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# Submandibular sialolithiasis case report with 10 years follow up

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**Abstract**
Sialoliths are one of the most commonly encountered anomalies of the salivary glands in routine practice. Submandibular glands are more frequently affected due to certain anatomic and physiologic reasons. Usually may not exhibit clinical signs and symptoms and can be discovered as an incidental finding on dental radiographs. Otherwise they may present with symptoms such as submandibular pain of swelling. Swelling during meal times is pathognomic of this disease. We present case of sialolithiasis of the submandibular gland with ten years follow up managed via the intraoral surgical approach.

**Keywords:** Sialolithiasis; Salivary Calculi, Salivary Stone, Submandibular gland.

## Introduction
Sialolithiasis are calcified organic masses that form within the secretory system of the salivary glands [1]. 92% of the sialoliths occur in the submandibular gland, 6% in parotid gland and 2% in both sublingual and minor salivary gland [2]. It is more common in the males, with 2:1 male predilection reported the average age of the affected patients is 40.5 years. The true prevalence of silolithiasis is difficult to estimate as many cases are asymptomatic [1, 2].
Usually, sialolith measures from 1mm to less than 1cm. However, occasionally they can present in more than 1.5 cm size [3]. Mostly, sialolithiasis is clinically characterized by local pain and oedema during meals. Occasionally larger Sialoliths which almost completely obliterate the duct may result in reduced salivary flow, spontaneous bleeding and purulent discharge from the duct [4]. Small sialoliths are known to spontaneously expel through the ducts by stimulation of the salivary flow either by performing local massage or using mechanical or chemical sialogogues [5]. On the other hand, multiple or massive sialoliths often require major approaches such as surgery, lithotripsy and sialoendoscopy [6]. In this report we present a case of submandibular sialolithiasis which persisted for ten years before it was surgically retrieved.

## Case Report
A 39 year old male reported with a chief complaint of swelling in left submandibular region during meals in the year 2006. The swelling persisted for 30 minutes after food and then gradually subsided on its own. It was variable in size. On examination, there was mild swelling in left submandibular region and was tender on palpation. Ductal orifice was not inflamed. Expressed saliva was normal in viscosity and there was no evidence of pus discharge. There was no other foci of active odontogenic infection. Therefore clinical differential diagnosis of left submandibular sialolith and submandibular sialadenitis was made.
Mandibular true occlusal radiograph displayed an oval shaped radiopacity with smooth outline measuring 7-8 mm in the floor of the mouth. Internal structure was non homogenous with multiple layers of calcification (Figure 1). Hence based on the history, clinical examination and radiographic findings the diagnosis of submandibular sialolith was established. The patient was presented with all the treatment options but he opted out of surgery. Hence the patient was provided with symptomatic treatment such as antibiotics (Amoxicillin), analgesics (combination of Diclofenac, Paracetamol and Serratioppeptidase) and Sialogogues. Following this regimen the patient's symptoms subsided within seven days. The patient was followed up Initially every 6 months and then yearly basis and he remained asymptomatic. In the year 2016 he presented with persistent and painful swelling in the left submandibular region.

On examination, the skin over the swelling appeared to be inflamed and had smooth surface texture (Figure 2). On palpation there was rise in the local temperature, tenderness and firm in consistency. Submandibular lymphadenitis was present.

On intra oral examination, the floor of the mouth was raised, orifice of the Wharton's duct was inflamed and pus discharge was evident from it (Figure 3). Mandibular true occlusal radiograph displayed no change in the size of the sialolith. On ultrasonography of neck it was seen that the left submandibular gland was enlarged in size and hypoechoic (Figure 4). A calculus measuring 8.7 mm was seen in the lumen of the Wharton's duct with evidence of duct dilatation (Figure 5). The sialolith was removed intra orally under local anesthesia (Figure 6). The healing was uneventful.

**Fig 1:** Mandibular True Occlusal Radiograph Showing Calcified Sialolith in the Floor of the Mouth Region

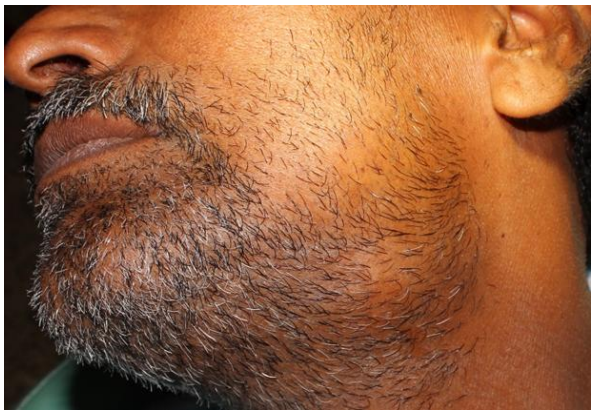

**Fig 2:** Extraoral Photograph showing swelling in the submandibular region

**Fig 3:** Intraoral photograph showing inflamed orifice of the left Wharton's Duct

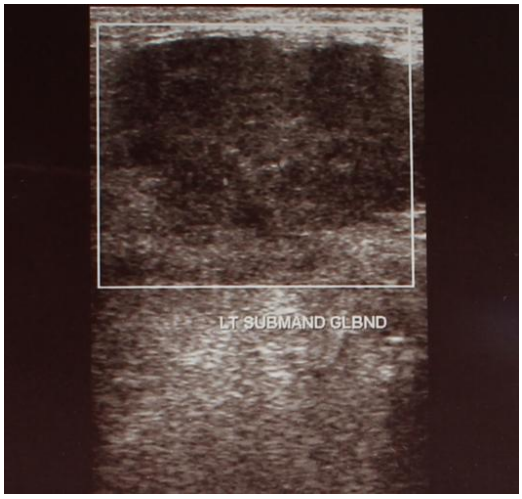


**Fig 5:** Ultrasound Image showing enlarged hypoechoic submandibular gland


**Fig 5.1:** Ultrasound image showing dilated left Wharton's Duct (left) and Sialolith within the ductal lumen (right)

**Fig 6:** Calcified Sialolith after surgical extraction.

## Discussion

Sialoliths are deposition and condensation of calcium salts primarily calcium phosphate in the form of hydroxyapatite with small amounts of magnesium carbonate and ammonium [7]. The definite etiology of our case remains unknown. So far various etiologies have been reported in the literature. Broadly the etiological factors can be classified into those favouring retention of saliva and changes in the salivary composition. Retention of saliva may be due to irregularities in the salivary ductal system, local inflammation, drug induced such as anticholinergics and diuretics and dehydration [1]. Sialolith formation can occur in two stages; central core formation and layered periphery formation. Initially, mineral salts bound by certain organic substances precipitate either in the duct lumen or in the acini to form the

central core. Thereafter, in the second phase some organic and inorganic materials deposit around the central core in layers. Parotid and submandibular stones are thought to frequently form around a nidus of inflammatory cells or foreign body and a nidus of mucous respectively [8].

Another theory mentions an unknown metabolic phenomenon which can lead to precipitation of salivary calcium and phosphate ions by increasing the salivary bicarbonate content, which in turn alters the calcium phosphate solubility. Schroder et al. studied the salivary composition of 40 patients with sialolith. They discovered that the salivary ion concentration particulary salivary calcium, phosphorus and magnesium was markedly high in patients with sialolithiasis as compared to healthy controls [9]. A retrograde theory suggested that any substance particle or bacteria of the oral cavity that had migrated into the salivary ducts in retrograde fashion, can act as a nidus for further calcification [7]. Marchal F et al. further suggested that retrograde theory is more of a possibility for submandibular sialoliths because migration of particles is easier due to the sphincter-like mechanism in the first 3 cm of the Wharton's duct [8].

Salivary calculi related to the submandibular gland are more common than the parotid gland due to some factors like the direction of salivary flow against gravity, a longer and more tortuous structure of Wharton's duct and the higher calcium and mucin content of saliva produced in the submandibular gland [10, 11]. Calculi are more often found within the Wharton's duct, than at the hilum of the duct or inside the gland [7]. Submandibular gland sialolithiasis is generally asymptomatic in nature. The symptoms include pain and swelling of the involved gland caused by the accumulation of saliva due to blockage of the lumen of Wharton's duct by a salivary calculus. Recurrent infections may occur due to the ascent of bacteria into the parenchyma of the gland [7]. Our patient remained fairly asymptomatic for ten years with occasional exacerbations which were successfully managed by providing symptomatic relief.

The diagnosis of Sialoliths is established by clinical features combined with radiographic interpretation, though newer more sophisticated techniques are now available. We lay more emphasis on diagnosis by the traditional method, based on the patient's history and clinical examination and supplemented by radiographic findings. Commonly majority of the patients will present with pain and swelling of the involved salivary gland at the times of meals or in response to any other salivary stimuli. In cases where the calculi almost completely obliterates the duct may lead to retention and development of microbial species. In such cases pus may be seen draining from the duct accompanied with signs of systemic infection. A posterior to anterior bimanual palpation of the floor of the mouth can reveal submandibular sialoliths in majority of the cases. In the present case, the stone was present inside the Wharton's duct, but was not palpable on the floor of the mouth.

Occlusal radiographs are extremely useful in showing radiopaque stones unless otherwise there are radiolucent stones. Occlusal radiographs are readily available, inexpensive and cause minimal radiation exposure to the patient [1]. Ultrasound is also increasingly employed for imaging of salivary gland calculi. The main advantages being it is non-invasive, ability to visualise non calcified calculi and less costly than other advanced imaging. According to Katz P et al. it has sensitivity and specificity between 90% and 95% respectively [12]. The drawback of this imaging modality is that there is no enough evidence supporting the sensitivity of ultrasound in detecting calculi and stones less than 2 mm in size may not produce an acoustic shadow and may remain undetected [1]. In the present case, ultrasound was performed to precisely locate the position of the sialolith within the duct and to also assess the submandibular gland. Other imaging modalities such as computed tomography and cone beam computed tomography are also routinely used to detect sialoliths. Their major advantage being high sensitivity towards detection of micro sialoliths [1]. However, as they are expensive and result in higher radiation exposure to the patient as such these imaging techniques were not used. After removal of sialolith it is recommended that patients be advised to have a diet rich in proteins, liquids and acidic pH foods to prevent recurrence or formation of new sialoliths in the glands [13].

## Conclusion

The history, clinical and radiographic examinations are crucial for diagnosis Sialolithiasis. Sialoliths should be considered as one of the differential diagnosis wherein the patients present with submandibular and facial pain particularly when it is related to mealtime. Although advanced imaging modalities are available occlusal radiographs are still useful in diagnosing sialoliths.

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