Case Report - Surgical Pathology

Unusually large sialolith of Wharton's duct

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ABSTRACT

The formation of calcific concretions in the salivary duct or glands is a common disorder, especially in the submandibular glands. Most of the salivary calculi are small in size, in contrast to those that reach several centimeters, which are reported as megaliths or giant calculi in the literature. They may occur in any of the salivary gland ducts but are most common in Wharton's duct and the submandibular gland. This report presents clinical and radiographical sign of an unusually large sialolith. There was painless swelling on the floor of the edentulous mouth and patient was unaware of it. Radiographical examination revealed large irregular radio-opaque mass superimposed on right canine and premolar areas. This case report describes a patient presenting with an unusually large submandibular gland duct sialolith, the subsequent patient management, the aetiology, diagnosis and its treatment.

Keywords: Sialolith, submandibular salivary gland, wharton's duct

INTRODUCTION

Sialolithiasis is the most common disease of salivary glands. It is estimated that it affects 12 in 1000 of the adult population.[1] Males are affected twice as much as female patients.[2] Children are rarely affected but a review of the literature reveals 100 cases of submandibular calculi in children aged 3 weeks to 15 years old. Most salivary calculi (80%-95%) occur in the submandibular gland, whereas 5% to 20% are found in the parotid gland.[3] The sublingual gland and minor salivary glands are rarely (1%-2%) affected.[3] Intraductal stones are more common when compared to intraglandular stones. The submandibular gland is most frequently involved because of its anatomic location, long tortuous duct with a narrow orifice compared to the main portion of duct. Along with these factors, alkaline saliva rich in mucin also contributes to the stone formation. 40% of parotid and 20% of submandibular stones are not radiopaque and sialography/ diagnostic sialoendoscopy may be required to locate them[2] [Figure 1]. Salivary calculi are usually unilateral and are not a cause of dry mouth.[2] They consist of mainly calcium phosphate with smaller amounts of carbonates in the form of hydroxyapatite, with smaller amounts of magnesium, potassium and traces of ammonium.[4] Submandibular stones are 82% inorganic and 18% organic material whereas parotid stones are composed of 49% inorganic and 51% organic material.[3] The organic material is composed of various carbohydrates and amino acids.[4] Bacterial elements have not been identified at the core of a sialolith.[4]

CASE REPORT

A 55-year-old, edentulous male presented to the department of Prosthodontics of Career post graduate Institute of Dental Sciences and Hospital Lucknow for fabrication of complete denture. During intra oral examination of the patient a firm mass in the canine premolar region of the left side of the floor of the mouth was noticed. The patient was unaware of the swelling and noticed it when it was pointed out by the prosthodontist. The patient was referred to department of Oral and Maxillofacial surgery for the management of this swelling. Oral examination was unremarkable.

Intraoral bimanual palpation revealed the presence of a hard swelling, approximately 3 cm in length, in the anterior oral floor of mouth [Figure 1]. The entity was not attached to the underlying structures. The oral mucosa was normal in texture and without erythema. A dental occlusal radiograph [Figure 2] and orthopantomogram [Figure 3] confirmed that the swelling was radio-opaque structure in the floor of the mouth. In occlusal radiograph the calcified structure was mimicking a canine tooth but the radio opacity was homogenous. In panoramic radiograph lower border of sialolith was showing two extension passing vertically downwards probably into ducts of Rivinus. On the basis of clinical and radiological findings, a diagnosis of left submandibular duct sialolith was made. As it was a large sialolith (megalith), we elected to remove the sialolith surgically under local anaesthesia [Figure 4]. Upward and medial pressure was applied at the submandibular area, and an intra-oral incision was placed directly over the sialolith to expose it. After sufficiently mobilizing the sialolith, it was delivered through the ductal opening taking care that the downward extensions from the sialolith do not break. After successful removal of the sialolith a 2.0-cm-wide opening remained which was marsupialized intraorally with No. 4 vicryl sutures placed along its margins. Postoperatively, the intraoral opening was regularly irrigated, and after 45 days, the ductal opening reduced to 3 mm. Scientific importance of the patient's disease has been explained to him and he consented for publication of his case. The sialolith recovered measured 35 mm in length and 30 mm circumference [Figure 5].

No postoperative complications were noted. Histopathological validation was performed and during decalcification process the lesion showed concentric lamellas. On the basis of clinical, radiographical and histopathological findings, the diagnosis was confirmed as submandibular megasialolith.

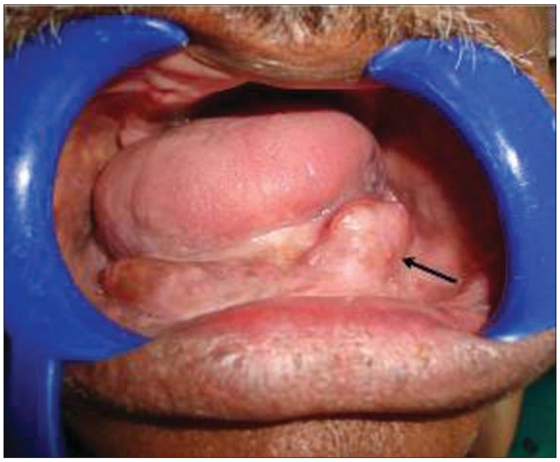

Figure 1: Intra oral view showing swelling in floor of mouth

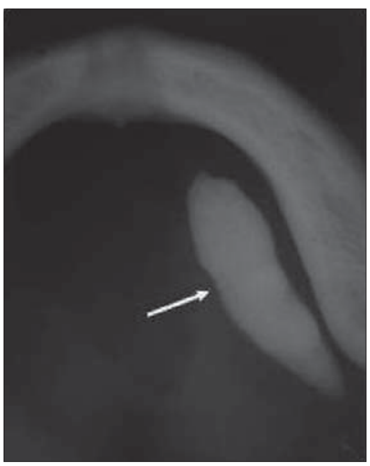

Figure 2: Occlusal view of mandible showing sialolith

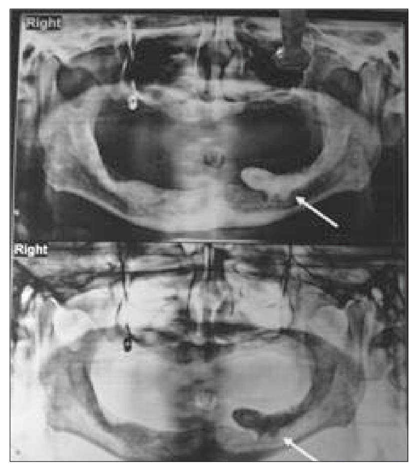

Figure 3: OPG showing sialolith

Figure 4: Intra operative photograph while removing sialolith

Figure 5: Removed sialolith

DISCUSSION

Salivary calculi are usually small and measure from 1 mm to less than 1 cm. They rarely measure more than 1.5 cm.[5] Mean size is reported as 6 to 9 mm. Giant sialoliths are rare and defined as the size of 3.5 cm or larger.[5] Large and giant calculi may perforate the floor of the mouth by ulcerating the duct or may result in a skin fistula by causing a suppurative infection. It is reported that the most common symptoms of sialoliths are recurrent pain and swelling of the associated gland during meals, as slow deposition and calcification of the stone usually does not block the flow of saliva completely. However, large sialoliths have been frequently reported in the body of salivary glands, they have rarely been described in the salivary ducts, particularly without any complaints from the patients.[1] In this report, clinical and radiological features of a large sialolith which was 35 mm in the size was presented. It was located into Wharton's ducts and the patient had no complains the swelling was noticed accidentally during intraoral examination when patient consulted for fabrication of complete denture. The largest sialolith reported in the literature was 70 mm in length in Wharton's duct and was described as having a "hen's egg" size.[5] Some unusual large salivary calculi may be seen without a long history; because the lesions were generally asymptomatic. It is believed that a calculus may enlarge at the rate of approximately 1 to 1.5 mm per year.

The exact aetiology 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 as a result of deposition of calcium salts around an initial organic nidus consisting of altered salivary mucins, bacteria and desquamated epithelial cells.[2] For stone formation it is likely that intermittent stasis produces a change in the mucoid element of saliva, which forms a gel. This gel produces the framework for deposition of salts and organic substances creating stone.[4] Traditional theories suggest that the formation occurs in two phases: a central core and a layered periphery.[6] 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 non organic material. Submandibular stones are thought to form around a nidus of mucous,[7] whereas parotid stones are thought to form most often around a nidus of inflammatory cells or a foreign body.[7] A retrograde theory for sialolithiasis has also been proposed.[6] Aliments, substances or bacteria within the oral cavity might migrate into the salivary ducts and become the nidus for further calcification. A case of stone formation around a vegetal nidus had been reported and this was histologically proven.[6]

Sialolithiasis typically causes pain and swelling of the involved salivary gland by obstructing the food related surge of salivary secretion. Calculi may cause stasis of saliva, leading to bacterial ascent into the parenchyma of the gland,[4] and therefore infection, pain and swelling of the gland. Some may be asymptomatic until the stone passes forward and can be palpated in the duct or seen at the duct orifice. It may be possible that obstruction caused by large calculi is sometimes asymptomatic as obstruction is not complete and some saliva manages to seep through or around the calculus.[1] Long term obstruction in the absence of infection can lead to atrophy of the gland with resultant lack of secretory function and ultimately fibrosis.

The treatment objective for giant sialoliths, as for the standard-sized stones, is restoration of normal salivary secretion. There are three ways in which we can treat patients with salivary stones: removal through the oral cavity, interventional sialoendoscopy, and resection of the gland. Our choice depends on the site, size, shape, number, and quality of the stones. The giant sialolith should be removed in a minimally invasive manner, via a transoral sialolithotomy, to avoid the morbidity associated with sialadenectomy.[3] Whenever the stone can be palpated intraorally, it is best to remove it through an intraoral approach.[1] The cardinal rule when performing stone removal from Wharton's duct is to first isolate the duct and then provide a longitudinal incision into the duct over the stone to retrieve it. In direct cut down approach to the stone, the initial incision is taken directly to the surface of the stone without primary isolation of the duct. Direct cut down is associated with the risk of ductal stenosis, except when the sialoliths are at the orifice of the duct or when there is a large stone in the submandibular gland pushing the gland upward and anteriorly. More anterior stones, 1 to 2 cm from the punctum, can be removed by cutting directly into the stone in the longitudinal axis of the duct while carefully protecting the lingual nerve.

Giant sialoliths are accompanied by long-standing salivary gland sialadenitis resulting in a grossly fibrotic and poorly functioning gland. However, after elimination of the obstruction, the apparent resiliency of the submandibular gland results in no adverse symptoms. Submandibular gland removal is indicated only when there is a stone of substantial mass (12 mm or more) within the gland itself that is not surgically accessible intraorally and when there are small stones present in the vertical portion of Wharton's duct from the comma area to the hilum. Surgical removal of gland is also indicated in situation where opening of the duct surgically created recurrent infection of the gland due to ingress of oral fluids.

CONCLUSION

There are various methods available for the management of salivary stones, depending on the gland affected and stone location. Newer treatment modalities such as extracorporeal shock wave lithotripsy (ESWL) and more recently the use of endoscopic intracorporeal shockwave lithotripsy (EISWL), in which shockwaves are delivered directly to the surface of the stone lodged within the duct without damaging adjacent tissue (piezoelectric principle) are effective alternatives to conventional surgical excision.[8] Transoral CO_2 laser sialolithectomy can also be used with a low incidence of complications, and can be readily managed on an out-patient basis. The laser is set up in continuous mode at 4–6 W with a focusing spot. Locating the stone was accomplished by manual palpation or lacrimal probe insertion with or without the aid of radiological images.

Other techniques for sialolithiasis fragmentation have been described, such as electrohydraulic and pneumoblastic devices. Electrohydraulic devices, initially described as promising, have been proven to be of low efficiency at low voltages. At higher voltages, although we have found that destruction was possible, injuries of the duct wall have been described and the technique criticized. Pneumoblastic devices are based on the delivery of mechanical energy to the stone. While no clinical trials using this technique have been published for salivary gland calculi, in vitro studies tend to emphasize the risks of wall perforations of the duct.[9]

The endoscopic revolution has not spared salivary gland pathology. In the early 1990s, endoscopy of salivary gland emerged. Sialendoscope is now available in four generations: free optical fiber, flexible endoscopes, and two generations of semirigid endoscopic devices of various diameters. The last generation of sialendoscopes could be called "all-in-one" sialendoscopes because they have an integrated irrigation channel that may also be used for introducing small-sized operating instruments. The endoscopic system includes diagnostic and interventional sialoendoscopy, a papillary dilator, forceps, grasping wire basket (3, 4 or 6 wires), and an electrohydraulic lithotripter. Custom-made forceps measuring 0.8 mm have also been designed and may be used to retrieve small salivary calculi or for taking biopsies within the salivary ductal system.

However, for giant sialoliths, transoral sialolithotomy with sialodochoplasty or sialadenectomy remains the mainstay of management.

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