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Case Report

HILAR SIALOLITH OF THE SUBMANDIBULAR GLAND – A CASE REPORT

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ABSTRACT

Salivary stones also known as sialoliths are calcified deposits in the salivary glands. Sialolithiasis is the most common disease affecting the salivary glands. Submandibular glands being the most common site, the sialolith near the hilum is much rarer as compared to the intraductal ones. Salivary stones consist of an amorphous mineralised nucleus, surrounded by concentric laminated layers of organic and inorganic substances. The organic components of salivary stones include collagen, glycoproteins, amino acids and carbohydrates. The major inorganic components are hydroxyapatite, carbonate apatite, whitlockite and brushite. In this case report, we present a case of a 26-year-old male who complained of prandial pain and swelling, with no evidence of calcified mass on imaging. Multiple sialoliths were present on surgical approach near the hilum of the gland.

Key words: Salivary stones, Sialoliths, Sialolithiasis, Submandibular gland, Hilum of the gland.

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INTRODUCTION

The term sialolithiasis derived from the Greek words, sialon meaning saliva and lithos meaning stone [1]. More than 80% of the sialoliths occur in the submandibular gland or its duct, 6% in the parotid gland and 2% in the sublingual gland or minor salivary glands. Salivary gland/duct stones or sialoliths are calcifications that accumulate within the salivary gland parenchyma and associated ductal systems [2]. They develop from a mineralization nucleus of debris including bacterial colonies, shed ductal epithelial cells, cell remnants, mucus plugs and foreign bodies.

Sialolithiasis is the most common disease of the salivary glands. It is estimated that its incidence is 12 per 1000 adult individuals [3, 4]. Sialolith constitutes deposition of calcium-rich salts around a central nidus, which may consist of desquamated epithelial cells, foreign bodies, or bacteria and their decomposition products [5]. Most patients present with a single stone but multiple stones occur in 32 per cent of cases in the parotid gland and 22 per cent in the submandibular gland [6]. Bilateral stones occur in around 2.2 per cent of cases. Sialoliths are typically more common in middle-aged males but some studies suggest a male to female ratio of 1:1 and with ages ranging from 12 to 93 years [7]. The most frequent clinical presentation is swelling and pain in the area of the affected gland [2, 6].

Sialoliths can often be detected on palpation, especially when they are located above the mylohyoid muscle or in the buccal mucosa and lip [1, 2]. The

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submandibular gland system is more susceptible to sialolithiasis because of anatomic and physiological features of the gland itself, where saliva is more alkaline and presents higher concentration of calcium and phosphate in the form of carbonates, besides higher amounts of mucin, providing higher viscosity and favoring adherence around foreign bodies [8]. Patients with sialolithiasis involving the duct of a major salivary gland may complain of moderate to intense pain, particularly at mealtimes, due to the increased salivary flow rate, associated with enlargement of the gland.

The duct occlusion blocks or decreases the free flow, leading to saliva accumulation in the gland, which under pressure causes pain and swelling. Depending on the calcification degree of the calculi, it can be visible in conventional radiographs, being radiopaque and observable at any point of the duct or inside the gland itself. Calculi in the terminal portion of the submandibular main duct are better visualized through occlusal radiography. In panoramic or periapical radiographs, the calcification image may appear superimposed on the mandible; therefore, it may be mistaken for an intrabony lesion [9].

Based on the calcification degree of certain lesions, not all calculi can be visualized in conventional radiographic examinations; in this case, other imaging examinations may be necessary such as sialography, ultrasound, computed tomographic scan, and magnetic resonance imaging [10-12]. In the presence of sialolithiasis, the sialography examination shows interruption in the contrast image or image straightening [13]. Hilar stones tend to become very large before becoming symptomatic. Ductal stones are elongated in shape whereas hilar stones tend to be oval [14]. In this case report we present a 26-year-old male patient who reported to our department with a chief complaint of prandial pain and with no obvious clinical pathology.

CASE REPORT

A 24-year-old male patient reported to the dental OP with a chief complaint of pain and recurrent swelling in his right lower jaw region for past 6 months. Patient gave a history of pain associated with the swelling. Swelling appeared and increased in size during meal time. Spontaneous regression followed by reappearance during meal time was noticed.

On intraoral examination, the floor of the mouth appeared elevated on the right side. (Figure - 1) On bimanual palpation tenderness was evident in the right submandibular gland. Reduced salivary flow was evident through the orifice of the Wharton's duct on the right side.

A provisional diagnosis of obstructive sialadenitis of the right submandibular gland was considered and the patient was subjected to conventional imaging modalities (Figure - 2) and ultrasonography (Figure - 3) of the region of the Wharton's duct which did not reveal any abnormality or any calcified mass in the region of the submandibular gland or along the course of its duct.

A Sialogram (Figure - 4) was advised, following cannulation of the duct and injection of contrast media a lateral view radiograph was taken which showed a filling defect in relation to the course of the submandibular duct and did not reveal evidence of any calcified mass. It returned with a report stating imaging features in favour of soft calculus (radiolucent) in proximal portion of right Wharton's duct with obstructive features.

A surgical procedure was planned and the Wharton's duct was sequentially explored beginning from the proximal portion extending till the gland. Multiple calcified sialoiths less than 5 mm was found near the hilum of the gland (Figure - 5). Thus, a diagnosis of sialolithiasis was given and the specimen was sent for histopathological evaluation. Clinical features and surgical evidence of silaoliths lead to a final diagnosis of Sialolithiasis of the right submandibular gland. Patient is presently under follow-up and has not complained of any recurrent symptoms.

Fig 1. (Mirror image) Right side of the floor of the mouth presenting with an elevated appearance.

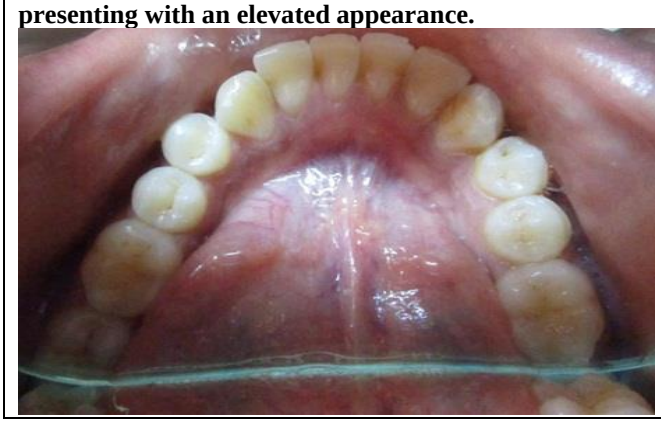


Fig 2. Mandibular occlusal view shows no evidence of calcification

Fig 3. Utrasonogram of the right submandibular gland showing a normal glandular architecture

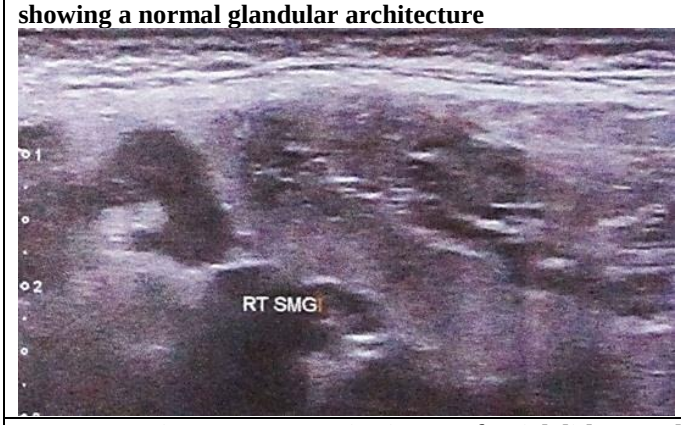


Fig 4. Sialogram showing a filling defect distal to the hilum of the gland with no evidence of calcified mass

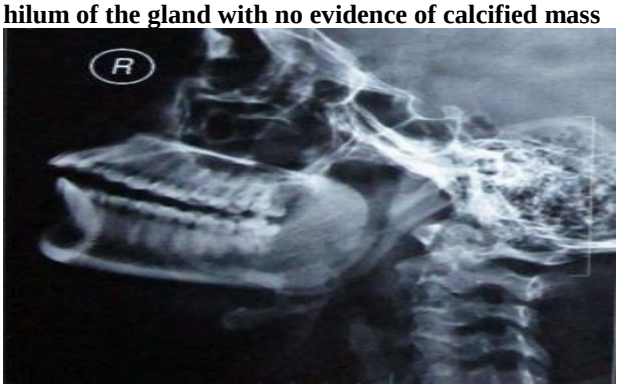
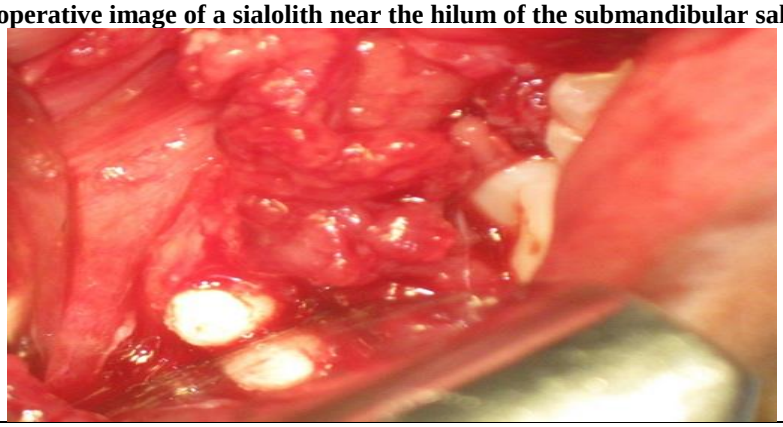


Fig 5. Intraoperative image of a sialolith near the hilum of the submandibular salivary gland



DISCUSSION

Salivary stones or sialoliths are calcified structures or concretions located in the parenchyma or ductal system of the salivary glands. Swelling associated with pain is the most common symptom in submandibular stones. Submandibular stones are usually located in the duct (80–90%), of which 57% is located in the hilum and 34% is located in the distal duct. Characteristics for sialolithiasis are episodes of pain and swelling during mealtime which may persist for a few hours, followed by long episodes of remission (weeks or months) [5, 6]. Submandibular sialoliths measuring less than 1 cm in greatest dimension are quite common but larger sialoliths are rare [4, 6, 17].

There are several factors that may contribute to the increased incidence of sialoliths in the submandibular gland. These include the more viscous mucus content of the saliva and the high concentration of calcium phosphate. This creates a more alkaline pH which not only favours the solid-liquid phase exchange of calcium phosphate species in the mouth and maintenance of the dentition but also causes precipitation of the more reactive species dibasic calcium phosphate dihydrate / brushite. This process is helped by the ascending course and narrow orifice of Wharton's duct compared to the calibre of the duct itself, both of which encourage stagnation of saliva. The initial radiographic examination of sialoliths is usually undertaken with plain films. Lustmann found that sialoliths were detected in 94.7 per cent of cases using intra-oral radiographs alone.[7] Large and well mineralized calculi are visible on plain radiographs but small or partially mineralized calculi may remain undetected [17]. Blatt (1964) in his study found that around 20 per cent of sialoliths remain unseen on plain film examination due to a low mineral content [18].

The exact etiology and histopathogenesis of salivary gland calculi is still an enigma. It is commonly thought that calcium salt deposition begins around an initial organic nidus consisting of altered salivary mucins, bacteria and desquamated epithelial cells [19, 20]. It is also further postulated that calculus formation occurs in two phases; one as a central core of precipitated salts bound by organic solution and another as a layered deposition of organic and inorganic material.[16] Predisposing factors are salivary stagnation, increased alkalinity of saliva, infection from oral cavity and physical trauma to the submandibular duct or gland. The size of the calculi in submandibular gland are variable and found commonly between 3–8 mm and occurs in the duct, hilum or glands with or without glandular atrophy or degeneration.

Sialolithiasis of the submandibular gland can be completely asymptomatic.[1] Common symptoms vary from a painless swelling, moderate discomfort to severe pain with large glandular swelling accompanied by trismus and usually associated with eating. Sialoliths are commonly 1-10 mm in size, but giant sialoliths (greater than 3.5 cm) have been reported occasionally [21].

Sialoliths developing in the hilus of the submandibular gland tend to be oval and may grow larger before becoming symptomatic reflecting the dynamics of fluid flow around the developing stone and the ductal structure [22].

The pain and swelling are caused by the obstruction of the salivary flow in the affected gland, resulting in accumulation of saliva and a subsequent increase in intra-glandular pressure. Several studies showed higher calcium concentrations in saliva in patients with salivary stones than in a healthy control group [23-26]. Saliva of patients with salivary stones contains reduced concentrations of the crystallisation inhibitors like phytate, magnesium and citrate, which may predispose to the formation of salivary stones [27]. The affected gland may feel firm and tender. In case of a submandibular gland, the affected side of the floor of the mouth may be elevated and inflamed [28].

As the submandibular salivary gland is positioned postero-inferiorly, a mandibular oblique lateral radiograph may be useful for visualization.[22]Sialoliths are well visualized on panoramic and periapical radiographs but can be obscured with superimposition over the roots of the premolar and molar teeth and muscle attachment ridges on the cortices of the mandible. Intraductal, large stones will usually show an anteriorly inclined stone due to the ascending course of the duct from the flexure in the lingual fossa to the anterior floor of the mouth [29].

Apart from conventional radiographs other imaging techniques that may be used to diagnose sialoliths include sialography, ultrasound, computed tomography and magnetic resonance sialography. Sialography is rarely indicated and should be restricted to those cases with a suspected ductal stricture or other obstruction but without a calcification visible on routine imaging. Until proven otherwise it is prudent to consider and exclude the presence of multiple sialoliths in any patient presenting with a sialolith. Computed tomography when there are multiple stones or when the stone is situated in a site which is not accessible for intra oral clinical examination.

In smaller sized stones, conservative management such as moist heat, increased intake of fluids, sialagogues and gentle massage of the gland towards the gland duct opening can allow spontaneous release of the stone [1,2]. A small sialolith near the orifice of the duct can be removed by widening of the orifice with a lacrimal probe [7]. Surgical removal is necessary in case of large sialolith. Sialoliths in the gland duct can often be removed without damage to the gland but intraglandular sialoliths generally require removal of the gland [30]. Stone removal in the posterior part of the duct or removal of the gland may lead to complications such as damage to the lingual and hypoglossal nerves or bleeding into the floor of the mouth. Haemorrhage in the floor of the mouth can lead to major complications and can even be life threatening. Therefore, postoperative observation is vital. This procedure is usually performed under general anaesthesia to control bleeding and to protect the lingual and hypoglossal nerves. Other treatments used successfully in the management of sialoliths include interventional sialendoscopy with wire-basket extraction for small sialoliths (< 4 mm) and fiberoptic laser lithotripsy with basket retrieval for larger sialoliths (> 4 mm) [13]. In any retrieval procedure within the ductal system, care must be taken to ensure that the stone does not track proximally and be lost to the extraction process. It should be made a priority to ensure both the duct and adjacent anatomical structures are not damaged to the extent of causing significant scarring or other anatomical deficit on healing.

CONCLUSION

Sialolithiasis is a common obstructive disorder of the salivary gland and should be suspected if the submandibular salivary gland is involved in the pathology. Ideal imaging techniques are necessary to identify their exact location. Immediate removal with minor or no trauma to the gland, duct or surrounding vital structures using the optimal technique is imminent to restore gland function and to promote good oral health of the patient.

STATEMENT OF HUMAN AND ANIMAL RIGHTS

All procedures performed in human participants were in accordance with the ethical standards of the institutional research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. This article does not contain any studies with animals performed by any of the authors

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Nil

CONFLICT OF INTEREST

No interest

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