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# SIALOLITHIASIS GIANT OF THE SUBMANDIBULAR GLAND TREATED BY SURGICAL EXCISION: CASE REPORT

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#### ABSTRACT

The giant sialoliths are defined as those that exceed 15 mm in one of their dimensions. In an asymptomatic context the location in the body of the submandibular gland has been frequently described. Giant sialoliths of more than 20 mm are rare. We report the case of a 67-year-old woman who gradually developed duct's hilum sialolithiasis on a non-functional gland that led to submandibulectomy. Careful diagnosis helps to make a reasonable choice of surgical technique. Surgery should be performed with the minimal invasive method to avoid complications associated with surgical techniques.

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### INTRODUCTION

Sialolithiasis is one of the most common diseases of the salivary glands [1]. It accounts for over 50% of the major salivary gland disorders. It is characterized by an obstructive lithiasic deposit in a salivary gland or in its excretory duct. The clinical presentation is generally characterized by local swelling, pain, infection of the affected area and dilation of the salivary duct. Sialolithiasis usually affects adults between the third and fourth decades of life, with a frequency of 12/1000 [1] [2]. Men represent about two-thirds of the patients [1] [2]. In 80 to 90% of the cases the sialolithiasis sit in the submandibular gland, whereas in 10 to 20% they relate to the parotid [1] [2]. The giant sialoliths are defined by a diameter of at least 1.5 cm. They are rare and few studies are found in the medical literature [1]. This observation is in addition to the rare case of giant sialolithiasis described in the literature.

### **CASE REPORT**

The patient is a 67-year-old woman with no notable antecedents, who consulted for a left mandibular swelling. At the anamnesis, the swelling began about two years ago and it was painful. It was initially marked by salivary colic at mealtimes with fluctuating size; Over the last nine months it has steadily increased in volume. The combined bi-manual palpation of the submandibular region and the buccal floor showed a hard, painful, well limited and mobile gland with a stony appearance from the posterior half of the lateral region

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of the buccal floor. The massage of the submandibular gland did not allow the flow of any purulent fluid at the orifice of the Wharton duct. Examination of the other main salivary glands was normal. In the CT imaging a voluminous and bony density lithiasis, extended in the antero-posterior axis of the submandibular gland, on the inner face of the left horizontal branch of the mandible was detected; She measured about 22X12mm (Fig.1).



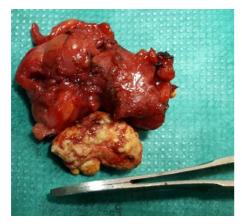
Figure 1 Axial CT scan showing a giant sialolithiasis of the submandibular gland.

Antibiotic and anti-inflammatory treatment was instituted over a period of eight days, followed by a pre-anesthetic evaluation; surgical treatment under general anesthesia consisted of left submandibular removal by a cervical approach. This surgery was made difficult by the voluminous size of the lithiasis and the glandular fibrosis which masked the lingual nerve (Fig.2).



**Figure 2** Intraoperative view showing sialolithiasis in the hil of the submandibular gland.

After glandular excision, the lithiasis had a gray-yellow color, a compact and hard structure (Fig.3). The closure of the submandibular region was performed in two planes on Redon drain. No postoperative complications were reported and the patient was discharged after 48 hours of hospitalization. The histopathological study confirmed the diagnosis of non-specific lithiasic sialadenitis.



**Figure 3** The specimen image showing the compact appearance of the sialolithiasis.

# **DISCUSSION**

The presence of giant calculi is extremely rare and most of the sialoliths do not exceed 1.5 cm of larger diameter [1] [2]. Although the exact etiology is unknown, it is believed that sialoliths occur as a result of the deposition of mineral salts around an initial nidus consisting of altered salivary mucin, bacteria or desquamated epithelial cells [3].

The stagnation of the salivary flow, the high alkalinity and the increased calcium content predispose to the formation of sialoliths. Salivary lithiasis are most often found in the submandibular gland than in the parotid gland due to factors such as the direction of salivary flow against gravity, a longer and more tortuous Wharton duct structure, and increased content In calcium and mucin of the saliva produced in the submandibular gland [4]. Most calculi occur in Wharton duct, whereas the rest are found at the ductal hilum or within the glandular tissue. Sialoliths of submandibular gland are rarely radioluscent and are recognized as filling defects in sialography [5]. The sialoliths usually remain in the gland and enlarg, they rarely migrate to the oral cavity or fistulate in the cervical region [1] [2]. The ability of a calculus to grow and become a giant sialolith depends mainly on the reaction of the

affected duct. When the sialolith is located in a duct able to dilate and allow normal salivary flow around the stone, sialolith may increase in size until it becomes giant and remains asymptomatic for a long time [6]. Giant sialoliths are usually organized hard concretions, with a porous aspect, vellow color and hard texture. They usually have an oval or long shape and they may appear hyperdense to radiographic images [6]. Submandibular gland sialolithiasis generally have an asymptomatic evolution. Symptoms occur at mealtimes whenever the lumen of the Wharton duct is completely obstructed with epithelial proliferation around the lithiasis. The resulting salivary stasis then causes pain and swelling of the diseased gland; Then a bacterial propagation occurs in the glandular parenchyma exposing to recurrent infections. In the long term this obstructive and infectious process can lead to atrophy of the gland with ultimately fibrosis and loss of the secretory function as in this observation. The bi-manual palpation of the submandibular gland and the buccal floor makes it possible to feel the presence of a hard mass evoking the diagnosis a giant calculus. Medical imaging makes it possible to refine the diagnosis. Ultrasonography is a firstlevel diagnostic technique because it reveals highly mineralized ductal sialolithiasis with a diameter of at least 1.5 mm and an accuracy of 99% [6]. Conventional trans-occlusal endoral radiography seems more useful. The sialography makes it possible to detect the salivary calculi and the visualization of the whole duct system; However, sialography is not indicated in the case of acute infections, allergy to contrast medium or radio-opaque calculus in the distal part of the duct. CT is the radiological examination of choice for the detection of sialolithiasis. Sialendoscopy is a new method for directly visualizing the lithiasis [6] [7]. In terms of treatment, the literature indicates a variety of procedures such as transoral sialolithotomy with ductal reconstruction, interventional sialoendoscopy, extracorporeal shock wave lithotripsy, and gland resection, but also the use of sialogogues with external massage of the gland for the smallest lithiasis [1] [2]. The appropriate treatment method is chosen according to the size, location, number and mobility of the sialoliths, the permeability of the duct and the functional character of the gland. Small sialoliths can be removed through the duct orifice via bimanual palpation or by sialoendoscopy. Larger sialoliths or located in the duct require trans-oral surgical removal. Resection of the submandibular glands is indicated when a sialolith is larger (12 mm or more) and is located in the gland and can't be removed by trans-oral approach [8]. Other therapeutic means have recently been used: extracorporeal lithotripsy has been used whatever the size and position of the calculus in the salivary glands. Few contraindications are described for this method: acute infection, anomaly of the blood crass and taking anticoagulants. It is recommended to use only lithotripters specially designed for salivary lithiasis. [9]

# CONCLUSION

Giant sialolithiasis represent the ultimate evolution of an obstructive pathway of salivary excretion. Their diagnostic and therapeutic management is a challenge for the practitioner and requires a careful preoperative assessment. The choice of the surgical method fluctuates between a trans-oral sialolithotomy with sialodochoplasty which can preserve the secretory and excretory function of the gland and its duct. In

addition, radical submandibulectomy provides the solution if the gland is atrophic and non-functional.

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