


Prospective evaluation of the sialoadenectomy as treatment for giant sialolith in the submandibular gland: Case series

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Abstract:

Objective: The objective of this study is to report a series of cases of giant submandibular gland sialoliths treated by sialoadenectomy. **Methods:** Nine patients were admitted to the Maxillofacial Surgery Department of Nova Iguaçu General Hospital from 01/01/2012 to 01/05/16, presenting submandibular gland sialoliths larger than 1.5 cm without response to conservative treatment. All patients underwent sialoadenectomy through cervical access. **Results:** Patients presented a mean age of 51 years, with 7 being male and 2 being female. The size of the sialoliths removed ranged from 1.8 to 4.9 cm. None of the patients presented hypoglossal nerve complications, 2 had transitory marginal mandibular nerve palsy, and 2 patients had postoperative infections in the first week. **Conclusion:** We can conclude that giant sialoliths represent a greater difficulty for the treatment of sialolithiasis, where sialoadenectomy represents a resolutive therapy with few complications.

Keywords: Submandibular Gland Diseases; Calculi; Surgery, Oral

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INTRODUCTION

Sialoliths are defined as calcified structures that develop in the salivary glands or in their ducts. They may originate from the deposition of calcium crystals or be related to salivary retention due to anatomical factors (narrow and tortuous salivary duct) and the composition of the saliva itself. Sialolithiasis is a disease characterized by the development of sialoliths, which are also called salivary calculi, within the salivary glands or their ducts^{1,2}.

Sialolithiasis accounts for about 30% of salivary changes, and is the most common cause of submandibular gland obstruction, with an incidence in young and middle-aged adults (30 to 50 years old)³. The most affected site is the ductal system of the submandibular gland, which makes up 80 to 90% of the cases^{1,4}. A patient presenting with this condition may exhibit pain and edema in the path of the ductus or involved gland, depending on the site of the obstruction, with a slow evolution, and with the presence of sialoliths of diameter that rarely exceed 1.5 cm⁴⁻⁶.

The diagnosis of sialolithiasis of the submandibular gland is made by means of inspection and palpation of the buccal floor and submandibular region, and confirmation of the presence of sialoliths is performed by imaging tests such as panoramic and occlusal radiographs, ultrasonography and computed tomography^{5,7-9}.

The choice of treatment is directly related to the location and size of the salivary calculus^{1,6,10}. The location of the calculus in the posterior portion of the duct, internally of the gland, and a sialolith of large proportions, makes response to minimally invasive treatment difficult, requiring a more complex approach by extra-oral access to sialoadenectomy, which may lead to greater complications such as hypoglossal nerve, facial nerve, and salivary fistula lesions.

This study aims to analyze a series of 9 cases of giant sialoliths of the submandibular gland, where in all cases it was necessary to perform the sialoadenectomy to resolve the pathological process.

METHODS

This study was approved by the Research Ethics Committee of the Nova Iguaçu General Hospital, with the protocol CAAE (Certificate of Presentation for Ethical Appreciation) number 54545916.0.0000.5283, and final opinion No. 1,500,925 (Nova Iguaçu, Rio de

Janeiro, Brazil). An analysis of 9 cases of sialoadenectomy performed as a treatment of sialolithiasis of the submandibular gland in cases of giant sialoliths was carried out.

The patients of this case series sought care in the department of oral and maxillofacial surgery of Nova Iguaçu General Hospital from 01/01/2012 to 01/05/16. All patients underwent clinical examination and computed tomography to confirm the diagnosis and analysis of the sialolith location (Parenchyma or duct). This study selects 9 patients who presented sialoliths larger than 1.5 cm in a region of submandibular gland (Fig.1).



Figure 1. Computed tomography demonstrating giant sialolith in the left submandibular gland.

The surgical procedure was performed under general anesthesia with nasotracheal intubation. A baseline was marked 2.0 cm below the lower border of the mandibular angle, containing a 3.0 cm extension, followed by the infiltration of 1: 200.000 adrenaline without local anesthetic for hemostasis control.

The submandibular approach is performed with a skin incision and blunt dissection until the superficial layer of the deep cervical fascia is reached. At this point a careful dissection begins, until complete exposure of the submandibular gland, with possible ligation of the facial vessels. With the aid of an Allis clamp, the gland

is drawn to allow dissection of the medial part, with special attention to the duct and possible contact with the hypoglossal nerve.

The gland was removed completely, and the hemostasis of the remaining tissues with electrocautery was performed if necessary (Figs. 2 and 3). The remnant of the duct of the submandibular gland needs to be identified and connected with Cotton 2.0. A simple suture of the muscular planes is then performed with 3-0 polyglactin 910 acid thread, and the skin is subjected to an intradermal suture with Nylon 5.0.

The patients attended postoperative follow-up in the first week immediately after surgery, and at least once a month until the conclusion of the first 6 months. During the postoperative period, movements of the tongue and facial mimesis, surgical access scarring, and possible decrease of salivary flow were evaluated.

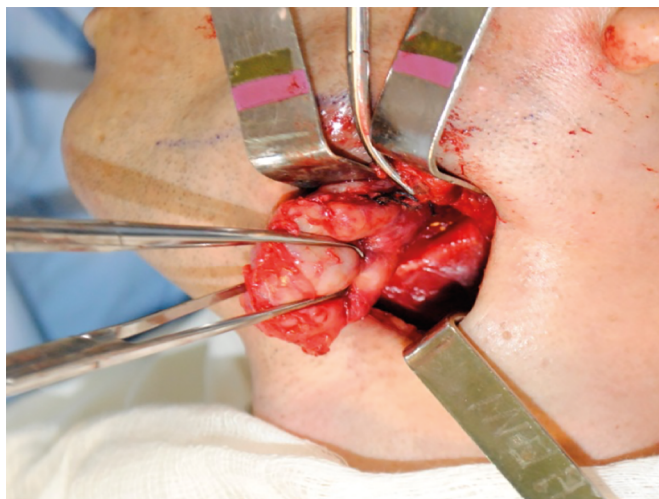


Figure 2. Stabilization and dissection of the submandibular gland.

RESULTS

The results of the analysis are shown in Table 1. Of the total 9 patients, 7 were males and 2 females, with ages ranging from 41 - 73 years and a mean age of 51 years. In four cases the sialoliths were in the proximal portion of the duct of the submandibular gland, and in another 4 they remained internally in the parenchyma of the gland. The sialoliths had part of its structure in the parenchyma of the gland, and part in the Wharton duct, in only one case. The largest sialolith found was 4.9 cm and the smallest 1.8 cm, with a mean of 3.2 cm.

Regarding nerve damage, 2 patients presented paralysis of the mandibular marginal branch of the facial nerve in the immediate postoperative period, but after



Figure 3. Sialolith with approximately 4 cm removed from the submandibular gland parenchyma.

3 months of follow-up no deficit was found. No patient presented complications related to the hypoglossal nerve. One black patient presented keloid in the surgical access, without the necessity for re-intervention. When questioned about the sensation of “dry mouth” for evaluation of decreased salivary flow, only 1 patient reported this complication. Two patients presented postoperative infection in the 1st week post-procedure, and were treated by drainage during suture removal, and oral antibiotic therapy 500 mg of Amoxicillin with 100 mg of clavulanate potassium (Clavulin® - GlaxoSmithKline Brasil Ltda - Rio de Janeiro - Brazil)

DISCUSSION

The high incidence of sialolithiasis related to the submandibular gland can be explained by the alkaline pH, mucous secretion and high concentration of calcium of this gland, remembering that sialolithiasis is one of the most common pathologies of salivary glands in middle-aged patients. Because of its long duct, which surrounds the mylohyoid muscle, and secretion in the anti-gravitational sense, may also contribute to the sialolithiasis prevalence¹⁰.

Facial edema is the most frequent sign of the patient with this alteration, accompanied by pain and swelling of the affected glands during meals, and fever and purulent drainage intra- or extra-buccal may be present¹. All patients in this study sought hospital attention because they presented facial volume increase,

Table 1. Table demonstrating the patients treated with sialoadenectomy of the submandibular gland with size of the sialoliths, complications and follow-up.

| | Genre | Age | Local | Size | Hypoglossal nerve paresis | Marginal branch paresis | Dry mouth | Other complications (Infection, trismus, scar, lingual nerve disturby) | Follow-up |
|----------|--------|-----|---------------------|--------|---------------------------|-------------------------|-----------|--|-----------|
| Case - 1 | Male | 45 | Parenchyma | 4,0cm | No | No | No | Infection | 2 years |
| Case - 2 | Female | 42 | Duct | 1,9cm | No | Yes | No | No | 8 months |
| Case - 3 | Male | 51 | Duct | 3,4cm | No | No | No | No | 1 year |
| Case - 4 | Female | 73 | Duct and parenchyma | 4,9cm | No | No | No | No | 6 months |
| Case - 5 | Female | 54 | Parenchyma | 3,8cm | No | No | YES | Scar | 2 years |
| Case - 6 | Male | 51 | Parenchyma | 2,8 cm | No | No | No | Infection | 2 years |
| Case - 7 | Male | 59 | Duct | 1,8cm | No | Yes | No | Infection | 9 months |
| Case - 8 | Male | 41 | Duct | 2,5cm | No | No | No | No | 1,6 years |
| Case - 9 | Male | 48 | Parenchyma | 3,5 | No | No | No | No | 1 year |

*infection, trismus, scar, lingual nerve disturbs.

pain, and, in 3 cases, had a history of fever and purulent intrabuccal secretion.

Conservative treatment of sialolithiasis is based on increased salivary flow so that the calculation is expelled intra-buccally, using sialogogues, fluid ingestion, and manual stimulation of the gland⁴. This therapy is more effective with sialoliths smaller than 1.5 cm, and in the distal portion of the duct, but all cases operated on in this study performed conservative therapy while waiting for the surgical procedure (10-days maximum) as an attempt to avoid the more morbid procedure.

Sialoliths can be removed by intraoral approach even if they are considered giant sialoliths, but in that case, they need to be located in the most distal portion of the Wharton duct, near to the oral cavity^{5,11}. In 2013, Chang et al.¹¹ conducted a comparative study of the intraoral and extraoral approaches to sialoadenectomy, in which it was found that the procedure time and sensorial complications of the lingual nerve were higher in the intraoral group, but the hospitalization and paralysis time of the facial nerve was higher in the extraoral group. In our study we used the cervical approach, in which the results were similar to that of Chang et al. because facial paralysis was found transiently in 2 cases, and we did not obtain any changes related to the lingual nerve. Although the advantages of intraoral approach, our choice was by cervical approach because of its facility to expose the submandibular gland, a shorter procedure time and a low rate of complications.

The minimally invasive surgery for treatment of sialolithiasis is undoubtedly via sialoendoscopy^{12,13}. Hasan and Curran¹² obtained a 95% success rate in the removal of sialoliths by sialoendoscopy. In this procedure, the stone is broken in small fragments however, in giant sialoliths this is impossible to occur beside that there is not enough ductal dilatation for its removal.

Sialoadenectomy represents the most morbid procedure and it is indicated in cases of giant sialoliths, smaller sialoliths located into the gland parenchyma, and at the proximal portion of the duct when doesn't respond to the conservative treatment^{5,10}.

About the complications of this procedure, paralysis of the hypoglossal nerve or facial nerve can limit functionally and socially the patient's life^{4,15}. In the present case series, occurred facial nerve paralysis in 2 cases however, was transient and the movements returned spontaneously. In 2 cases, the hypoglossal nerve was identified during the surgical procedure and the sialolith was in the proximal portion of the duct, requiring a deeper dissection of the tissues. Despite this, no patient presented alterations in the hypoglossal nerve functions.

Decreased salivary flow after sialoadenectomy may be a late complication after sialoadenectomy of the submandibular gland¹⁵. In this case series, only one patient reported this complication, but the patient used antidepressant medication, which may also be associated with this symptom.

Two patients had infection in the immediate postoperative period. This complication was associated with the formation of dead space during the suture of the procedure. In cases where the surgeon suspects the formation of a hematoma or is unable to reduce the dead space in the suture, the use of a Penrose Drain in the first 2 postoperative days may be beneficial¹⁶.

In this series of cases, we can conclude that the giant sialoliths represent a greater difficulty in the treatment of sialolithiasis, especially when located into the parenchyma of the submandibular gland. For these cases, sialoadenectomy was presented as a resolutive technique and with low morbidity.

REFERENCES

1. Ledesma-Montes C, Garcés-Ortiz M, Salcido-García JF, Hernández-Flores F, Hernández-Guerrero JC. Giant sialolith: case report and review of the literature. *J Oral Maxillofac Surg.* 2007;65:128-30.
2. Graziani F, Vano M, Cei S, Tartaro G, Mario G. Unusual asymptomatic giant sialolith of the submandibular gland: a clinical report. *J Craniofac Surg.* 2006;17:549-52.
3. Lee LT, Wong YK. Pathogenesis and diverse histologic findings of sialolithiasis in minor salivary glands. *J Oral Maxillofac Surg.* 2010;68:465-70.
4. Kraaij S, Karagozoglu KH, Forouzanfar T, Veerman EC, Brand HS. Salivary stones: symptoms, aetiology, biochemical composition and treatment. *Br Dent J.* 2014;217:E23.
5. Arunkumar KV, Garg N, Kumar V. Oversized submandibular gland sialolith: a report of two cases. *J Maxillofac Oral Surg.* 2015;14(Suppl 1):116-9.
6. Bodner L. Giant salivary gland calculi: diagnostic imaging and surgical management. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 2002;94:320-3.
7. Oliveira Tde P, Oliveira IN, Pinheiro EC, Gomes RC, Mainenti P. Giant sialolith of submandibular gland duct treated by excision and ductal repair: a case report. *Braz J Otorhinolaryngol.* 2016;82:112-5.
8. Leung AK, Choi MC, Wagner GA. Multiple sialoliths and a sialolith of unusual size in the submandibular duct: a case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* 1999;87:331-3.
9. Rai M, Burman R. Giant submandibular sialolith of remarkable size in the comma area of Wharton's duct: a case report. *J Oral Maxillofac Surg.* 2009;67:1329-32.
10. Arslan S, Vuralkan E, Çobanoğlu B, Arslan A, Ural A. Giant sialolith of submandibular gland: report of a case. *J Surg Case Rep.* 2015;2015. pii: rjv043.
11. Chang YN, Kao CH, Lin YS, Lee JC. Comparison of the intraoral and transcervical approach in submandibular gland excision. *Eur Arch Otorhinolaryngol.* 2013;270:669-74.
12. Hasan W, Curran A. Sialoendoscopy in the management of salivary gland disorders-4 years experience. *Ir Med J.* 2014;107:120-1.
13. Baurmash HD. Re: Iidas et al. Plunging ranula as a complication of intraoral removal of a submandibular sialolith. *Br J Oral Maxillofac Surg* 2001; 39: 214-216. *Br J Oral Maxillofac Surg.* 2001;39:486-7.
14. de Carvalho AS, Dedivitis RA, de Castro MA, Nardi CE. Submandibular gland excision. *Rev Col Bras Cir.* 2015;42:14-7.
15. Springborg IK, Møller MN. Submandibular gland excision: long-term clinical outcome in 139 patients operated in a single institution. *Eur Arch Otorhinolaryngol.* 2013;270:1441-6.
16. Laverick S, Chandramohan J, McLoughlin PM. Excision of a submandibular gland: a safe day case procedure? *Br J Oral Maxillofac Surg.* 2012;50:567-8.