Case Report

# Unusual giant sialolith of Wharton's duct

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## Abstract

Salivary gland calculi account for the most common disease of the salivary glands. Most of the salivary calculi are small in size. Some calculi that reach several centimeters 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 in the submandibular gland. This report presents clinical and radiographical sign of an unusually large sialolith. A patient came with pain in the floor of mouth. There was a swelling on floor of mouth on the left side. Radiographical examination revealed large irregular radio-opaque mass superimposed on left lateral incisor to molar areas. This case report describes a patient presenting with an unusually large submandibular gland duct sialolith, the subsequent patient management, the etiology, diagnosis, and its treatment.

**Key words:** Sialolith, submandibular salivary gland, Wharton's duct

## INTRODUCTION

Sialolithiasis is the most common disease of salivary glands caused by the obstruction of a salivary gland or its excretory duct by a calculus. It is estimated that it affects 12 in 1000 of the adult population.[1] Males are affected twice as much as females.[2] It may occur at any age but there is a peak incidence in fourth, fifth, and sixth decades.[3]

Majority of salivary calculi (80–95%) occur in the submandibular gland, whereas only 5–20% are found in the parotid gland. The sublingual gland and minor salivary glands are rarely affected (1–2%).[4] Sialoliths located in the duct are usually elongated, while those situated in the gland or hilus tend to be round or oval.[3] The size of the salivary calculi may vary from <1 mm to a few cms in largest diameter. Most of the calculi (88%) are <10 mm in size, whereas only 7.6% are larger than 15 mm.[3] They consist of mainly calcium phosphate with smaller amounts of carbonates in the form of hydroxyapatite, with smaller amounts of magnesium, potassium, and ammonia.[5]

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## CASE REPORT

A 45-year-old male reported to the outpatient department of VSPM's DCRC, Nagpur, with the chief complaint of pain and swelling in the floor of mouth on the left side since 1 month. Detailed history revealed that it has started as a small swelling which used to increase before meals 1 month back. It gradually increased within a month.

Intraoral examination revealed a well-defined elongated swelling of approximately 2.5 cm × 1 cm in size in the floor of the mouth in relation to lower left incisors to first molar region. Overlying mucosa was inflamed and the swelling was hard in consistency and tender on palpation [Figure 1]. Radiographic evaluation included cross-sectional mandibular occlusal view which revealed a large well-defined

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elongated homogenous radio-opacity in the floor of the mouth on the left side in relation to lower left canine to first molar region. It was approximately 2.5 cm × 1.5 cm in size [Figure 2].

On the basis of clinical and radiological findings, a diagnosis of left submandibular duct sialolith was made. As it was a large sialolith, we elected to remove the sialolith surgically under local anesthesia [Figure 3]. The sialolith removed measured 25 mm (i.e. 2.5 cm) in length [Figure 4].

## DISCUSSION

The exact etiology and pathogenesis of salivary calculi is largely unknown. Genesis of calculi lies in the relative stagnation of calcium-rich saliva.[2] Salivary calculi are usually small and measure from 1 mm to <1 cm. They rarely measure more than 1.5 cm. Mean size is reported as 6–9 mm.[3] In this report the sialolith was in the Wharton's duct and the patient had pain before and during meals.

**Figure 1:** Swelling in the floor of mouth on the left side

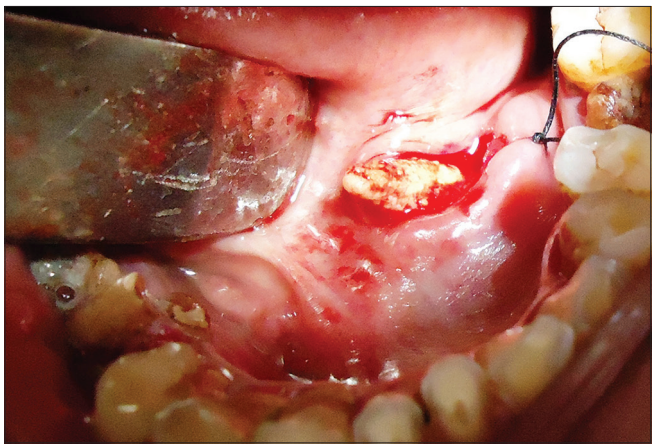

**Figure 3:** Removal of the sialolith from Wharton's duct

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 a stone.[5]

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 nonorganic material.[7] Another theory has proposed that an unknown metabolic phenomenon can increase the saliva bicarbonate content, which alters calcium phosphate solubility and leads to precipitation of calcium and phosphate ions.[5] 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.[6] Salivary stagnation,

**Figure 2:** Mandibular occlusal radiograph showing submandibular salivary duct calculus


**Figure 4:** The removed sialolith measured 2.5 cm × 1.5 cm in its largest dimension

increased alkalinity of saliva, infection or inflammation of the salivary duct or gland, and physical trauma to salivary duct or gland may predispose to calculus formation. Submandibular sialolithiasis is more common as its saliva is (i) more alkaline, (ii) has an increased concentration of calcium and phosphate, and (iii) has a higher mucous content than saliva of the parotid and sublingual glands. In addition, the submandibular duct is longer and the gland has an antigravity flow.[1]

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[5] and therefore infection, pain, and swelling of the gland at meal time.

Careful history and examination are important in the diagnosis of sialoliths. 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 and a uniformly firm and hard gland suggests a hypofunctional or nonfunctional gland.[8]

Imaging studies are very useful for diagnosing sialolith. The best view for visualizing radiopaque stones is a standard mandibular occlusal radiograph.[8]

Other traditional diagnostic methods include sialography, ultrasound, computed tomography, and scintigraphy for sialoliths.[9]

Sialoendoscopy can be used for both diagnostic and treatment purposes.[10]

In this report, as the lesions were observed clearly in occlusal radiographs, no further investigations were performed for diagnosis.

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.[4] Whenever the stone can be palpated intraorally, it is best to remove it through an intraoral approach.[1]

For giant sialoliths, transoralsialolithotomy with sialodochoplasty or sialadenectomy remains the mainstay of management.[8]

In some cases, excision of the entire gland is required. Submandibular gland removal is indicated if (1) the gland has been damaged by recurrent infection and fibrosis, (2) there is a stone of substantial mass within the gland itself that is not surgically accessible intraorally, (3) there are small stones present in the vertical portion of Wharton's duct from the comma area to the hilum, (4) the size of an intraglandular stone reaches 12 mm or more as the success of lithotripsy may be $<20\%$ in such cases.[8]

## CONCLUSION

There are various methods available for the management of salivary stones, depending on the gland affected and stone location. Transoral sialolithotomy remains mainstay of the treatment for giant sialolith in the duct of submandibular gland. Also, patients should be followed up regularly as recurrence has been reported in the literature.

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### Conflicts of interest
There are no conflicts of interest.

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