CASE REPORT

Sialolithiasis and submandibular gland excision: Report of cases

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Abstract

Sialolithiasis is the most common salivary gland disorder, and it consists in an inflammatory process related with a partial or total duct obstruction. The submandibular gland is the most affected one, representing 80% of the incidence rate. The treatment varies and depends on the location, number and size of the sialolith and the surgeon’s experience. In this report of two cases of sialadenitis due sialolithiasis, the sialoliths were located in the glandular’s parenchyma and posterior region of the Wharton’s duct and were treated by submandibular gland and sialolith excision using submandibular approach. Despite of the variety of techniques available to the treatment of sialolithiasis, the submandibular gland excision with the sialolith demonstrated to be a well-indicated and secure technique to treat sialadenitis due sialolithiasis, when the calculi is located in areas accessible by the intraoral approach and when the gland is not functional. The surgical technique is easy to perform, but careful is necessary not to damage important structures around to the submandibular gland.

Keywords

Oral pathology, oral surgery, sialolith, submandibular gland

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Introduction

Sialolithiasis is the most common salivary gland disorder, and it consists in an inflammatory process related with a partial or total duct obstruction. The submandibular gland is most affected, representing 80% of the incidence rate.¹² Salivary calculi are usually unilateral, and children are rarely affected. It is estimated that 12 in 1000 of adults are affected by this disorder.²³

The treatment varies and depends on the location, number and size of the sialolith and the surgeon’s experience. It can be done by extracorporeal shock wave lithotripsy, endoscopic intracorporeal shock wave lithotripsy, sialendoscopy, transoral CO₂ laser sialolithectomy, simple incision on the mucosa along the duct, electrohydraulic or pneumoblastic devices or in some cases, gland excision.²⁴

The present paper presents two cases of sialadenitis secondary to sialolithiasis in the submandibular gland treated by gland and calculi excision as well as discusses the indications for such therapeutical option.

Case Reports

Case 1

A 13-year-old, female patient presented to Oral and Maxillofacial Surgery Department of the General University Hospital of the University of Cuiaba with a complaint of a swelling below the mandible and tongue at the right side associated to an unpleasant feeling below the tongue when she ate acid or sour foods [Figure 1a]. Previous history revealed surgical treatment attempt to this condition elsewhere 3 years previously. Bimanual palpation revealed a firm swelling, approximately 3 cm in length in the posterior oral floor of
the mouth, which could be clinically observed to cause an asymmetry due to an augmentation at the submandibular right area. The overlying oral mucosa was healthy in color and without erythema. On the patency test, was observed Warthor’s duct obstruction in the right side. Occlusal radiographic revealed no radiopacities on the tubular region. Computed tomography (CT) did not reveal any other detail other than an enlargement of the submandibular gland. Considering the hypothesis of sialocele, the patient was submitted twice to a marsupialization procedure, one under local anesthesia and other under general anesthesia, without success. Another CT and also an ultrasonography were performed, at this time revealing the presence of radiopaque structures similar to calculi inside the submandibular gland and at the posterior region of its duct [Figure 1b]. The diagnostic was then suggested to be sialolithiasis. Thus the procedure of gland excision was indicated under general anesthesia through a submandibular approach [Figure 2a and b]. During the surgical procedure, two stones were identified (one inside the gland and another in the duct) and an inflammatory glandular tissue was observed around the calculi located in the gland parenchyma [Figure 2c and d]. The histological analysis confirmed the diagnosis as a sialadenitis secondary to a sialolithiasis. In a post-operative control of 6 months, the patient evaluated with normal tongue’s movement and sensibility as well as mimic facial expressions preserved [Figure 3].

Case 2
A 50-year-old, female patient presented to the Oral and Maxillofacial Surgery Department of the General University Hospital of the University of Cuiaba with a complaint of an unpleasant feeling below the tongue when she ate acid or sour foods. During the physical examination, a firm swelling was observed by bimanual palpation associated to the right submandibular gland with no patency of the Warthor’s duct. A CT was taken, and it revealed the presence of two radiopaque structures compatible with calculi at the posterior duct’s region and inside the glandular parenchyma. Surgical removal of the gland was indicated, but the patient did not come to the surgical procedure and gave up on the treatment. One year later, she presented to the local emergency hospital with an abscess on the submandibular region [Figure 4a]. Another CT was taken and revealed the same two calcified structures previously observed [Figure 4b]. Surgical drainage was performed under general anesthesia to resolve the infection and the patient were referred to further follow-up to program the submandibular gland excision. Although the patient waited almost 1 year to return, definitive treatment was accomplished. During the elective surgical removal of the submandibular gland, a fibrous tissue was found around the posterior region’s duct, a large sialolith was removed inside the glandular parenchyma and another was removed from inside

Figure 1: Pre-operative observation of a discrete swelling at the right submandibular area (a); computed tomography observation of two calculi (red arrows, b)

Figure 2: Submandibular approach, when the inflammatory aspect of the gland could be observed (a), the duct was properly separated from adjacent structures for ligation (b), a calculus could be observed inside the gland (c) and the gland already removed beside the other calculus (d).

Figure 3: At 6 months post-operative follow-up, facial nerve functions preserved (a and b) as well as a good esthetic outcome of the scar at the submandibular area (c)
de Wharthon’s duct at its posterior region [Figure 5]. The histological analysis confirmed the diagnosis as a sialadenitis secondary to a sialolithiasis. In a post-operative control of 6 months, the patient presented with normal tongue’s movement and sensibility as well as mimic facial expressions preserved [Figure 6].

Discussion

There are some theories to the etiology of sialolith formation. Some believe that it occurs as a failure of the process of void intracellular microcauculi through the duct system, acting as a nidus for stone formation.[5] Others believe in a deposition of calcium salts around an organic nidus (altered salivary mucins, bacteria and desquamated epithelial cells) or around aliments, substances or bacteria which migrate into the salivary duct.[2,3]

The submandibular gland is most affected because its salivary composition, higher mucus levels than the other major salivary glands, a greater concentration of phosphate and calcium, an increased alkaline composition and due the long duct, that has a curvature over the posterior border of the mylohyoid muscle and leads an antigravity flow.[2,6]

The most common form of inflammation of the submandibular gland is the acute sialadenitis, and obstruction of the salivary duct is the most frequent cause of sialadenitis.[1] It has common symptoms like pain and swelling when food that increases the salivary secretion is eaten. Due to the inability of proper salivary secretion, this salivary stasis may cause infection of the submandibular gland due to bacterial migration that can happen into the parenchyma of the gland.[7] Long periods of obstruction without infection can lead to gland atrophy, lack of secretory function and ultimately to gland fibrosis.[3]

In the first case hereby presented, the patient did not had any previous infection related to the affected gland, but two different characteristics were found in the same gland: On the anterior region, there was an inflamed glandular tissue involving the calculi with an associated fibrosis due previous manipulation; and on the posterior region, an atrophied glandular tissue suggesting lack of function. In the second case, there was an atrophied gland and a fibrosis around the initial portion of the Wharthon’s duct, probably due to the episode of infection presented by the patient almost 1 year before the surgery.

For diagnostic purposes, a professional can use of some exams, like sialography, mandibular occlusal radiographs, CT scan and ultrasonography or sialendoscopy. In both cases, we used CT scan to diagnosis and located the position of the calculi. In the first case, the first CT was not contributory to diagnosis but the second CT and the ultrasonography were helpful as they revealed two sialoliths inside the glandular parenchyma, informing their size (40 mm and 30 mm) and indicated increased vascularity of the glandular parenchyma. In the second case, although only the CT scan was necessary as the calculi were already seen at the first exam.

The treatment for this condition depends on the size, location and number of the sialoliths. Extracorporeal shock wave lithotripsy can be used to stone fragmentation, but this technique is more effective in parotid than in submandibular stones, because it is easier to locate the calculi and position the lithotriptor in the parotid’s region.[5] Some authors believe that surgical gland excision with the sialolith is the gold standard technique when the gland presents as non-functional.[2] In the two cases presented here, the submandibular glands presented
no patency on the Wharton’s duct of the affected glands. During surgery, it was possible to clinically observe the aspect of an atrophied gland. Many studies have been presenting the sialendoscopy as a new form of manipulation of these calculi,[1,2,4,10] but this technique requires specific equipment and exists a risk of stone fracture during manipulation and dissemination of the calcified material into the gland, increasing the chance of recurrence.[2] To sialoliths located on the two anterior thirds of the Wharton’s duct or those whose are palpable by intraoral, they can be removed by a linear incision on the oral mucosa along the duct or by transoral CO₂ laser.[2,4] There are no studies proving that electrohydraulic or pneumoblastic devices are good options for management of sialoliths.[11]

Surgical approaches to management the submandibular gland are planned to allow direct access and a good view of the gland and in areas of natural skin creases, for an esthetic scar. The submandibular gland is located in the submandibular triangle that is delimited, anteriorly, by the mylohyoid muscle; posteriorly by the hypoglossus muscle; and by the anterior and posterior bellies of the digastric muscle and the mandible.[12] The classical incision to access this gland is the transcervical or Risdon’s approach. The surgeon must be careful with some important anatomical structures during the procedure such as the marginal mandibular nerve, that is a branch or the facial nerve and passes superficial to the fascia of the submandibular gland; the lingual nerve, branch of the trigeminal nerve, which loops under the Wharton’s duct running a lateral to medial course and go deep and superior to this gland; and the hypoglossal nerve, twelfth cranial nerve, which passes anterior and medial to the lower third of the gland under the posterior belly of the digastric muscle.[12] In both cases, the classic submandibular approach was used, and the patients did not experiment any nerve damage to these nerves. The post-operative scars were almost invisible, and the patients were satisfied with the result of the treatment.

Conclusion

Despite the variety of techniques available to the treatment of sialolithiasis, the submandibular gland excision with the sialolith demonstrated to be a well-indicated and secure technique to treat sialadenitis due sialolithiasis, when the calculi is located in areas accessible by the intraoral approach and when the gland is not functional. The surgical technique is easy to perform, but careful is necessary not to damage important structures around to the submandibular gland.

References