialadenitis and sialolithiasis, only a single study to date has investigated its use in the setting of idiopathic chronic sialadenitis. The earlier European series found sialolithiasis to be a frequent (20%) cause of idiopathic sialadenitis, which differed from our observations in the North American population. The primary objective of this study was to describe the diagnostic and therapeutic roles of salivary endoscopy in the management of idiopathic obstructive sialadenitis of a single major salivary gland without identifiable etiology on examination and imaging, and to compare these findings to early studies. In addition, we present our long-term clinical outcomes in these patients following salivary endoscopy.

**MATERIALS AND METHODS**

**Patient Data and Statistical Analysis**

The medical records of 258 consecutive patients who underwent salivary endoscopy between November 2008 and May 2012 in the Department of Otolaryngology–Head and Neck Surgery at the Medical University of South Carolina (MUSC) were reviewed. Patients included presented with recurrent inflammation and swelling of a single major salivary gland, without identifiable etiology on both physical examination and radiographic imaging. Patients were excluded if they satisfied any of the following criteria: age < 18 years; prior history of extrusion or removal of stone or foreign body; history of major salivary gland surgery or trauma; prior radioiodine treatment or radiation therapy to the head and neck; history or laboratory findings of autoimmune disorder; or involvement of multiple glands. Data were collected from a department quality assurance database on patient demographics and clinical characteristics, including age, gender, gland involved, presenting symptoms, medical history, preoperative imaging studies, date of procedure, intraoperative findings and interventions, and complications from salivary endoscopy. Long-term outcomes were obtained from either the last clinic visit or follow-up surveys via mail. The study was approved by the MUSC institutional review board (IRB).

Statistical analysis was performed with Sigma Stat 3.5, SPSS 21.0, Sample Power 2.0, and Sigma Plot 10.0 (IBM Corporation, Armonk, NY). Disease information and demographic variables such as age, ethnic group, and sex were summarized by means of summary statistics. Fisher’s exact test was used to compare proportions from independent populations.

**Sialendoscopy Technique**

All salivary endoscopies were performed by a single surgeon (M.B.G.) in an outpatient surgical center operating room. Patients were consented for endoscopic-assisted transoral salivary surgery, with possible conversion to open transcervical surgery if the procedure could not be successfully performed via the transoral route. All patients were placed under general anesthesia with transoral intubation (parotid gland) or nasotracheal intubation (submandibular gland) as indicated. Antibiotics and steroids were administered intravenously prior to the procedure. The surgical technique involved visualization and serial dilation of the parotid or submandibular ostia with a series of salivary duct probes, followed by wider dilation with a tapered salivary duct dilator. A 0.8 mm diagnostic sialendoscope (Karl Storz, Culver City, CA) was used to make an initial pass, with larger (1.1 mm diameter; 1.6 mm diameter) endoscopes with instrument channels inserted as needed. The ductal lumen from the salivary ostia to the first and second order ducts were examined thoroughly to identify possible sources of obstruction.

Additional endoscopic-assisted interventions were performed as indicated based on the findings of the initial diagnostic endoscopy. Dilation of the salivary duct was performed in cases involving narrowing of the lumen due to stenosis (circumferential lengthwise narrowing of the main duct < 1.1 mm, as confirmed by inability to advance a 1.1 mm endoscope) or strictures (focal short segment scar bands extending across lumen).

The definition of a stenotic segment as < 1.1 mm is based on earlier anatomical studies that showed the mean diameter of a dilated Wharton’s and Stensen’s ducts to be greater than 1.2 mm. In case of stenosis, a guidewire (Karl Storz, Culver City, CA) was passed through the stenotic ductal segment through the working channel of a 1.1 mm endoscope. The ductal segment was then progressively dilated over the guidewire with a series of silastic bougies sized 4, 5, 6, and 7 French (Cook, Bloomington, IN). In cases of ductal stricture, the intraluminal scar or web was perforated with a micro hand drill, followed by passage of the guidewire and dilation with bougies. Steroid irrigations were performed (5 ml of 10 mg/ml triamcinolone acetonide) through a 23-gauge angiocatheter or through the Cook bougie for irrigation of the ductal epithelium or following ductal dilation. Salivary stents (4.0 French; Sialotechnology, Ashkelon, Israel) were placed in cases of stenosis or stricture of the ostium, or if an iatrogenic perforation was made in the distal main duct. Small salivary stones were captured with a wire basket and removed through the salivary ostium. Biopsy of a salivary gland was performed as an additional diagnostic modality in patients with complaints of xerostomia and xerophthalmia and/or intraglandular adenopathy on imaging but previous negative Sjögren’s syndrome serology. Botox injections (100 units botulinum toxin type A in 2 ml saline over 3 sites) were administered via ultrasound guided needle placement for temporary symptomatic relief in cases of stenosis/stricture that were too proximal (intraglandular) in location to be treated with endoscopic intervention. The expected period of relief from Botox is estimated to be 3 to 4 months with reduction in effectiveness after this time.

**RESULTS**

**Patient Demographics**

A total of 51 patients presented with idiopathic chronic sialadenitis of a single major salivary gland, representing 20% of all patients treated with endoscopic-assisted management of salivary gland obstructions between November 2008 and May 2012. The patients had a mean age of 54.4 years (range, 23–75), and included 29 females (57%) and 22 (43%) males.

Moderate to heavy caffeine use (≥ 16 ounces of coffee per day) was noted by 26 (57%) patients. Only eight
(16%) patients were current smokers. Twenty-two patients (43%) were currently taking one (19 patients) or more drugs (3 patients), which may contribute to reduced salivary output. Interestingly, only two (9%) of these patients had the sensation of having xerostomia. The principal drug classes were diuretics (13 patients), namely hydrochlorothiazide; or antidepressants (12 patients), most commonly selective serotonin-reuptake inhibitors.

The mean duration of symptoms was 22.8 months (range, 1–102). The parotid gland was involved in 38 (75%) of cases and submandibular gland in 13 (25%) of cases. The majority (89%) of patients reported their symptoms as being intermittent and about half (48%) of patients reported worse swelling during meals. The most common symptoms experienced by patients were pain (93%), foul taste or discharge (58%), and dry mouth (25%). None of the 23 (45%) patients previously tested had positive Sjögren’s serologies (SSA, SSB). Antibiotic therapy had been used in 82.4% of patients prior to presentation with incomplete resolution of symptoms.

**Diagnostic Radiography**

Preoperative diagnostic imaging evaluation was performed on all patients. The primary modalities included ultrasonography in 61% of patients and computed tomography (CT) in 31% of patients. Two (4%) patients underwent magnetic resonance imaging (MRI), and another two (4%) patients received sialography.

The most common finding on diagnostic imaging was ductal dilation which was observed in 24 (47%) patients (Fig. 1). Eleven (22%) patients had normal glandular imaging. Other radiographic findings included enlarged intraglandular lymph nodes (20%), possible but not obvious sialoliths (20%), enlarged salivary gland (8%), inflammation of the salivary gland (8%)

**Endoscopic Findings**

All patients underwent diagnostic salivary endoscopy, with stenosis (duct < 1.1 mm greatest diameter) being the most common finding in 30 (59%) patients (Fig. 2). Stricture (scar tissue/webs across lumen) was present in 17 (33%) patients and inflammatory debris (mucous plugs) with sialodochitis (duct wall inflammation) was present in 8 (16%) patients. Occult stones were found to be the source of obstruction in 4 (8%) cases. Anatomic anomaly (ductal duplication) with stenosis was observed in one patient (2%). Nine patients (18%) had multiple abnormal findings. No patients had a normal examination. The anatomic locations of the obstruction in order of frequency were the main duct (41%), intraglandular sites...
(second- and third-order ducts) (27%), hilum (18%), and salivary ostia (18%). Two patients (4%) had obstruction at two or more anatomic locations.

Endoscopic stone retrieval with basket was successful in the four cases of occult stone and eight cases of mucous plugs. Ductal dilation with a salivary bougie over a guidewire was performed in 40 (78%) patients. Steroid infusion of the ductal lumen was administered in 38 (75%) of patients with stenting of five (10%) patients. Botulinum toxin A (Botox) injection was performed in 16 (31%) patients (4 parotid glands and 12 minor glands) as an additional diagnostic modality in patients with complaints of xerostomia and xerophthalmia and/or intraglandular adenopathy on imaging but negative Sjögren’s serology. Five of the 16 (31%) biopsies showed lymphocyte infiltration of the salivary glands consistent with Sjögren’s syndrome.

Complications
There were two (3.6%) patients who experienced complications, both with perforation of the distal main Stensen’s duct just beyond the ostium. Both of these patients underwent placement of a salivary stent (4.0 French, Sialotechnology, Ashkelon, Israel), which bridged the perforation and was secured to the buccal mucosa with a 4.0 vicryl stitch. The stents were left in place for 2 weeks and then removed. Both patients reported resolution of symptoms without further complication. Salivary endoscopy was limited in one (2%) patient due to severely structured salivary ostia.

Outcomes
Patient outcomes were assessed at last follow-up visit or via mailed questionnaire after a mean follow-up time of 20 months (range, 4–45). A total of 31 (61%) patients reported complete symptom resolution after either a single (30 patients) or second endoscopy (1 patient). Fourteen (27%) patients reported improvement in symptoms with less severe and less frequent glandular obstruction after a single (10 patients) endoscopy or a second endoscopy (4 patients). Six (12%) patients experienced no improvement after endoscopy. Two patients (4%) without improvement elected to undergo gland excision.

DISCUSSION
Chronic sialadenitis is an infrequent, but not rare, salivary disorder characterized by glandular swelling and pain that often flares during mealtime or with intake of sour or acidic foods. Salivary stones are the most common source of chronic sialadenitis (80% of cases) and are usually readily identified on routine imaging with either ultrasonography (US) or CT. In the absence of an identifiable stone, imaging may reveal signs suggestive of obstruction such as ductal dilation or glandular enlargement from congestion, as seen in just over half of the patients in this study. However, additional specific causes such as ductal stenoses, strictures, fibromucinous plugs, ductal anomalies, foreign bodies, and small stones (≤2 mm) frequently fall below the detection threshold of US and CT. Although sialography has the potential to diagnose ductal stenoses and strictures, it is used infrequently due to the lack of skilled personnel, concern over contrast reactions, radiation exposure, patient discomfort, and ready access to less invasive imaging modalities.

Direct inspection of the salivary ducts with salivary endoscopy provides a method to identify the cause of obstruction in real time in the majority of cases of idiopathic salivary gland obstruction. Although Koch et al. noted normal architecture in 11 of 103 (11%) glands with swelling of unclear etiology, the present series noted at least one pathologic finding in 100% of inspected glands (Table I). When comparing our findings to that of Koch et al., the North American population in this study was significantly more likely to present with idiopathic obstruction and have parotid gland involvement. In addition, these patients had a significantly higher rate of stenosis and stricture on endoscopic examination and fewer occult stones. Reasons for this may include a higher rate of stone pathology overall in the German population, which is suggested by the greater number of submandibular glands that are more prone to stone formation. North American surgeons, therefore, need to have equipment and tools that allow for the effective management of strictures and stenoses available at the time of endoscopy for idiopathic chronic sialadenitis. Equipment that may be required includes endoscopes of various diameters, endoscopic hand drills, guidewires, tapered dilators, endoscopic balloons, micro forceps, laser microfibers, and salivary stents.

<table>
<thead>
<tr>
<th>Study Findings</th>
<th>University of Erlangen</th>
<th>MUSC</th>
<th>P value&lt;sup&gt;*&lt;/sup&gt;</th>
</tr>
</thead>
<tbody>
<tr>
<td>Patients with idiopathic obstruction</td>
<td>103/1900 (5.4%)</td>
<td>51/258 (20%)</td>
<td>0.0001</td>
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<tr>
<td>Female</td>
<td>54 (52.4%)</td>
<td>28 (57%)</td>
<td>N.S.</td>
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<tr>
<td>Mean age (years)</td>
<td>49.5</td>
<td>54.4</td>
<td>N.S.</td>
</tr>
<tr>
<td>Parotid gland</td>
<td>50 (48.5%)</td>
<td>38 (75%)</td>
<td>0.003</td>
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<tr>
<td>Submandibular gland</td>
<td>53 (51.5%)</td>
<td>13 (25%)</td>
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<td>Endoscopic Findings</td>
<td></td>
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<tr>
<td>Stenosis/stricture</td>
<td>58 (56.3%)</td>
<td>47 (92%)</td>
<td>0.0001</td>
</tr>
<tr>
<td>Occult stones</td>
<td>21 (20.3%)</td>
<td>4 (7.8%)</td>
<td>0.06</td>
</tr>
<tr>
<td>Inflammatory Debris/sialodochitis</td>
<td>11 (10.7%)</td>
<td>8 (15.7%)</td>
<td>N.S.</td>
</tr>
<tr>
<td>Ductal anomaly</td>
<td>4 (3.9%)</td>
<td>1 (2.0%)</td>
<td>N.S.</td>
</tr>
<tr>
<td>Normal anatomy</td>
<td>11 (10.7%)</td>
<td>0</td>
<td>0.016</td>
</tr>
<tr>
<td>Outcomes</td>
<td></td>
<td></td>
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<tr>
<td>Symptoms improved/resolved</td>
<td>93 (89.3%)</td>
<td>45 (88.2%)</td>
<td>N.S.</td>
</tr>
<tr>
<td>Gland excision</td>
<td>2 (2%)</td>
<td>2 (4%)</td>
<td>N.S.</td>
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*Fischer’s Exact Test.
MUSC = Medical University of South Carolina; N.S. = nonsignificant.
Although salivary endoscopy helps to determine whether there is an anatomic cause of the inflammation, the underlying etiology of sialadenitis is often not evident. In a recent retrospective study of 153 patients, the authors observed a higher prevalence of smoking (44%) and diuretic use (20%) in patients with salivary stones compared to the general population, thereby offering support to the theory that stones largely arise in the setting of oral dryness. The underlying cause of obstruction in the 47 patients without stones in this study is less clear. Many of the patients had daily exposures to substances that lead to dry mouth such as moderate caffeine consumption (57%), drying medications (43%), and tobacco (16%), but it is unknown whether this predisposes to ductal stenosis or stricture. Future epidemiological studies of patient populations taking diuretics or antidepressants may help to determine whether these patient populations are at greater risk of chronic sialadenitis.

Additional causes of ductal stenosis and stricture include prior stones that have passed, previous salivary gland infection (e.g., mumps), congenitally small ducts, chronic ductal reflux, or an underlying inflammatory (e.g., autoimmune) condition. Five patients (10%) in the study were found to have lymphocytic infiltration of minor salivary tissue consistent with Sjögren’s on biopsy, although all had previously negative Sjögren’s serologies. This finding indicates that Sjögren’s syndrome is a potential underlying cause of at least a minority of cases of idiopathic chronic sialadenitis. Patients who meet diagnostic criteria for Sjögren’s syndrome have relatively advanced disease, having had symptoms for a mean of 5.2 years prior to diagnosis. With the advent of salivary endoscopy, many patients are seeking treatment of chronic glandular inflammation of shorter duration, and may therefore have negative serologies due to their presentation at an earlier stage of disease. In general, open biopsy of minor salivary gland tissue is considered a more sensitive test for Sjögren’s syndrome and may detect cases earlier in the course of the disorder.

Our current practice is to perform minor salivary gland biopsy in cases of idiopathic chronic sialadenitis with negative Sjögren’s serologies if the patient has complaints of xerostomia and xerophthalmia and/or intraglandular adenopathy on imaging. This practice is supported by the relatively high rate (31%) of positive findings in our sample of patients who underwent salivary biopsy.

Once the diagnosis is established, salivary endoscopy allows potential therapeutic management of obstructive sites with microinstruments passed directly through the endoscope. All but two patients (49/51; 96%) underwent endoscopic therapy via basket retrieval of a stone or mucous plug, and/or ductal dilation and steroid infusion. One of these patients had severe stenosis of the parotid ostium only allowing the tip of the endoscope, whereas the other patient had stricture of a very small third-order intraglandular duct. Both of these patients underwent ultrasound-guided injection of botulinum toxin, which successfully reduced glandular swelling and inflammation. Therapeutic salivary endoscopy had excellent midterm results, with 88% of patients noting resolution or significant improvement in symptoms after a mean follow-up time of 20 months and only two patients (4%) requesting salivary gland excision to date, which compares favorably to the previous study (Table 1).

CONCLUSION

The absence of stones on imaging is a relatively common finding in patients with chronic sialadenitis. Salivary endoscopy can assist in determining the anatomic cause of swelling in idiopathic cases, although the underlying pathophysiologic cause remains unclear in the majority of cases. Salivary endoscopy assists in therapeutic interventions that lead to symptom control and gland preservation in the vast majority of patients. Further study is needed to elucidate the underlying mechanisms of nonstone-related chronic sialadenitis, and to determine if early intervention with salivary endoscopy can improve longer term outcomes.

BIBLIOGRAPHY