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

Renan Veiga Araújo, Basílio Almeida Milani, [1]Ivan Solani Martins, [2]Eleazar Mezaiko Vilela Dias, [3]Wilber Edison Bernaola-Paredes

Department of Oral and Maxillofacial Surgery, Campo Limpo Medical Center, [1]Department of Oral and Maxillofacial Surgery, Hospital Sirio-Libanes, Sao Paulo, [2]School of Dentistry, University Federal de Goias, Goias, Brazil, [3]Department of Radiation Oncology, A.C. Camargo Cancer Center, Sao Paulo, Brazil

Abstract

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 extraoral 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

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, with an estimated prevalence of 1.2% in the general population. Among symptomatic sialoliths, 80% are found in the submandibular gland and over 50% of these are included in the hilum or

Address for correspondence: Prof. Wilber Edison Bernaola-Paredes, Department of Radiation Oncology, A.C. Camargo Cancer Center, Sao Paulo, Brazil.
E-mail: edison.bernaola@gmail.com

Received: 02-04-2020 **Revised:** 25-04-2020
Accepted: 28-06-2020 **Published:** 23-12-2020

Access this article online

Quick Response Code:

Website: www.amsjournal.com

DOI: 10.4103/ams.ams_102_20

How to cite this article: Araújo RV, Milani BA, Martins IS, Vilela Dias EM, Bernaola-Paredes WE. An extraoral surgical approach to treat chronic submandibular sialolithiasis - A case series. Ann Maxillofac Surg 2020;10:537-42.

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 long, tortuous, ascending duct of the submandibular gland (Wharton), and its thick, mucoid secretion, to a large extent, contributes to the formation of sialoliths. Moreover, the abovementioned duct is 3–4 mm in diameter and is approximately 50 mm long.[7] The submandibular duct originates from the distal portion of the parenchyma and curves under the posterior border of the mylohyoid muscle, and its path continues under the lingual nerve in the posterior region of the floor of the mouth.[3]

The extraoral surgical approach continues to be a technique used in the treatment of sialolithiasis; however, its use involves esthetic and functional limitations. A discussion about its applicability may help oral and maxillofacial surgeons undergoing training to clarify doubts about this relevant issue.

The aim of the present case series was to describe our experience with these two cases of sialolithiasis in the submandibular gland, treated surgically through the extraoral approach, and to compare our clinical results after the surgery performed, with those found in the current literature.

Case Reports

Case 1

A 69-year-old male patient presented at the Campo Limpo Medical Center with a complaint of pain in the right submandibular region. During clinical assessment, we found an increase in volume in the region of the right submandibular space, with pain on palpation in the area as shown in Figure 1.

According to the patient's medical records, his preoperative assessment classification was American Society of Anesthesiologists – Physical Status III due to his history of High Arterial Blood Pressure (HABP), cardiomyopathies, and diabetes mellitus Type II.

In the axial, coronal, and sagittal sections of computed tomography (CT) images, a well-circumscribed, high-density area was observed, localized in the region of the submandibular trigone, as visualized in Figure 2.

Moreover, three-dimensional imaging reconstruction was performed in order to visualize the size of the sialoliths that were found to be 22.1 mm × 18.7 mm, as shown in Figure 3.

Surgical treatment was performed under general anesthesia (GA), based on the excision of the sialolith from the glandular parenchyma of the submandibular gland. Thus, the incisional or Risdon approach was used, with the incision being made 2 cm below the lower edge of the mandible to protect the mandibular marginal nerve (facial motor nerve branch), and extending it to approximately 4 cm from the angle of the mandible, in the direction from the posterior to anterior region.

After surgical incision, the deep layers, including subcutaneous tissue, platysma muscle, and superficial layer of the deep cervical fascia, were exposed, until the capsule covering 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

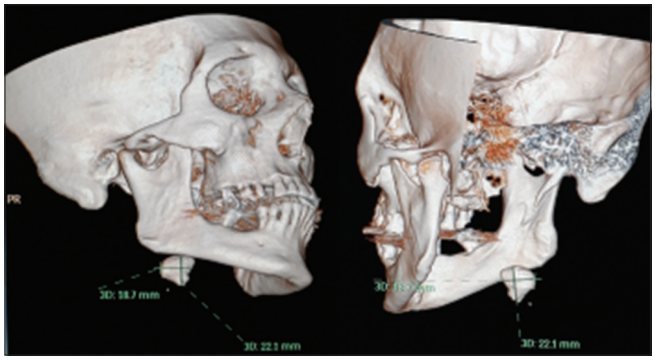

Figure 3: Three-dimensional imaging reconstruction showing the size of the sialoliths that were found to be 22.1 mm × 18.7 mm

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]

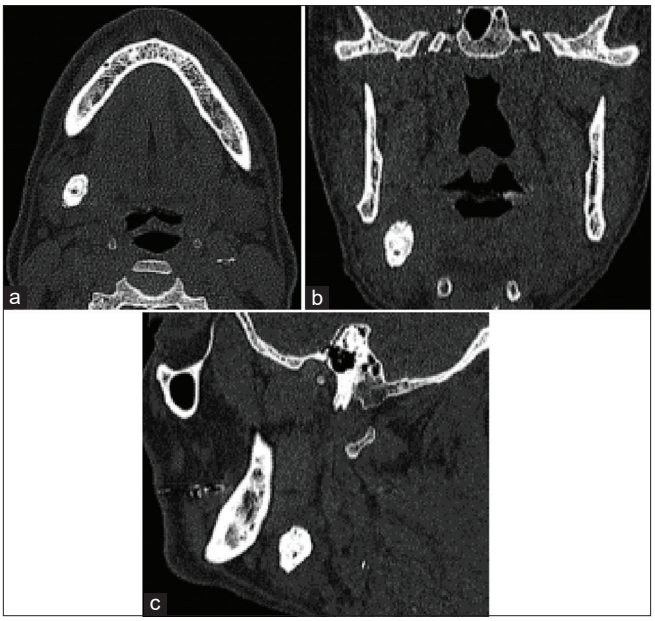

Figure 2: Computed tomography sections showing well-circumscribed area (hyperattenuating) close to the submandibular gland. (a) Axial, (b) coronal, (c) sagittal

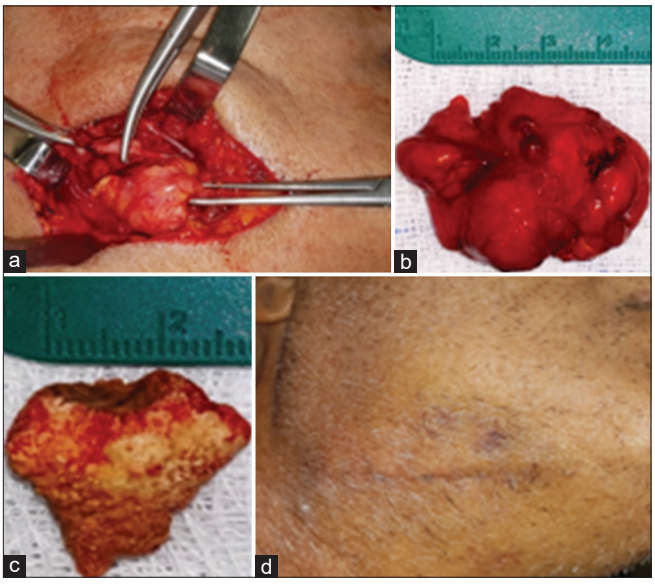

Figure 4: Extraoral surgical management: (a) Risdon approach, (b) submandibular gland removed, (c) sialoliths included in parenchyma of the gland were completely removed, (d) follow-up after 2 months showing evidence of scar at the surgical site

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

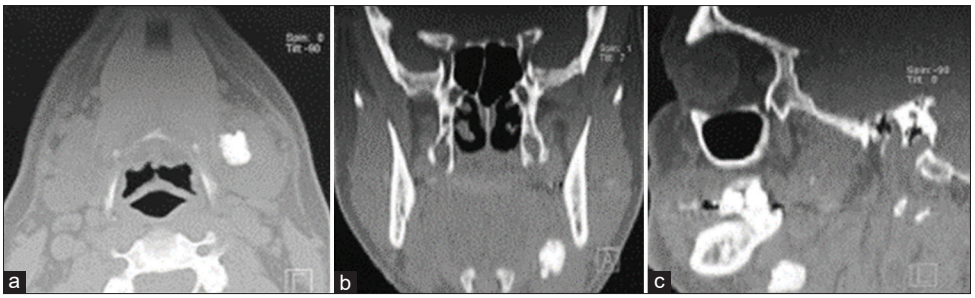

Figure 6: Computed tomography assessment showing hyperattenuating area surrounding the left submandibular gland: (a) Axial section, (b) Coronal section, and (c) Sagittal section

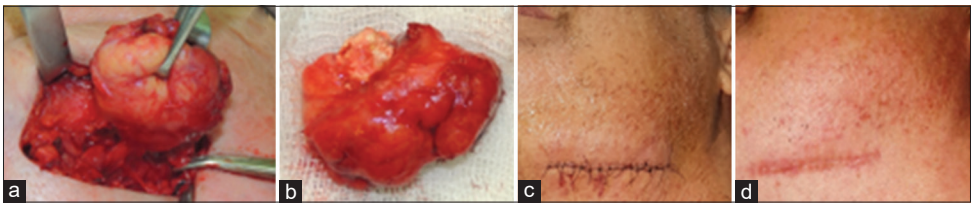

Figure 7: (a) Extraoral surgical removal of the left submandibular gland, (b) Sialoliths placed at the center of glandular parenchyma, (c) Clinical aspect after 7 days of postoperative evaluation, (d) Follow-up of 2 months after extraoral surgical approach; it was possible to visualize a fibrous scar

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 clinic 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 fiberscope 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.

References

1. Gomes RT, Naves M, Carmo M, Aguiar M. Sialadenitis: A review of literature about etiology, diagnosis and treatment. Odont Arquives 2006;42:257-336.
2. Binar M, Gokgoz MC, Aydin U, Yavan I, Karahatay S. Chronic sialadenitis due to the stone inside the accessory duct of submandibular gland. Surg Radiol Anat 2017;39:1165-8.
3. Schapher M, Mantsopoulos K, Messbacher ME, Iro H, Koch M. Transoral submandibulectomy for deep hilar submandibular gland sialolithiasis. Laryngoscope 2017;127:2038-44.
4. Pachisia S, Mandal G, Sahu S, Ghosh S. Submandibular sialolithiasis: A series of three case reports with review of literature. Clin Pract 2019;9:1119.
5. Vieira MB, Pereira RA, Santos Júnior NB. Subcapsular resection of submandibular salivary gland. Rev Bras Otorrinolaringol 2001;67:825-8.
6. Lima de AN, Milani B de A, Massaine LF, Souza de AM, Jorge WA. Sialolithiasis in submandibular salivary gland: a case report. Brazilian Journal of Oral and Maxillofacial Surgery 2013;13:23-8.
7. Foletti JM, Graillon N, Avignon S, Guyot L, Chossegros C. Salivary calculi removal by minimally invasive techniques: A decision tree based on the diameter of the calculi and their position in the excretory duct. J Oral Maxillofac Surg 2018;76:112-8.
8. Lele SJ, Hamiter M, Fourrier TL, Nathan CA. Sialendoscopy with intraductal steroid irrigation in patients with sialadenitis without sialoliths. Ear Nose Throat J 2019;98:291-4.
9. Yiu AJ, Kalejaiye A, Amdur RL, Todd Hesham HN, Bandyopadhyay BC. Association of serum electrolytes and smoking with salivary gland stone formation. Int J Oral Maxillofac Surg 2016;45:764-8.
10. de Carvalho AS, Dedivitis RA, de Castro MA, Nardi CE. Submandibular gland excision. Rev Col Bras Cir 2015;42:14-7.
11. Hong KH, Kim YK. Intraoral removal of the submandibular gland: A new surgical approach. Otolaryngol Head Neck Surg 2000;122:798-802.
12. Coniglio AJ, Deal AM, Bhate O, Hackman TG. In-office versus operating room sialendoscopy: Comparison of outcomes, patient time burden, and charge analysis. Otolaryngol Head Neck Surg 2019;160:255-60.
13. Achim V, Light TJ, Andersen PE. Gland preservation in patients undergoing sialoendoscopy. Otolaryngol Head Neck Surg 2017;157:53-7.
14. Sousa de RI, Santos dos MG, Oliveira JM de S, Mendonça VB de A, Pereira JV. Pleomorphic adenoma in the submandibular salivary gland: A case report and literature review. Brazilian Journal of Oral and Maxillofacial Surgery 2013;13:9-14.
15. Çukurova İ, Arslan İB, Bulğurcu S, Demirhan E. Transoral versus

transcervical approach to submandibular gland: Techniques and outcomes. Kulak Burun Bogaz Ihtis Derg 2015;25:319-23.
16. Zenk J, Constantinidis J, Al-Kadah B, Iro H. Transoral removal of submandibular stones. Arch Otolaryngol Head Neck Surg 2001;127:432-6.
17. Ichimura K, Nibu K, Tanaka T. Nerve paralysis after surgery in the submandibular triangle: Review of University of Tokyo Hospital experience. Head Neck 1997;19:48-53.