

Case report of giant sialolith (megalith) of the Wharton's duct

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Abstract

Sialolithiasis is the most common disease of salivary glands. Its estimated frequency is 1.2% in the adult population. Sialoliths most commonly occur in the submandibular glands. The sublingual gland and minor salivary glands are rarely affected. The sialolith usually measures from 1 to <10 mm. Giant sialoliths are classified as those exceeding 15 mm in any one dimension. In literature, large sialoliths or megalith (>15 mm) of Wharton's duct have rarely been reported. This case report describes a patient presenting with an unusually large sialolith (megalith) of Wharton's duct, which was 37 mm × 16 mm in the size, the subsequent patient management, the etiology, diagnosis, and its treatment.

Key words: Calculi, megalith, sialolith, submandibular, Wharton's duct

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Introduction

Sialolithiasis is the most common disease of the salivary glands characterized by the development of salivary stones, known as calculi or sialoliths, in the salivary ducts or in the affected salivary gland.^[1-3] Its estimated frequency is 1.2% in the adult population. Males are affected twice as much as female patients.^[2,4] Sialolithiasis occur most commonly between the third and sixth decades of life, and only 3.0% of all sialolithiasis cases occur in the pediatric population.^[5] More than 80.0% of salivary sialoliths occur in the submandibular gland or in its duct, whereas 5.0–20.0% are found in the parotid gland. The sublingual gland and minor salivary glands are rarely (1.0–2.0%) affected.^[1,2] The submandibular gland is most frequently involved because of its longer course and

the angulation of Wharton's duct. Sialolithiasis is clinically characterized by local pain and swelling, reduced salivary flow, restricted mouth opening, and purulent discharge. The swelling is usually correlated to meals when salivary secretion is enhanced.^[6-8] Small sialoliths can be spontaneously expelled through the stimulation of the salivary flow by performing local massage or the administration of sialogogues. However, multiple or massive sialoliths often require major surgical procedures, such as lithotripsy, sialadenectomy, and sialotomy.^[6,9] The size of sialoliths may vary from less than 1 mm to a few cm in largest diameter, with most sialoliths being <10 mm in size.^[3,5,10] In literature, giant sialoliths are classified as those exceeding 35 mm in any one dimension.^[3] A giant sialoliths (megalith) (>35 mm) in the submandibular duct have rarely been reported.

In this case report, the clinical, radiological findings, and surgical treatment of an unusually large sialolith (megalith)

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of submandibular gland duct (37 mm × 16 mm in the size) is presented.

Case Report

A 35-year-old man presented to Ordu University, Faculty of Dentistry, Department of Oral and Maxillofacial Surgery for painful and persistent purulent discharge from unilateral submandibular ducts despite medical therapy. The patient

had no other significant medical history. In addition, in patient, there are no medical conditions associated with the formation of sialolith. On examination, there was a tense and sensitive submandibular salivary gland and visible swelling in the anterior part of the left side of the mouth floor [Figure 1]. Computerized tomographic (CT) scan and radiographs were obtained. The radiograph showed an elongated radiopaque structure superimposed on the left

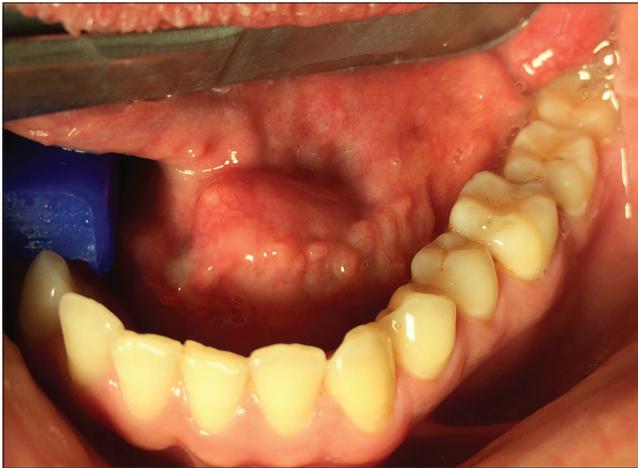


Figure 1: Preoperative intraoral photograph showing a swelling at the region of the left Wharton's duct

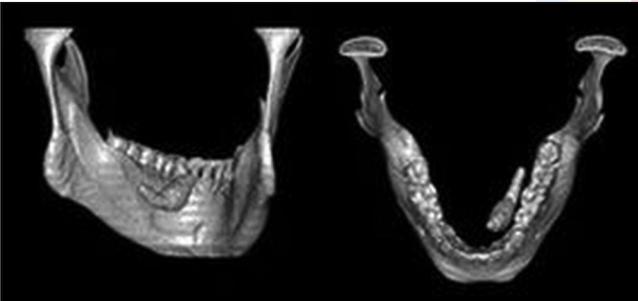


Figure 3: Three-dimensional computerized tomographic-scan shows large sialolith localized within the left Wharton's duct



Figure 5: Giant sialolith, measuring 37 mm × 16 mm



Figure 2: Panoramic radiograph shows large radiopaque mass at the left mandibular area



Figure 4: Intraoperative photograph while removing sialolith

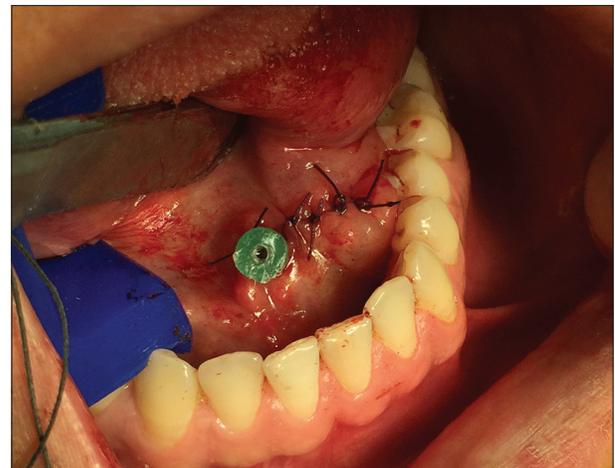


Figure 6: The duct cannulated with a catheter for purulent discharge

premolar areas [Figure 2]. A three-dimensional CT scan of the mandibular region showed the presence of a cylindrical and elongated sialolith located within the left Wharton's duct [Figure 3].

Following an informed consent, the sialolith was removed via intraoral approach under general anesthesia [Figure 4]. The incision was performed directly over the sialolith to expose it. In order to move the sialolith close to the incision, upward, and medial pressure was applied to the submandibular gland. The sialolith was carefully dissected from the floor of the mouth. Examination showed a hard, yellow, elongated stone-dimensions 37 mm long and 16 mm wide [Figure 5]. Following removal of stone, a drainage catheter was inserted into the duct for purulent discharge from submandibular gland [Figure 6]. An antibiotics (amoxicillin + clavulanate, 2000 mg/day), and an analgesic (naproxen sodium, 1100 mg/day) were prescribed postoperatively for 5 days. The drainage catheter was removed after 24 h. Reassessment of the patient took place on the 7th postoperative day. The left submandibular gland was found to be normal, and a clear salivary flow from the healed duct was observed.

Discussion

Sialolithiasis is the most common disease of the salivary glands and a major cause of salivary gland dysfunction. It is estimated that it affects 12 in 1000 of the adult population.^[1,2] The sialoliths can occur at any age, but it appears more frequently in patients in the third to sixth decades of life; sialolithiasis in children is considered rare.^[3,5] According to Bodner and Sheva,^[1] as the patient grows older and consumes more medication, in many cases, reduced secretory activity, alterations of electrolyte concentrations, impairment of glycoprotein synthesis, and structural deterioration of cell membranes of the salivary glands are seen. Thus, all of these may contribute to the underlying mechanisms for the higher incidence of calculi among the elderly.

The exact etiology and pathogenesis of salivary calculi are largely unknown. However, it is believed that salivary calculi develop as a result of deposition of calcium salts around a nidus of bacteria, mucus, or desquamated cells.^[2,4] Salivary stagnation, increased alkalinity and calcium content of the saliva, inflammation or infection of the salivary duct or gland, and physical trauma to the salivary duct or gland may predispose to calculus formation.^[4] The deposition of salivary calculi is not associated with systemic diseases involving calcium metabolism.^[5] Submandibular gland sialolithiasis is the most common (80.0–90.0%), followed by parotid gland sialolithiasis (5.0–20.0%).^[8] It is explained by the fact that the saliva of the submandibular gland is more alkaline, has an increased concentration of calcium

and phosphate, and has a higher mucous content than saliva of the parotid and sublingual glands. In addition, the submandibular excretory duct is wider in diameter and longer than the Stensen's duct, and the salivary flow in the submandibular gland is against gravity.^[3,5]

The submandibular sialolith typically causes increasing obstruction of salivary secretion, which leads to swelling, pain and infection of the gland, and finally requires surgical intervention.^[1,10] The swelling is usually worsened by meals. This is explained by an elevated intraglandular pressure resulting from an increased salivary secretion in the obstructed gland by the sight and smell of food.^[8] However, if the duct adjacent to the sialolith is able to dilate, allowing normal secretion of saliva around the stone, it might be asymptomatic and thus allowing the growth of a giant calculus eventually.^[1,10] In other words, the ability of a sialolith to grow and become a giant sialolith depends mainly on the ability of the duct to dilate to accommodate the stone. Sialoliths are generally small and measure from 1 to <10 mm. They rarely measure more than 15 mm. Giant sialoliths are rare and are defined as having the size of 35 mm or larger.^[3] Lustmann *et al.*^[8] reported that sialoliths in 78.8% of the cases in their study measured <10 mm account while those measuring 10-15 mm and >15 mm, respectively accounted for 13.6% and 7.65% of their cases. Giant sialoliths of the submandibular duct (>35 mm) have been very rarely reported in the literature.^[2,10]

The treatment objective of giant sialoliths, as for the standard-sized calculi, is the restoration of normal salivary secretion. Different treatment options may be selected according to the size and location of the sialolith.^[2] The sialolith should be removed with a minimally invasive procedure, usually through a transoral sialolithectomy, to avoid morbidity associated with sialadenectomy.^[1,4,5,10] As in our patient, whenever the sialolith can be palpated intraorally, the best option is to remove it through an intraoral approach.^[2,5,10] The ability to palpate the calculus, regardless of its location or size, is considered to be the most important factor in the successful transoral removal of the stone.^[10] However, submandibular gland excision is recommended in cases of intra-glandular sialoliths, which are inaccessible via a transoral approach.^[8] Conservative methods of treatment such as extraction with sialographic control using the sialoendoscope, laser intraductal lithotripsy, extracorporeal shock wave lithotripsy, and piezoelectric surgery techniques should be considered as an alternative to surgical excision, in particular for little sialoliths.^[9]

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Conflicts of interest

There are no conflicts of interest.

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