An Extraoral Surgical Approach to Treat Chronic Submandibular Sialolithiasis - A Case Series

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Introduction
Sialadenitis is an inflammation of the major salivary glands and its etiology can include infections or noninfectious causes. All the inflammatory processes that affect salivary glands, such as pain, increased gland volume, and reduced salivary flow, have been described.\(^1\) The main agents associated with salivary gland infections are viruses such as mumps, Coxsackie A, echovirus, choriomeningitis, parainfluenzae, and Cytomegalovirus, and others of bacterial origin. In many cases, Staphylococcus aureus is the most common causative agent of acute infections, and may be associated with Streptococcus and other agents.

Consequently, for an acute bacterial infection to occur, stenosis or sialolithiasis would be the factor that interrupt the normal salivary flow, and allow contamination in the microenvironment of the salivary duct system.

Salivary stasis or decreased salivary flow is considered the main factor in the development of sialoliths. Other risk factors are reduced fluid intake, dehydration of any origin, smoking, prolonged illnesses, diuretics, and antihypertensive drugs that reduce salivary flow.\(^2\)

Sialolithiasis is the most common cause of sialadenitis in the submandibular gland, in which the highest incidence of this condition occurs, among the major salivary glands. This could be explained by the anatomy of Wharton’s duct, and the chemical composition of the saliva produced by this gland. There are several alternatives and techniques for the treatment of sialolithiasis, including lithotripsy, sialoendoscopy, and conservative removal of the sialoliths or complete removal of the submandibular gland, through the transoral and extra oral routes for access to the gland. To determine the form of treatment, characteristics such as topography, diameter, and location of the sialolith in the duct are observed. The aim of this case series was to show our experience gained in two clinical cases of submandibular gland excision through an extraoral approach, using the submandibular access technique. In addition, we discussed the cause of sialolithiasis in these patients and after follow-up, compared the clinical results we obtained with this technique with those reported in the current literature. The submandibular approach or Risdon access continues to be a safe approach to removing the submandibular gland, as it is a commonly used technique and obtained satisfactory results, as shown in these cases. However, the major disadvantages were the less favorable esthetic results and paralysis of the marginal mandibular branch of the facial nerve.

Keywords: Salivary glands, sialadenitis, sialolithiasis, submandibular glands

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Received: 02-04-2020 Revised: 25-04-2020
Accepted: 28-06-2020 Published: 23-12-2020
close to the hilum, with a rate ranging between 1:30,000 and 1:10,000 annually.\[2,3\]

In general, patients suffering from sialolithiasis have a painful lesion sensitive to palpation and radiating pain that could begin in the retromolar region and spread to the jaw angle, whereas several other patients have manifested an increase in volume in the same region accompanied by decreased salivary flow. This history of pain or/and swelling in the salivary glands, especially during meals, suggests this diagnosis.\[4\]

The submandibular gland is the salivary tissue most frequently affected and removed due to this inflammatory process.\[5\] According to gender and age data, the male population, aged between 40 and 60 years, showed high incidence.\[6\]

The case report describes a 69-year-old male patient who presented at the Campo Limpo Medical Center, Sao Paulo, Brazil, with complaints of pain in the submandibular region as shown in Figure 6. The submandibular gland was reached. Blunt dissection was performed superiorly and medially around the gland, making it possible to identify the mylohyoid muscle. Finally, the submandibular gland containing an intraparenchymal sialolith measuring approximately 22.1 mm × 18.7 mm was removed, as shown in CT scans, and the duct was linked to its posterior portion. Primary wound closure was performed according to the anatomical layers, including the fascia and platysma, subcutaneous tissue, and skin closure, as shown in Figure 4.

**Case 2**

A 51-year-old male patient presented to the Oral and Maxillofacial Surgery Outpatient Clinic, Campo Limpo Medical Center, Sao Paulo, Brazil, with complaints of pain in the left submandibular area, with onset 1 week ago. During oral clinical examination, the patient experienced swelling and pain that increased on palpation. The patient reported that pain increased when he ate solid and acid foods. Local swelling is visualized in Figure 5.

CT assessment was performed for the purpose of establishing the extent of the lesion inside the gland and to determine its volume. Axial, coronal, and sagittal sections showed a circumscribed, radiopaque hyperattenuating area, localized in the submandibular gland region as shown in Figure 6.

As previously mentioned in the description of the extraoral surgical approach in Case 1, this was performed in Case 2 under GA, as shown in Figure 7a and b. An intraparenchymal sialolith measuring 15 mm × 10 mm was removed. After surgery, the postoperative care resulted in good improvement by reducing pain and edema so that the patient was discharged from the hospital on the 2nd day of hospitalization. On return after 7 days, he complained of mild pain in the left submandibular region and edema related to the surgical approach; however, the surgical wound was in the process of repair, as shown in Figure 7c and d. The patient showed facial mimicry with slight deficit of facial motor control; normal tongue movements and sensitivity were recorded after 2 months of follow-up.
**Figure 1:** Profile of the submental vertex, in which well-circumscribed increase in volume in the right submandibular area may be observed.

**Figure 2:** Computed tomography sections showing well-circumscribed area (hyperattenuating) close to the submandibular gland. (a) Axial, (b) coronal, (c) sagittal.

**Figure 3:** Three-dimensional imaging reconstruction showing the size of the sialoliths that were found to be 22.1 mm × 18.7 mm.

**Figure 4:** Extraoral surgical management: (a) Risdon approach, (b) submandibular gland removed, (c) sialoliths included in parenchyma of the gland were completely removed, (d) follow-up after 2 months showing evidence of scar at the surgical site.

**Figure 5:** Local swelling in the left side of the submandibular area.

**Discussion**

Chronic sialadenitis may be caused by sialoliths, mucous plugs, ductal stenosis, kinks in the salivary ducts, infections, immune-mediated or autoimmune diseases, radioiodine therapy, radiotherapy, and rarely duct compression by tumor.[8]

Sialolithiasis is characterized by the obstruction of a salivary gland or its excretory duct, due to the formation of calcified mass-denominated stones or sialoliths.[6]

The etiology of sialolith formation is unknown; however, the authors have claimed that stones are formed by the deposition of calcium salts around an organic matrix composed of mucin, altered by bacterial infection and squamous cell remnants. However, the etiology of sialolithiasis remains unclear; a
A retrospective review study showed that smoking and serum electrolytes, such as sodium and potassium, were associated with larger salivary stone development because of their capacity to contribute to decreased salivary flow and to reflect volume depletion, respectively.\[9\]

During extraoral examination, the submandibular gland is usually not noticed, but when enlarged, it is the first clinical sign that manifests as a palpable mass in the submandibular region.\[10\] Frequently, the clinical manifestations of inflammatory diseases are pain, fever, purulent secretion through the Wharton duct, and rapid growth.\[2,3,10\] In our case series, both patients showed a similar clinical condition with a well-defined increase in volume in the submandibular region, which was painful on palpation, with exacerbation of pain while feeding.

Among auxiliary examinations performed to confirm the presence of sialoliths, ultrasound scanning is widely used. Detection by CT of the submandibular glands is highly accurate. However, sialography has been less frequently indicated up to now.\[2\] Nowadays, magnetic resonance imaging using contrast is the gold standard examination for the identification and localization of salivary gland sialoliths.\[2,11\] Moreover, sialoendoscopy, a minimally invasive technique for the treatment of obstructive sialadenitis that is more widely used by otorhinolaryngologists, offers direct visualization of the ducts and direct access to these stones. There are other less invasive treatments, such as lithotripsy,\[7\] which was used in a recent study for the treatment of sialoliths in the parenchyma and hilum regions and showed favorable results. However, when used for removing sialoliths over 8 mm in diameter, lithotripsy showed poor results and required the use of a transcervical approach. In both cases treated with submandibulectomy, the sialoliths were larger than 8 mm in size.\[3\] Other current retrospective case series\[12\] found no differences between two different approaches (in-office versus operating room sialoendoscopy) for managing inflammatory salivary gland diseases, with regard to improving the symptoms and reducing the rates of recurrence. Similar results have previously been shown with the preservation of a high number of glands after sialoendoscopy performed without requiring any additional procedures.\[13\]

The main nonneoplastic submandibular salivary gland diseases that justify surgical management are the pleomorphic adenoma (PA) and chronic sialadenitis associated with the presence of sialoliths.\[5,10,14\]

With regard to this case series, Risdon access was used to remove the submandibular gland, by making an incision 1.5–2 cm from the lower edge of the mandible, to avoid the risk of injury to the marginal mandibular branch of the facial nerve. The deep layers were exposed until the capsule covering the submandibular gland was reached. Thus, ligation of the facial artery, vein, and Wharton’s duct and removal of the gland were performed; sutures were placed by planes; and, finally, skin closure was performed.

Other studies have stated that the extraoral approach to removal of the submandibular gland was relatively simple and widely accepted; however, new approaches, such as transoral access, have been developed. This therapeutic modality is usually performed with a fibrescope or endoscope, and the practitioner must be familiar with this technique because it is more difficult to remove or link important structures such as vessels and nerves by means of this management technique.\[15\]

In 2001, Zenk et al. conducted a case series study on 231 patients, for the removal of sialoliths from the submandibular gland and Wharton’s duct, by using a transoral technique.\[16\] However, unlike the studies conducted by Hong
and Kim,[11] who removed the gland through the transoral access, Zenk et al. used the transoral access to remove the sialolith, without the use of more restricted devices such as the fiberscope. The most important aspect of Zenk et al.’s study was that although the sialolith had been located in the hilum or glandular parenchyma, it was removed through the transoral access, preserving the salivary gland. In 115 patients, the sialolith was distal to the posterior border of the mylohyoid muscle; in 6% of these cases (14 patients), fragmentation of the sialolith occurred, making it impossible to remove the fragments through the transoral access. Furthermore, in three of the patients, the procedure was performed through the extraoral access.

In a retrospective study conducted at the University of Tokyo, with 133 patients submitted to removal of the submandibular gland affected by sialolithiasis, PA, and other benign neoplasms, a group of 87 patients showed irreversible glandular inflammatory disease. Moreover, after performing the transcervical approach, clinical complications such as paralysis of the marginal branch of the mandible, paresthesia of the lingual nerve, and paralysis of the hypoglossal nerve[17] appeared after postoperative follow-up, however, they were found to be transitory. In two cases of patients operated in this study, paralysis of marginal nerve did not exceed 2 weeks; moreover, there were no cases of hypoglossal nerve paralysis or lingual nerve paresthesia.

At present, the biggest disadvantage of extraoral access is relative to the healing process and remnant scar in the region of the skin incision, which can often be minimized by parallel relaxing incisions or in relaxed tension lines of the skin.[14]

**Conclusion**

In spite of esthetic considerations and possible functional complications, an extraoral surgical approach to removing the submandibular gland with sialolithiasis in the parenchyma, continues to be used by several oral and maxillofacial surgery departments in Brazilian domains. These procedures have shown satisfactory clinical results, and are simple for oral and maxillofacial surgeons to perform, as shown in our case series.

When this management was compared with other surgical techniques, Risdon’s access showed a major esthetic disadvantage relative to the skin scar observed in one case. Furthermore, we encourage further prospective studies to be conducted to assess the multimodal approach and to elucidate the role played by each procedure in reducing the esthetic and functional complications.

**Acknowledgment**

The authors thank the Department of Oral and Maxillofacial Department, Campo Limpo Medical Center, Sao Paulo, Brazil (BR), including the technical team and oral and maxillofacial surgeons Bruno da Silva Mesquita and Lisana Massaine, who participated in the work at the outpatient clinic and supported this work.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patients have given their consent for their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published, and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

**Financial support and sponsorship**

The author Wilber Edison Bernaola-Paredes is student fellow and supported with an scholarship by the National Council for Scientific and Technological Development of Brazil (CNPq) (140071/2019-9).

**Conflicts of interest**

There are no conflicts of interest.

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