

CASE REPORT

Multiple bilateral submandibular gland sialolithiasis

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Abstract

Sialolithiasis accounts for the most common etiology of salivary gland obstruction which leads to recurrent painful swelling of the involved gland which often exacerbates while eating. Stones may be encountered in any of the salivary glands but most frequently in the submandibular gland and its duct. Simultaneous sialolithiasis in more than one salivary gland is rare, occurring in fewer than 3% of cases. Seventy to 80% of cases feature solitary stones; only about 5% of patients have three or more stones, the case report which we are presenting here had three submandibular sialoliths involving both the submandibular glands which were removed by intraoral approach and no post-operative complications were noted.

Key words: Bilateral, multiple, sialolithiasis, submandibular

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Introduction

Sialolithiasis is the second most common salivary gland disease; the most common disease of submandibular glands in middle-aged adults, incidence being 12 in 1,000 of the adult population. Males are affected twice as much as females.^[1] There is no left or right predominance.

Submandibular glands are most commonly involved. Intraductal stones occur more commonly than intraglandular stones.^[2] Simultaneous sialolithiasis in more than one salivary gland is rare, occurring in fewer than 3% of cases. Seventy to 80% of cases feature solitary stones; only about 5% of patients have three or more stones,^[3] the case report which we are presenting here had three submandibular sialoliths involving both the submandibular glands.

The stones themselves are typically composed of calcium phosphate or calcium carbonate in association with other salts and organic material such as glycoproteins, desquamated cellular residue, and mucopolysaccharides. Bacterial elements have not been identified at the core of a sialolith.^[4] Some factors inherent to the submandibular

gland tend to favor stone formation there like longer and larger caliber duct, flow against gravity, slower flow rates, and higher alkalinity along with higher mucin and calcium content of the saliva. The submandibular gland hosts the largest stones with the largest reported one being 6 cm in length. Most submandibular stones are found in the salivary duct (75-85% of cases). Hilar stones tend to become very large before becoming symptomatic. Ductal stones are elongated in shape whereas hilar stones tend to be oval.^[3]

Case Report

A 22-year-old female patient reported to Department of Oral and Maxillofacial Surgery with a complaint of pain in the floor of the mouth since 6 months aggravated during meals. On examination by bimanual palpation of the floor of the mouth firm to hard mass was felt in the region opposite to 33 and 34 and another mass was felt opposite to 35 and 36 [Figure 1]. The patient was further sent to radiological investigation, occlusal radiograph confirmed the clinical diagnosis of submandibular sialolithiasis, with

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Figure 1: Pre-operative intra oral photograph showing a bulge at the region of left Wharton's duct opening



Figure 2: Pre-operative occlusal radiograph showing 3 sialoliths, two on the left and one on the right

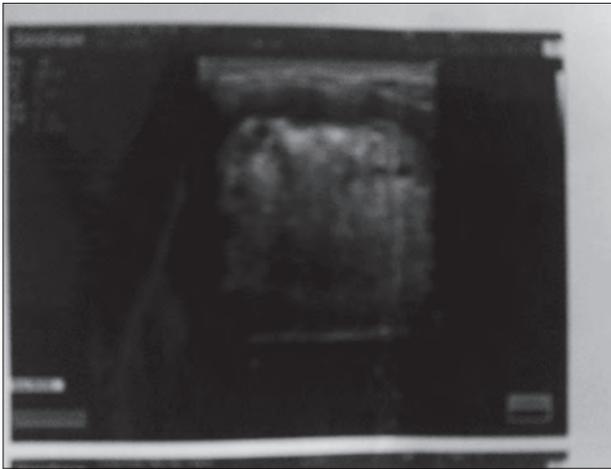


Figure 3: Ultrasonography

another sialolith in the right submandibular salivary gland duct [Figure 2]. Ultrasonography report proved the same [Figure 3].

The sialolith present opposite to 33 and 34 was removed under local anesthesia with adrenaline with the incision placed along the long axis of the duct as it was very close to the ductal orifice and superficial [Figure 4]. While the other two sialoliths were planned under general anesthesia by intraoral approach and were removed successfully [Figures 5-7], post-operative occlusal radiograph was clear with no sialoliths left out [Figure 8]. A follow up of the same patient after three months showed no recurrence.

Discussion

It is believed that salivary calculi develop as a result of deposition of mineral salts around a nidus of bacteria, mucus, or desquamated cells. Salivary stagnation, increased alkalinity of the saliva, increased calcium content of the saliva, infection or inflammation of the salivary duct or



Figure 4: Intra operative photograph showing excision of sialolith on right side

gland, and physical trauma to the salivary duct or gland may predispose to calculus formation.

Diagnosis of sialolithiasis is easy due to simple and obvious clinical features, but sometimes sialolithiasis of the submandibular gland can be completely asymptomatic. Common symptoms vary from a painless swelling, moderate discomfort to severe pain with large glandular swelling accompanied by trismus and usually associated with eating. Anyway, in order to establish the right treatment, imaging studies are always necessary. Ultrasonography is the simpler method, which demonstrates the sialolith with high accuracy. It has also been reported that sialoliths smaller than 3 mm may not be detected during ultrasonographic examination, as they will not produce acoustic shadows. Digital sialography and subtraction sialography have increased the sensitivity and specificity of conventional sialographic technique, which are considered the gold standard. The major advantage of these newer techniques is the production of an image without the superimposition of overlying anatomical



Figure 5: Intra operative photograph showing excision of sialolith on left side



Figure 6: Excised sialoliths



Figure 7: Post-operative intra oral photograph showing good healing

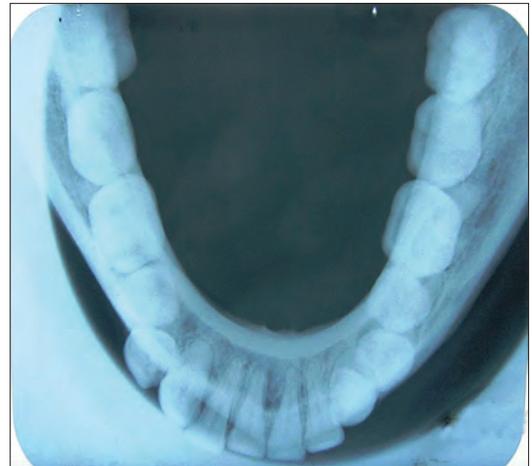


Figure 8: Post-operative occlusal radiograph showing no sialoliths

structures; the disadvantage being the need to use contrast agents that simulates conventional sialography. These agents may expose the patient to radiation hazards, cause pain associated with the procedure, perforate the duct's wall, and may be contraindicated during acute infections. Based on a review of the literature most of the sialoliths are usually of 5 mm in maximum diameter and all the stones over 10 mm should be reported as a sialolith of unusual size.^[5]

The algorithm for the treatment of sialolithiasis depends upon the location and size of the sialolith. Patients presenting with sialolithiasis may benefit from a trial of conservative management, especially if the stone is small. The patient must be well-hydrated and the clinician must apply moist warm heat and gland massage, while sialogogues are used to promote saliva production and flush the stone out of the duct. With gland swelling and sialolithiasis, infection should be assumed and a penicillinase-resistant anti-*Staphylococcal* antibiotic prescribed. Most stones will respond to such a regimen, combined with simple sialolithotomy when required.^[6]

Sialodochoplasty can be performed to remove the submandibular sialoliths, which are located close to the orifice of Warthin's duct. To remove the stones distal to the punctum, a transverse incision can be made distally on the stone taking care not to injure the lingual nerve. In the management of large sialoliths, which are located in the close proximal duct, extracorporeal shock wave lithotripsy (ESWL) can be considered. Endoscopic intracorporeal shock wave lithotripsy (EISWL) is also gaining importance because of less damage to the adjacent tissues during the procedure. Sialadenoscopy, which is a non-invasive technique, can be used to manage large sialoliths as well as ductal obliteration. CO₂ laser, because of its advantages of minimal bleeding, less scarring, clear vision, and minimal postoperative complications, is gaining its popularity in the treatment of sialolithiasis.^[6]

Conclusion

Though various diagnostic and treatment variabilities are available, still the conventional surgical removal holds good,

here is a case report of a patient with multiple submandibular sialoliths involving both the submandibular glands which is a rare occurrence treated surgically with no post-operative complications.

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