

## Case Report

### Case report: A Giant Calculus of the Submandibular Gland

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#### Abstract

Sialolithiasis is a pathological condition characterized by the formation of one or more calculi in the salivary glands. It is the most common disease of the salivary glands after mumps. The calculi usually range in size from 1 mm to 10 mm. They have rarely been reported to be larger than 15 mm. We describe a patient with a large calculus of the submandibular gland measuring 40 mm by 9 mm managed completely in the emergency department and present a review of the current literature related to the management of giant calculi.

**Keywords:** Emergency Department Management; Giant Calculus; Sialolithiasis

#### Introduction

Since they are very rare, few reports of large calculi of the salivary glands have been described in the literature. Salivary gland calculi generally present with localized swelling and pain. Occasionally, patients may present with a complication such as infection of the affected gland [1]. Pain often becomes worse while eating. The diagnosis is established by history and clinical examination and confirmed with X-ray, US, CT scanning and sialography. Management of salivary gland calculi includes medical, surgical, lithotripsy and endoscopic treatments.

We present a case of one of the largest salivary gland calculi reported to date that was entirely managed in the emergency department using a rapid surgical treatment to be described.

#### Case report

An otherwise healthy, non-smoking, 48 year old, male patient presented to the emergency department of the University Hospital of Verona with the sensation of a “stone” in the floor of the mouth and swelling in the right submandibular region. These symptoms had worsened over the last week. The patient had previously visited the emergency department for right submandibular swelling with purulent discharge from Wharton’s duct and pain on the right half of his face. On that occasion, the ENT specialist had recommended

antibiotics and corticosteroids for 7 days and outpatient follow-up. Five days later he came back in the ED, the physical examination revealed a swelling localized to the right side of the floor of the mouth and a stone in the right Wharton’s duct that was solid to the touch, yellow and was not adherent to any deeper structure (Figure1).



**Figure 1:** The large stone perforated Wharton’s duct on the right side of the mouth. The apical fragment is missing after attempting stone removal.

The diagnosis of a large salivary calculus of Wharton’s duct was confirmed by an intraoperative X-ray that demonstrated a radiopaque mineralized oval formation about 40 mm long and 9 mm wide located within Wharton’s duct. After an unsuccessful attempt to remove the calculus using traction with forceps (only a small fragment was removed), it was removed by sialodochotomy through an intraoperative approach in the emergency department.

Under regional anesthesia the orifice of the salivary duct was surgically enlarged with a 2-3 cm incision, in this way the large calculus was easily removed (Figures 2-3). After successful removal of the stone the incised mucosa was rotated outward with an everything suture in order to keep the ductal orifice open (marsupialization).



**Figure 2:** The extracted stone appeared yellowish, hard and curved.



**Figure 3:** The calculus (included the fractured, detached segment) measured 40 mm in length, its composition is phosphate salt.

The patient was discharged with antibiotic therapy after 2 hours of observation.

The chemical-physical analysis laboratory showed that the composition of the calculus was phosphate salt as the main component with an organic matrix. At one week follow-up, the symptoms had completely resolved and normal drainage of the submandibular gland was noted.

## Discussion

Sialolithiasis is the second most common disease of the major salivary glands after mumps [2], accounting for approximately 30% of all salivary gland disorders. Approximately 0.01%-1.0% of the population is affected, with a higher incidence in males between the ages of 30 and 60 years (Sialolithiasis: mechanism of calculi formation and etiologic factors [3,4]. The most common location of salivary stones is the submandibular gland.

Calculi generally consist of a mixture of different calcium phosphates together with an organic matrix [5]. Salivary stones

consist of an amorphous mineralized nucleus, surrounded by concentric laminated layers of organic and inorganic substances. The organic components of salivary stones include collagen, glycol proteins, amino acids and carbohydrates. The major inorganic components are hydroxyapatite, carbonate apatite, whitlockite and brushite [6]. The etiologic factors can be classified into two different groups: on the one hand, saliva retention due to morpho-anatomic factors (such as salivary duct stenosis, salivary duct diverticuli, etc.), on the other, saliva composition factors (such as supersaturation, crystallization inhibitor deficit, etc.) [5].

The calculi usually range in size from 1 mm to 10 mm. They have rarely been reported to be larger than 15 mm. The largest sialolith reported in the literature was 70 mm in length and located in Wharton's duct and was described as being the size of a "hen's egg" [7].

It is believed that a calculus may enlarge at the rate of approximately 1 to 1.5 mm per year [8]. Salivary calculi are associated with localized swelling, pain, and infection of the affected gland [1]. The pain typically becomes worse while eating. Large salivary calculi, however, have been reported to remain asymptomatic for many months prior to presentation [9].

Diagnosis of large salivary lithiasis is often made by history and physical examination. Special studies can be used to confirm the diagnosis and plan treatment. Plain radiography will detect opaque stones (80 to 95% of sialoliths), with intra-oral occlusal radiographs particularly useful. Computerized tomography (CT) scanning is more expensive, but has been described as the most accurate non-invasive technique for defining the location of stones. Sialography allows the whole duct system to be visualized, demonstrating calculi of all sizes and also glandular damage from chronic obstruction. Ultrasound provides an excellent, non-invasive method for detecting sialoliths. Stones that are greater than 1.5 mm and of high mineral content are reported to be identifiable on ultrasound with an accuracy of 99% [10]. In cases of clinically evident large sialoliths, ultrasound imaging may aid treatment planning by the detection of additional smallstones.

The methods of treatment for salivary calculi depend on their location, number, shape and size. The goal of treatment is restoration of normal salivary secretion [11]. The methods of treatment of salivary stones include medical management, surgical procedures, lithotripsy and interventional sialo-endoscopy [1].

Medical management consists of hydration, compression and massage, antibiotics for infected glands, anti-inflammatory agents and corticosteroids [12]. Surgical management includes sialodochotomy, which is a well-reported technique for the intra-oral removal of ductal stones, including large calculi. It consists of intra-oral extraction of ductal calculi with a linear incision on the mucosa along the salivary duct, which makes the calculus bulge, facilitating its removal [13].

Usually this procedure is performed under general anesthesia because possible complications include duct stenosis and lingual nerve damage.

Submandibular gland excision through an external approach is the other surgical option recommended in cases of substantial intra-glandular calculi, which are inaccessible via a trans-oral approach. Also, when multiple small stones are present in the vertical portions of Wharton's duct, sialadenectomy is recommended [11]. This approach is also recommended in situations where opening of the duct surgically created recurrent infection of the gland due to ingress of oral contents [8]. Excision of the gland is reported to carry a risk of up to 8% for temporary or permanent marginal mandibular nerve palsy [11]. Surgical methods of stone removal are more effective than medical management [5]. The advent of several minimally invasive techniques for removal of salivary gland calculi have made traditional excision of the salivary gland unpopular. However, several authors still believe that surgical excision of the gland with the calculi remains the gold standard of treatment for intraglandular calculi with an associated non-functional gland [13].

The recently proposed trans-oral removal of palpable hilar sub-mandibular stones by means of extended duct dissection or direct hilar incision (possibly under endoscopic control) now represents one of the main therapeutic options for sub-mandibular stones [14].

In our case we decided to manage the patient with a low invasive-surgical approach without the use of the operating room. The decision of this approach was taken after the analysis of several factors:

1. Patient's compliance
2. In the emergency department the necessary equipment was available: scalpel, bipolar, vacuum cleaner, head-lamp, specific medical and nursing staff
3. Calculus was clearly visible and partially extruded
4. The intraoral incision could be performed with full control of the nervous, vascular structures and submandibular gland as distant from the area of incision

The aim of lithotripsy is to breakdown the calculi into smaller fragments that do not block the flow of saliva and can be washed away. Lithotripsy should be considered as the initial approach in cases of calculi in the submandibular gland, except for calculi localized to the proximal part of Wharton's duct where a surgical approach (marsupialization) combined with medical therapy is preferable [1].

Sialendoscopy is now an established intervention for stone removal, and has been used for large salivary calculi. The incorporation of extracorporeal short-wave lithotripsy with endoscopic removal has also been shown to be an effective modality and an alternative to conventional excision [11].

Patients should be educated regarding the mechanism of their underlying pathology and instructed on methods of maintain-

ing control over them by emphasizing the value of hydration and excellent oral hygiene, which lessens the severity of the attacks and prevents dental complications. A diet rich in proteins and liquids including acidic food and drinks is also advisable in order to prevent the formation of further sialoliths of the salivary glands [15].

## Conclusion

Giant salivary gland calculi are rare; for this reason there are few case reports in the literature. The age, gender, and location of the calculus in our patient are in line with previous reports. However, the size of the calculus is among the largest described. This case demonstrates that even large calculi can be, in specific cases, successfully managed immediately and entirely in the emergency department leading to reduced surgical and anesthetic risks as well as lower costs.

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