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report

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# Brazilian Journal of OTORHINOLARYNGOLOGY

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## CASE REPORT

### Giant sialolith of submandibular gland duct treated by excision and ductal repair: a case report<sup>☆,☆☆</sup>



### Sialolito gigante de ducto da glândula submandibular tratado por excisão e reparo ductal: relato de caso

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## Introduction

Sialolithiasis is one of the most common diseases of the salivary glands.<sup>1,2</sup> It is a condition characterized by an obstructive phenomenon in a salivary gland or in its excretory duct due to a calculus.<sup>1</sup> The clinical presentation is usually characterized by local swelling, pain, infection of the affected area, and dilation of the salivary duct.<sup>1</sup> Sialolithiasis usually affects adults between the third and fourth decades of life, with a frequency of 12:1000.<sup>3</sup> The number of cases in male patients is about twice that of female patients.<sup>3</sup> It is estimated that 80–90% of cases occur in the

submandibular gland, while 10–20% occur in the parotid gland.<sup>3</sup> The size of the calculi varies from <1 mm to a few centimeters. Although the frequency of sialolithiasis is relatively high, the occurrence of giant sialoliths, larger than 1.5 cm in any diameter, is rare. For this reason few studies are found in the pertinent medical literature.<sup>1,4</sup>

This report describes a case of giant sialolith in a 42-years-old male, addressing the clinical features, the diagnosis, and the ductal repair surgery performed to restore salivary flow.

## Case report

The patient, a 42-year-old black man, attended a dental appointment in March of 2014. After routine radiographic examination, he was referred for a consultation with an oral and maxillofacial surgeon, in April of 2014. During anamnesis the patient denied any previous diseases. He reported only an uneventful surgery on the right leg. The physical examination showed an ankyloglossia and, during palpation, a hardness in the right submandibular salivary gland. To further investigate the case, imaging exams were requested (Fig. 1A). A provisional diagnosis of sialolithiasis in the right submandibular gland duct was suggested.

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**Figure 1** (A) Computed tomography scan (axial aspect) revealing a mineralized tissue with heterogeneous density and dimensions of  $3.0 \times 1.0$  cm, approximately. (B) Three-dimensional image of the sialolith and the mandible.

Since the sialolith had exuberant dimensions, an excision followed by the reconstruction of the submandibular gland duct was proposed. Blood tests and surgical risk exams were requested for the patient.

On May 21, 2014, the surgical procedure was conducted by an intra-oral approach. The sialolith was removed by curettage after direct incision of the duct. A partial mineralization favored the fragmentation of the distal portion of the calculus. A true salivary gland cyst was removed in association with the calculus (Fig. 2).

For the treatment of ankyloglossia, a tongue frenectomy was performed. To restore the salivary flow, a No. 8 urethral catheter was placed in the residual duct path. The mucosa was sutured around the catheter using a Vicryl 3-0 suture in order to repair the duct of the submandibular gland.

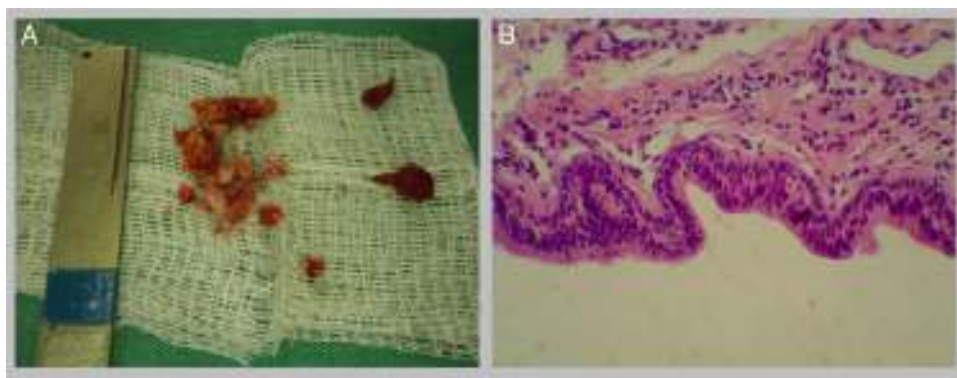
The other tissues were sutured in anatomical planes and there were no complications during the surgical procedure.

Two days after the surgery, an ultrasound showed that the catheter was inside the submandibular gland duct (Fig. 3). After milking of the gland, the presence of crystalline liquid flowing from within the tube was noted (Fig. 4). Eight days after the surgery, the patient reported an increase in salivary volume and the occurrence of contractions in the submandibular gland region.



**Figure 3** Ultrasound showing the catheter inside the salivary gland duct.

The sutures and the drain were removed fourteen days after the surgery. A gland milking maneuver showed copious salivation, indicating that the performed surgical technique succeeded in reconstructing the ductal structure. The



**Figure 2** (A) The fragmented stone is seen on the left. On the right side, there is a tissue corresponding to the salivary cyst. (B) Histological section of the cyst showing oncocytic epithelium compatible with ductal epithelium (hematoxylin and eosin stain,  $400\times$ ).

**Table 1** Comparative table of consulted cases.

Author	Sialolith size	Symptoms	Removal method	Age	Gender
Gupta et al. (Case 1)	2.8 cm × 1.1 cm	Intermittent, dull aching pain, and swelling in left submandibular area during meals	Surgically removed via intraoral approach under local anesthesia and transposition of ductal opening	48	Male
Gupta et al. (Case 2)	1.9 cm × 5.0 cm	Swelling in mouth associated with pain over left side of face during intake of food	Surgically removed via intraoral approach under local anesthesia and transposition of ductal opening	45	Female
Iqbal et al.	3.5 cm × 3.0 cm	Asymptomatic	Surgery under local anesthesia, intra-oral approach with marsupialization	55	Male
Dalal et al.	1.8 cm × 6.0 cm	Pus discharge and continuous pain of pricking and sharp nature, radiating to the tongue with restricted tongue movement	Sialolithotomy via intraoral approach under local anesthesia	40	Female
Fowell & MacBean	4.1 cm	Pain in the right floor of mouth and submandibular region, exacerbated by swallowing	Excision of the right submandibular gland and stone via a standard extra-oral approach	58	Male
Krishnan et al. (Case 1)	3.4 cm	Recurrent pain and swelling over eight years that increased during meals. In the last two years presented asymptomatic	Sialolithotomy via intraoral approach under local anesthesia. The wound was left to heal by secondary intention	41	Male
Krishnan et al. (Case 2)	2.5 cm	Multiple episodes of pain and swelling in the left lower part of the mandible, during the past four to five years, especially at meal times	Surgically removed through a transoral approach, with sharp dissection under local anesthesia	32	Female

follow-up appointments within two months of the surgery showed no complications or complaints.

## Discussion

Sialolithiasis is a disease that can affect any age group, with a higher prevalence in male adults.<sup>2,5</sup> It mainly affects



**Figure 4** The catheter and the sutures are in the correct surgical placement.

the submandibular gland.<sup>6</sup> Despite being a common disease, the presence of giant calculus is extremely rare and most sialoliths do not exceed 1.5 cm.<sup>3,5</sup> The calculus in the present case had dimensions of approximately 3.0 cm × 1.0 cm, thus considered a giant sialolith.<sup>1</sup>

The reported symptoms usually are pain and swelling in the gland, which worsen during the meal time (Table 1).<sup>2-4,6,7</sup> In the current report, the patient remained asymptomatic despite the exuberant dimensions of the stone.

According to Jensen<sup>8</sup> and Cawson et al.,<sup>7</sup> salivary stones can be associated with the presence of true salivary gland cysts. Such lesions occur due to the obstruction of the salivary flow, followed by a proliferation of the duct epithelium that surrounds the stone. The present specimen presented a squamous and oncocytic differentiation in accord with the literature.<sup>8</sup>

The pathophysiology of the stone formation is still poorly understood.<sup>3</sup> However, it is believed that the sialolith is formed after the deposition of calcium salts around a "niche" of organic material.<sup>7</sup>

In 80% of cases the submandibular gland is affected<sup>7</sup> due to a number of synergistic factors, such as: (a) the

composition of the saliva produced by the gland, which is more alkaline and with a major concentration of calcium<sup>6</sup>; (b) the salivary flow occurs against gravity<sup>2,9</sup>; and (c) the long and tortuous anatomy of the duct of the submandibular gland.<sup>6,9</sup> All these factors work together in the formation of the calculus in the submandibular gland.<sup>2,6,9</sup> In the authors' opinion, the occurrence of the sialoliths presented in the consulted literature is in line with their understanding.

Regarding the treatment, a less invasive procedure is of utmost importance in order to preserve the gland's function.<sup>2,4,7,9</sup> The pertinent literature indicated some surgical procedures such as trans-oral sialolithotomy, sialoendoscopy, extracorporeal shockwave lithotripsy, and resection of the gland.<sup>2,3</sup> For small sialoliths, conservative treatments using sialogogues and massage of the gland are also possible.<sup>7</sup> The current case showed the treatment of an exuberant calculus through an intra-oral approach associated with a ductal repair. Although Fowell et al.<sup>2</sup> concluded that sialoplasty is one of the main treatments for giant sialoliths, this technique has not been described or used by the authors consulted. They performed the removal of the sialolith with closure by secondary intention.

Among the possible surgical complications, one is injury of the mandibular nerve,<sup>2</sup> another is Wharton's duct stenosis.<sup>2</sup> There was no evidence of any of these complications in the present case. The ductal repair maintained salivary flow between the gland and the oral cavity.

The surgical removal of sialolith varies between surgeons. The preferred approach is mostly performed through intra-oral intervention (Table 1).

## Conclusion

The present case report described the removal of a giant sialolith. To the best of the authors' knowledge, this case

is unique with regard to the surgical ductal repair after the excision of a salivary stone.

## Conflicts of interest

The authors declare no conflicts of interest.

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