Report of 2 Rare Cases of Bilateral Submandibular Sialolithiasis in Patients with Autoimmune Disorders

SUMMARY

Sialolithiasis is a condition characterized by the obstruction of a salivary gland or its excretory duct by calcareous concretions. It is a common disease of major salivary glands and mainly affects the submandibular gland. Bilateral submandibular sialolithiasis, however, is a rare condition. No systemic disorders, with the exception of gout, were found to be related to salivary stone formation.

We present 2 patients presenting with bilateral submandibular sialolithiasis. In the first case, the patient was treated with removal of the submandibular gland on the left side and removal of the salivary stone, intraorally, on the right side. In the second case the patient underwent bilateral submandibular gland removal due to the location of the stones. However, no decrease in salivary flow was noted following the operation. We also review the literature on submandibular sialolithiasis.

Keywords: Sialolithiasis, bilateral; Submandibular Salivary Gland, removal; Sialadenitis

Introduction

Salivary glands are divided into major and minor salivary glands. The rate of secretion of individual glands ranges from barely perceptible during sleep to as high as 4 ml/min on maximal stimulation. It has been reported that of total salivary secretion (1.5 l/day), approximately 75% is secreted by the submandibular glands, 20% by the parotid glands and the rest by sublingual and other salivary glands1. Furthermore, salivary secretions from individual types of glands differ. The parotid gland secretes protein rich saliva with enzymes like amylase, while the submandibular gland secretes mucin rich saliva, which is useful in lubricating the bolus of food before ingestion2.

Sialolithiasis is a condition characterized by the obstruction of a salivary gland or its excretory duct due to the formation of calcareous concretions. It is a common disease of the major salivary glands. It mainly affects the submandibular gland (80-90%) and to a lesser degree the parotid gland (5-20%). The sublingual and the minor salivary glands are rarely affected. It may occur at all ages but there is a peak incidence during the 4th, 5th and 6th decades3.

The classic treatment of sialolithiasis includes antibiotics and inflammatory agents, hoping for a spontaneous stone expression through the papilla. In cases of anterior located submandibular stones, a sialodochoplasty is the treatment of choice, whereas submandibular gland removal is indicated if the sialoliths are not accessible intraorally. In cases of posterior located or intraglandular parotid stones, a conservative approach is usually adopted, probably because parotidectomy for infectious conditions is associated with a high incidence of facial nerve complications4. External lithotripsy, initially described by Iro et al5, is becoming popular and success rates up to 75% for the parotid and up to 40% for submandibular gland are reported6. Sialoendoscopy is also a relatively recent technique for the removal of salivary stones. Other techniques for salivary stones fragmentatin using electrohydraulic and pneumoblastic devices have been described, but their use is not recommended due to injuries of the canal wall that have been reported7,8.

It has been found on autopsy that 1% of the population has sialolithiasis9. However, bilateral sialolithiasis is an extremely uncommon condition. In Tholen’s series of 97 cases of sialolithiasis, only 1 was bilateral10, whereas in Levy’s series, only 4 patients among 180 had bilateral sialolithiasis11. As far as the
submandibular gland is concerned, Mela et al\textsuperscript{12} found only 3 cases of bilateral submandibular sialolithiasis among 245 patients. Lustmann et al\textsuperscript{13} reported an incidence of 1\% of bilateral submandibular sialolithiasis in autopsy materials. We present 2 rare cases of bilateral submandibular sialolithiasis with concurrent sialadenitis.

### Report of 2 Cases

#### Case 1

A 40-year-old male came to our department complaining of a history of recurrent swelling in the submandibular region on both sides. Past medical history revealed psoriasis, whereas family history was unremarkable. Review of systems revealed no pathologic findings and vital signs were within normal limits. On physical examination, a mild swelling of the submandibular regions was discernible, which felt firm and revealed no tenderness on palpation. The overlying skin was freely mobile and no signs of cellulitis were present. On bimanual palpation calculi were palpable in both submandibular ducts.

The mandibular occlusal radiograph and panoramic radiograph showed calcifications within the left submandibular gland and radiopacities in both submandibular ducts. A CT scan was ordered to better delineate the size and extent of calcifications (Fig. 1).

A diagnosis of bilateral submandibular gland sialolithiasis with obstructive sialadenitis was made. The patient was placed on IV clarithromycin and was scheduled for left submandibular gland excision and right transoral stone removal with concurrent sialodochoplasty. The operation was carried out under general anaesthesia via nasotracheal intubation. On the right side, a longitudinal incision was made over the calculus, exposing it. The calculus was removed and a sialodochoplasty was performed (Fig. 2). The incision on the left side was made over a natural skin crease below the lower border of the mandible (Fig. 3). The submandibular gland was dissected out without complications. Healing occurred uneventfully.

#### Case 2

A 44-year-old female was referred for evaluation and treatment of submandibular swelling. The patient complained of submandibular swelling accompanied by severe pain below the right side of her jaw at mealtime. Her medical history revealed a 16-year history of Systemic Lupus Erythematosus (SLE) and mild to moderate hypertension, which was effectively controlled by medication. Review of systems was remarkable for mild hypertension. Laboratory exams revealed elevation of WBC count (15.2x10\(^9\)\/L) and positive ANA, antiSSA, antiSm and antiRNP antibodies due to her systemic disease.
On physical examination the right submandibular area was markedly oedematous, firm and tender. A calculus was palpable intraorally along the course of the right submandibular duct. The left submandibular gland and duct revealed no pathologic findings on palpation. Imaging consisting of panoramic radiograph and CT scan revealed bilateral submandibular sialolithiasis (Fig. 4).

Our patient had no symptoms in the left submandibular region. We informed her, however, that if the sialolith remained in place it would probably cause the same symptoms of obstructive sialadenitis that she had experienced on her right submandibular gland, and that another operation would be indicated to remove the gland. For all the reasons mentioned above, the patient agreed to have both the submandibular glands removed simultaneously. In view of the inaccessibility of the stones to simple intraoral surgery, bilateral submandibular gland removal was undertaken via nasotracheal intubation (Fig. 5).

The specimens were sent for histopathologic evaluation. The affected lobules of the right submandibular gland showed intralobular fibrosis and a considerable amount of chronic inflammatory cells infiltration. These findings confirmed the diagnosis of chronic sialadenitis. Microscopic examination of the left submandibular gland revealed mild inflammatory changes centred around small and large ducts.

The patient recovered uneventfully and 12 months later she was free of further symptoms. Despite the fact that both her submandibular glands were removed, she did not complained of decreased salivary flow or of any difficulties in chewing, speaking or swallowing.

Figure 4. Axial computed tomography demonstrating large bilateral calcifications in the salivary ducts

Figure 5. Bilateral incisions below the lower border of the mandible can be seen

Discussion

Sialolithiasis is the most common disease of the salivary glands in the middle-aged patients. It occurs mainly in the submandibular gland (80-90%) and to a lesser degree in the parotid gland (5-20%)3. Calculi formation is more common in submandibular gland because of its longer duct, its slower rate of saliva flow, its viscous secretions of high alkalinity and its high calcium content of saliva14.

The nucleus of the salivary stones is inorganic and is mainly composed of calcium carbonate. Around this nucleus, organic and inorganic substances accumulate in a laminated concentric fashion15. Submandibular stones are thought to form around a nidus of mucous, whereas parotid stones are usually formed around a nidus of inflammatory cells or around a foreign body16. It is thought that stagnation of calcium rich saliva, combined with unclear metabolic events, contribute to calculus formation17. Risak18 was the first to support that bacteria of the oral flora are associated with stone formation and Teymoortash et al15 identified oral bacteria (especially streptococcus species) in sialoliths. They state that these bacteria play a potential role in the etiopathogenesis of sialolithiasis.

Sialolithiasis occurs in the right and left submandibular gland with the same frequency, but simultaneous sialolithiasis in more than 1 gland is a rarity. That is because sialolithiasis is probably caused by local factors and not by systemic disorders. Past medical history and laboratory exams of both our patients revealed no evidence of hypercalcaemia or hyperparathyroidism or any other metabolic or hormonal disturbance. In fact, in reviewing the literature, gout is the only systemic disease
that is known to cause sialolithiasis. In this case the salivary stones are radiolucent because they are mainly composed by uric acid\(^7\). Disturbances in the viscous secretions of the salivary glands occur in patients with cystic fibrosis, although it is not known if these patients have an increased prevalence of sialolithiasis\(^8,9,10\). The coexistence of sialolithiasis and nephrolithiasis has also been mentioned by some authors\(^11,12\), but we found no correlation between sialolithiasis and autoimmune diseases, such as psoriasis and lupus in the literature.

Conventional radiographs, such as panoramic radiograph and mandibular occlusal radiograph, reveal submandibular sialolithiasis in 80-94.7\% of cases\(^13\). Ultrasound is helpful especially in parotid sialolithiasis since it detects 90\% of stones greater than 2 mm. High resolution CT scan has proven superior to conventional radiographs and ultrasound in detecting sialolithiasis, because its sensitivity makes it possible to detect recently calcified calculi which cannot be detected through conventional radiography\(^14,15\). This is the reason why it is advisable to request it always before surgical intervention.

The most common symptom of sialolithiasis is swelling followed by pain, which usually correlates with meals. The increased salivary secretion in the obstructed gland leads to an elevated intraglandular pressure, which causes pain to the patient\(^16\). Since obstruction is rarely complete, the gland swelling will subside to some degree, but in chronic gland obstruction, some swelling and induration may persist even after stone removal due to inflammation and fibrosis of the gland.

Bilateral submandibular sialadenitis may be due to obstructing calculi, low grade bacterial infection or a lympho-epithelial disease such as Sjögren syndrome\(^17\). If complete obstruction has occurred, local cellulitis may be present. The superficial layer of deep cervical fascia, which envelops the gland, limits the spread of infection. However, prolonged swelling and inflammation process can weaken the fascia and allow the infection to spread into the submandibular space\(^18,19\). The progression of infection from both glands caused by bilateral submandibular sialolithiasis, coupled with oedema of the floor of the mouth, can form a picture that is consistent with Ludwig’s angina and carries the same morbidity\(^20,21\).

The treatment of choice of the submandibular gland sialolithiasis is the removal of the obstructing stone by an intraoral approach. Stones in the anterior or middle third of the submandibular duct can be removed with an intraoral approach\(^22\). Sialendoscopy may be curative if the stone is located more distally in the duct\(^23\). While the mainstay of treatment of sialolithiasis is surgery, some authors implement a trial of conservative therapy, consisting of gland massage, adequate hydration and anti-staphylococcus antibiotics, especially when the stones are inaccessible to intraoral approach\(^24,25,26\). They state that the conservative approach is preferred to the extraoral gland removal because of the high risk of damage to nerves, such as the lingual, hypoglossal and facial nerve, during the operation and because of the visible scar that is left. On the other hand, one should keep in mind the complications that can result from recurrent sialadenitis. If an acute sialadenitis occurs, there is a danger of abscess formation, which can result in sinus and scar formation far more unsightly than the surgical scar. Furthermore, longstanding recurrent sialadenitis leads to formation of fibrous adhesions and distortion of normal anatomy\(^27\). Removal of the gland, if it is eventually needed, is far more complicated and carries a greater risk of causing damage to the regional nerves. Furthermore, Preuss et al\(^28\) have recently analyzed 258 unselected submandibular excisions and they found a low rate of transient palsies of the mandibular branch of the facial nerve (9\%) and lingual nerve (2\%). To date, it is not known that routine facial nerve stimulation devices could further reduce the risk of transient nerve palsy.

As we mentioned before, new endoscopic techniques allow the conservative removal of salivary stones in a considerable number of cases. However, it remains unclear whether the incidence of complications is lower in comparison with conventional surgical excision of the gland. Analysis of complications of interventional sialoendoscopy in a large series of patients revealed no major complications, but the long-term success rate of this procedure is still unknown\(^29\), although in the case of parotid sialolithiasis, endoscopy has proven to be superior to other techniques\(^30,31\). Recently, shock wave lithotripsy was introduced for the treatment of sialolithiasis. Shock waves crush the calculus \textit{in situ} and the fragments are rinsed through the duct by salivary flow. The results show that 40-50\% of patients are rendered stone free\(^32\), although the overall success rate in patients with parotid gland calculi was far better than in patients with submandibular sialolithiasis\(^33\). Furthermore, lithotripsy is performed with special and expensive equipment, which is not available in our hospital and this is the reason why we did not consider treating our patients with this technique. If these new procedures prove to be safe in long-term studies, they certainly promise a successful conservative treatment option for a large number of patients with submandibular sialolithiasis.

\section*{References}


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