Sialodocholithiasis - A Case Report and Review

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Abstract

A majority of sialoliths occur in the submandibular gland or its duct and are a common cause of acute and chronic infections. This report describes the case of a patient who had an unusual submandibular gland duct sialolith (calculus) that was completely obstructing the submandibular gland duct (sialodocholithiasis) and the use of the computed tomography and ultrasonography as a diagnostic aid and a surgical guide. Patients with sialolithiasis require definitive surgical treatment in most cases, which results in an excellent prognosis. Along with presenting the case report, this article also reviews the etiology, diagnosis, and various treatment modalities available for the management of salivary gland calculi depending on their site and size.

Keywords: Calculi, duct obstruction, salivary gland, sialography, submandibular gland

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Introduction

Sialolithiasis is the most common disease of the salivary glands and is the major cause of salivary gland dysfunction along with acute and chronic infection. For the diagnosis of sialolithiasis, a careful history and examination are important criteria that are to be considered. The patient should also give a positive history of pain and swelling in response to the concerned gland and also in response to salivary stimuli. In the case of complete obstruction, constant pain and swelling along with pus discharge and signs of systemic infections are seen.[1]

Although large sialoliths have been reported in the body of salivary glands, they have been rarely been reported in the salivary ducts.[2-4]

Gout is the only systemic illness known to predispose to salivary stone formation[5] although in gout, the stones are made predominantly of uric acid.[6]

The anatomic location, long, torturous duct course, and a narrow orifice are the major causes of the submandibular gland being affected. Along with these factors, alkaline saliva rich in mucin and increase in the concentration of calcium and phosphate also contributes to the stone formation.[7,8] In addition, the submandibular duct is longer and the gland has an antigravity flow.[9] Stone formation is not associated with systemic abnormalities of calcium metabolism.[6]

Salivary calculi are usually unilateral and are not a cause of dry mouth.[10] Clinically, they are round or ovoid, rough or smooth, and of a yellowish color. They consist of mainly calcium phosphate with smaller amounts of carbonates in the form of hydroxyapatite, with smaller amounts of magnesium, potassium, and ammonia. This mix is distributed evenly throughout.[6] Submandibular stones are composed of 82% inorganic and 18% organic material as compared with the parotid stones, which are composed of 49% inorganic and 51% organic material[4] that is composed of various carbohydrates and amino acids.[6]

The exact etiology and pathogenesis of salivary calculi is largely unknown. Genesis of calculi lies in the relative stagnation of calcium-rich saliva. They are thought to occur because of the deposition of calcium salts around an initial organic nidus, consisting of altered salivary mucins, bacteria, and desquamated epithelial cells.[10,11] For stone formation, it is likely that intermittent stasis produces a change in the mucoid

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element of saliva, which forms a gel. This gel produces the framework for the deposition of salts and organic substances creating stone.[6]

Traditional theories suggest that the formation occurs in two phases: a central core and a layered periphery.[12] The central core is formed by the precipitation of salts, which are bound by certain organic substances. The second phase consists of the layered deposition of organic and nonorganic material.[13] Submandibular stones are thought to form around a nidus of mucous,[1] whereas parotid stones are thought to form most often around a nidus of inflammatory cells or a foreign body.[1,14,15] Another theory has proposed that an unknown metabolic phenomenon can increase the saliva bicarbonate content, which alters calcium phosphate solubility and leads to the precipitation of calcium and phosphate ions.[6,16] A retrograde theory for sialolithiasis has also been proposed. Aliments, substances, or bacteria within the oral cavity might migrate into the salivary ducts and become the nidus for further calcification.

Many studies reveal that males are more commonly affected than females. Persons in their middle age are more affected in the age group of 42–58 years. The submandibular gland (>80%) is most commonly involved, followed by parotid (6%), sublingual (2%), and minor salivary glands.

Bimanual palpation of the floor of the mouth, in a posterior to anterior direction, reveals a palpable stone in a large number of cases of submandibular calculi formation. Bimanual palpation of the gland itself can be useful, as a uniformly firm and hard gland suggests a hypofunctional or nonfunctional gland.[6] For parotid stones, careful intraoral palpation around Stensen's duct orifice may be beneficial.[6] Deeper parotid stones are often not palpable. When minor salivary glands are involved, they are usually in the buccal mucosa or upper lip, forming a firm nodule that may mimic tumor.[6,17]

Imaging studies are very useful for diagnosing sialolithiasis. Occlusal radiographs are useful in showing radiopaque stones. It is very uncommon for patients to have a combination of radiopaque and radiolucent stones.[18] Forty percent of parotid and 20% of submandibular stones are not radiopaque. For the patients showing signs of sialadenitis and those with deep parotid and submandibular stones, sialography is very beneficial. Sialography is, however, contraindicated in acute infection or in significant patient contrast allergy.[6] The conventional diagnostic radiographic techniques for salivary gland pathology are routine radiographs (occlusal and panoramic), sialography, ultrasound, xeroradiography, scintigraphy, and CT. These techniques (indirect) provide partial information concerning the presence of calculi and the glandular status.

Case Report

A 52-year-old male patient, with noncontributory cultural, social, and medical histories, reported with a chief complaint of pain in the left lower back region of the jaw for the past 6–7 days, which was dull, intermittent in nature, subsiding at night, and appearing back in the morning on brushing. The patient also gave a history of severe pain before having meals. Patient had a habit of chewing tobacco 2–3 times a day for the past 30 years.

On extraoral examination of the left submandibular region [Figure 1], small, diffuse swelling was noticed with no change in color of the overlying skin. On palpation, a firm, fixed, nontender, nonfluctuant swelling was felt. The swelling measured around 3 cm × 2.5 cm approximately. Intraoral examination of the floor of the mouth [Figure 2] revealed no significant findings. On milking the left submandibular gland, pus discharge from the submandibular duct was noticed. On hard-tissue examination, there were generalized attrition and cervical abrasions.

After a history and clinical examination of the patient, a provisional diagnosis of acute bacterial sialadenitis was made.

The patient was advised to get a left mandibular occlusal radiograph done.

The left mandibular occlusal radiograph [Figure 3] revealed a well-defined oval radiopacity, measuring around 4 cm × 2 cm in the floor of the mouth in relation to the 37 and 38 tooth region.

The patient was then advised for computed tomography (CT) scan (plain and contrast-enhanced CT) of the left submandibular gland and duct along with the ultrasonography (USG) of the neck.

CT scan of the left submandibular region [Figures 4 and 5] revealed enlarged and swollen left submandibular gland (30 cm × 23 cm), showing a mild heterogeneous enhancement on the postcontrast study. A radio-opaque oval calculus (11 mm × 6 mm) was evident in relation to the left submandibular region along the ductal region. Left submandibular lymphadenopathy was also noticed.

Figure 1: Compare swelling in left and right submandibular region

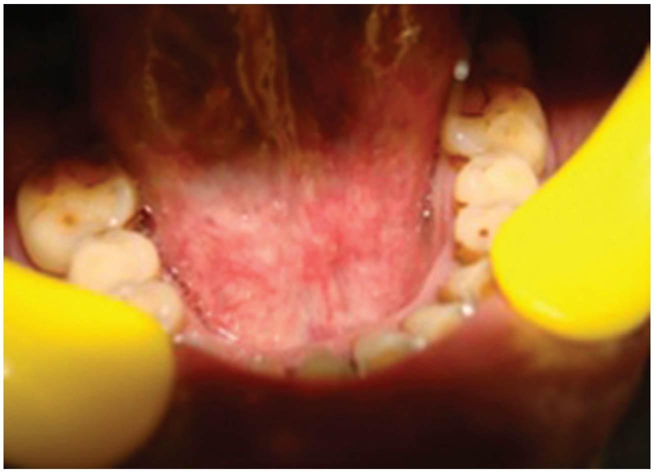

Figure 2: Intraoral view showing floor of the mouth

Figure 3: Occlusal radiograph showing salivary stone in relation to submandibular ductal region

Figure 4: Plain study computed tomography showing radiopaque salivary duct calculi

Figure 5: Intravenous contrast study computed tomography showing radiopaque salivary duct calculi

USG of the neck revealed [Figure 6] a large 11.3-mm radiodense shadow in the left submandibular region along with a small ill-defined hypoechoic area measuring around 15.0 mm × 15.3 mm, which was noticed in the left submandibular gland, indicating the possibility of calculus in the duct of the left submandibular gland causing obstructive inflammatory lesion in the gland.

Based on the history, clinical examination, radiographic examination, and advanced imaging techniques, the final diagnosis of sialodocholithiasis was made.

The patient was then surgically operated for the stone in the submandibular duct under local anesthesia without the removal of the submandibular salivary gland. After taking the radiograph, the specimen was sent for histopathological examination [Figure 7].

Treatment

Patients presenting with sialolithiasis may benefit from a trial of conservative management, especially if the stone is small.[6] 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 antistaphylococcal antibiotic prescribed.[1,14]

Almost half of the submandibular calculi lie in the distal third of the duct and are amenable to simple surgical release through an incision in the floor of the mouth, which is relatively simple to perform and not usually associated with complications.[19] If the stone is sufficiently forward, it can be milked and manipulated through the duct orifice. This can be done with the aid of lacrimal probes and dilators to open the duct. Once open, the stone can be identified, milked forward, grasped, and removed. This is then followed by the milking of the gland to remove any other debris in the posterior portion.[6]

The duct may need opening to retrieve the stone. This involves a transoral approach where an incision is made directly on the stone. In this way, more posterior stones, 1–2 cm from the punctum can be removed by cutting directly on the stone

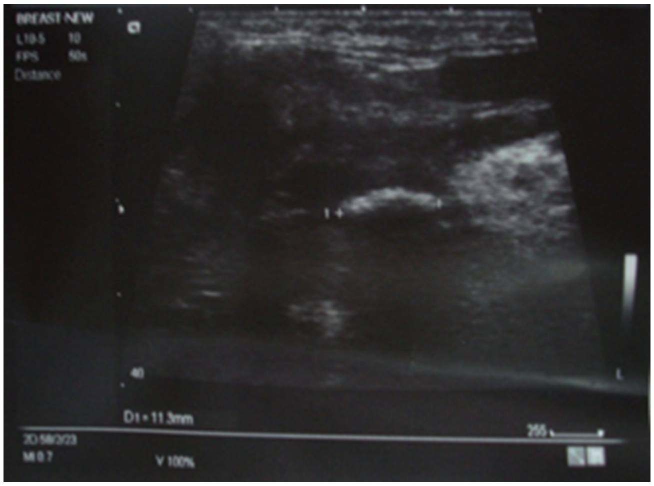

Figure 6: Ultrasonography of the submandibular duct region

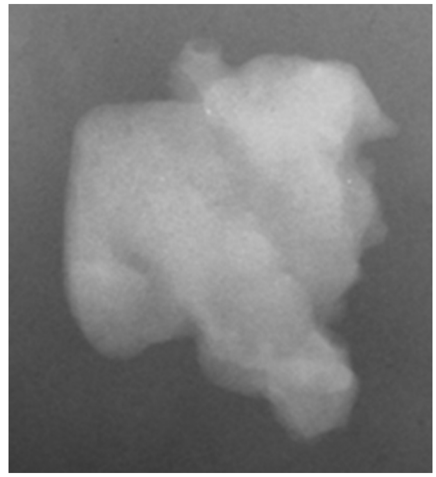

Figure 7 : Postoperative radiograph of the salivary calculi specimen

in the longitudinal axis of the duct. Care should be taken as the lingual nerve lies deep, but in close association with the submandibular duct posteriorly. Subsequently, the stone can be grasped and removed. No closure is done leaving the duct open for drainage. If the gland has been damaged by recurrent infection and fibrosis, or calculi have formed within the gland, it may require removal.

The accelerated use of endoscopy in various surgical fields, such as in the renal and biliary ducts, has enhanced the adoption and application of such techniques to major salivary glands.[20] Katz[21] first introduced a flexible mini-endoscope into the ductal system of the major salivary glands. Nahlieli and Baruchin[20] reported the usage of a rigid miniendoscope for the same purpose. Sialoendoscopy is a minimally invasive technique for the removal of calculi from salivary glands as well as an excellent diagnostic procedure.[20] Königsberger *et al*. and Yoshizaki *et al*.[22] reported that endoscopically controlled intracorporeal lithotripsy of salivary stones can be effectively used as a noninvasive therapy for sialolithiasis. Azaz *et al*.[23] and Lustman *et al*.[24] reported that treatment with CO_2 laser yields excellent results, with almost no bleeding, minimal scarring, clear vision of the operating site, noncontact surgery, carried out during acute stages, without spreading of infection, minimal postoperative pain and edema, and little discomfort through the healing period. However, Iro *et al*. and Tro *et al*.[25,26] reported that extracorporeal piezoelectric shock wave therapy seems likely to be a safe, comfortable, and effective minimally invasive nonsurgical treatment for salivary stones. Haring[27] reported that a salivary stone should always be removed. Lustman *et al*.[24] also reported the treatment of the obstructing stone by an intraoral surgical approach. The present case was surgically excised under local anesthesia.

Alternative methods of treatment have emerged such as the use of extracorporeal shock wave lithotripsy, and more recently, the use of endoscopic intracorporeal shockwave lithotripsy, in which shockwaves are delivered directly to the surface of the stone lodged within the duct without damaging adjacent tissue (piezoelectric principle).[25] Both extracorporeal and intracorporeal lithotripsy are gaining increasing importance in the treatment of salivary stone disease.[28] In extracorporeal piezoelectric lithotripsy, the average size of fragments produced is about 0.7 mm.[29] Extracorporeal salivary lithotripsy provides another therapeutic option that carries fewer risks than surgical removal of the affected gland, such as the risks of a general anesthetic, facial nerve damage, surgical scar, Frey's syndrome, and causes little discomfort to the patient while preserving the gland.[25]

In some cases, excision of the entire gland is required to prevent multiple recurrent episodes. If the gland is infected, the infection should be eliminated with antibiotics before surgical removal. According to numerous reports, even if only the submandibular gland of one side was removed, the salivation within the oral cavity was reduced, and thus, oral hygiene may be deteriorated.[30] According to reported studies, the patients underwent the transoral removal of submandibular stones, function of the submandibular gland was assessed by scintigraph, and it was found that after the removal of stones, 75% submandibular glands recovered normal functions.[31,32]

Conclusion

The conventional techniques retain their popularity as compared to the various advanced diagnostic and treatment modalities because of their easy to use techniques. Submandibular gland removal may be indicated following the failure of advanced techniques. Our case was a middle-aged patient with the stone in the duct of the submandibular gland unilaterally. The removal of stone/calculi was performed by the intraoral removal of submandibular stone while preserving the salivary gland.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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