Submandibular Duct Salivary Stone Perforating the Floor of the Mouth: Review of Literature and Case Series

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Abstract

Majority of salivary gland stones (sialoliths) occur in the submandibular gland (Wharton's duct and parenchyma) accounting for 80% of cases, due to its anatomic features. The purpose of this article is to report a case series of submandibular duct salivary stone perforating the floor of the mouth and review the related literature regarding "submandibular duct stone perforating the floor of the mouth".

This is 04 case report series reported to the clinic presenting with a tender hard mass beneath the tongue which is dull and intermittent pain while eating. Three cases had their submandibular stones transorally removed under local anesthesia with help of adson's forcep along with milking of the submandibular salivary gland. One case reported self-exfoliation of salivary stone early in the morning before the planned surgical procedure.

Conservative treatment with transoral removal of submandicular duct stone should be considered. This will preserve the submandibular gland with minimum surgical trauma to the duct and also bestow full post -surgical function of the gland.

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Introduction

The submandibular gland (Wharton's duct and parenchyma), which accounts for 80% of all calculi, is the site of the majority of salivary gland sialoliths. It frequently involves the Wharton's duct n the submandibular region[1-2]. The submandibular glands account for the majority of salivary stones (80-95%), followed by the parotid gland (5-20%) and, less frequently, the sublingual and minor salivary glands (1-2%)[3-5].

Salivary stones are mostly seen in the distal portion of the Wharton's duct[6]. This is due to the long, curved shape of the Wharton's duct, the horizontal flow of saliva in the opposite direction of gravity, high alkali, calcium and mucin concentration found in saliva that is produced by the submandibular gland and the opening of the duct being narrow increases the incidence of the salivary stone in the submandibular gland[7-8]

Clinically Salivary stones are yellowish in color, ovoid or round and usually small in size measures from 1mm to less than 1 cm. Sialoliths have been estimated to grow through deposition at a rate of 1 to 1.5 mm per year[9-10].

The pathophysiology of the salivary stone formation is not clear. Sialoliths are believed to form as a result of an initial organic made up of bacteria, desquamated cells, and salivary mucin followed by deposition inorganic material around the initial nidus[11].

Salivary calculi are diagnosed using

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clinical and radiographic findings. The patient complains of pain and swelling, especially after meals[12]. Occlusal and panoramic x-rays are commonly used to diagnose salivary calculi. Ultrasonography, Computed tomography (CT), Sialoendoscopy can be used for diagnostic purposes[13]. Smaller stone identification and the differentiation between acute and chronic obstruction are both easier to detect by MRI[12].

The treatment of submandibular salivary stone depends upon the location and size of the stone. Small salivary stone can be milked out through the orifices of the Wharton's duct by bimanual palpation, application of moist warm heat and administration of sialogogues will aid in flushing out the salivary stone through the duct [12]. Antibiotics must be prescribed in case of infection.[14]

A transverse incision is required to remove the stones distal to the punctum. Extracorporeal shock wave litrotripsy can be considered for large salivary calculi located in the close proximal duct[15]. Co2 is gaining popularity because of minimal bleeding and less post-operative complications. When the sialolith (12 mm or more) is located within the gland and intraoral surgical access is inadequate, submandibular gland resection is done as a last resort[16].

This study aimed to report four cases Wharton's duct salivary stone perforating the floor of the mouth, which requires conservative treatment without an incision to remove the salivary stone and to review the related literature to submandibular duct stone

Case report 1:

A 37-year-old female patient was seen at the oral surgery clinic at the college of dentistry in Jazan. With a hard mass beneath the tongue as their main complaint, along with dull, intermittent pain during eating particularly sour food since last 1 week in right submandibular area. The patient reports no prior history of pain in the right submandibular gland region and had no significant systemic medical problems.

Intro-oral examination reveals a yellow colored hard structure in the floor of mouth, perforating the opening of the submandibular duct (Wharton's duct) was seen and the duct opening was slightly inflamed. (Fig 1A). The ipsilateral submandibular gland was not enlarged. Radiographic examination was done. As occlusal radiograph was not possible, an IOPA was used in place of occlusal radiograph which revealed a single ovoid shaped radiopacity in relation to #44 and #45. (Fig 1B). The results of the laboratory tests were normal; the serum sodium, potassium, and calcium readings were normal, and the hemoglobin level was 13 g/dl. Based on the clinical and radiological findings diagnosis was made as right submandibular sialolithiasis.

The patient provided informed consent. The stone measuring 9x4mm was removed under local anesthesia with help of adsson's forcep.(Fig 1C,D) The patient was prescribed Augmentine 625 mg TID and Paracetamol 500 mg to be taken as needed. The patient was instructed to take soft food diet for few days. A follow up was done after 1 week reveals normal saliva drainage from the duct.

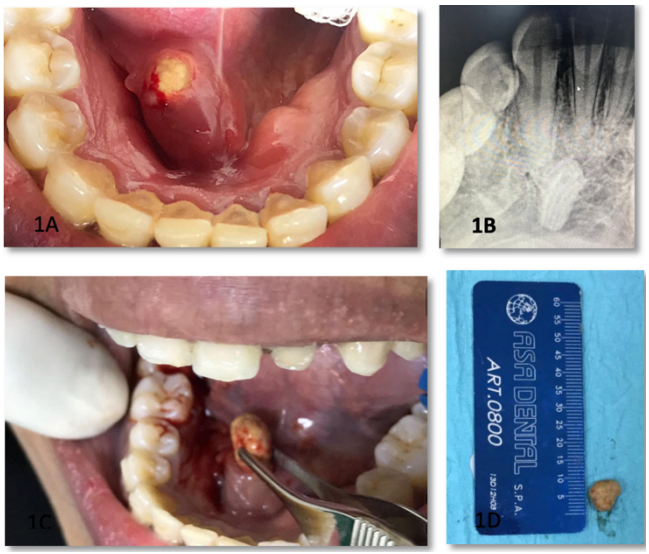

Figure 1A (Case 1) - Intraoral view: Sialolith perforating the floor of the mouth on intraoral examination , Fig 1B- Radiographic View: Intra Oral periapical view radiograph revealing a radiopaque stone, Fig 1C- Surgical removal of Submandibular duct stone and Fig 1D- Measurement of sailolith.

Case report 2:

A 24 years old healthy patient reports to a private clinic with the chief complaint of pain in right submandibular region below the tongue during the meal time and the pain gets relieved after 1 – 2 hours.

Local examination reveals a yellow colored hard structure in the floor of mouth, perforating the opening of the submandibular duct was seen.

(Fig 2A) During extra oral examination and palpation of the submandibular gland, the stone was seen to move upward, this hard structure measuring 12x3 mm was grasped with an Adson forceps and removed.(Fig 2B). As there was no need to surgically intervene in this case. A follow up was done after 1 week reveals normal saliva drainage from the duct.

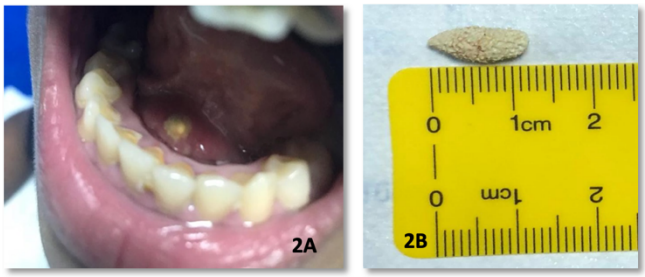

Figure 2A (Case 2) - Intraoral view showing part of the submandibular salivary stone seen out of the duct, Fig 2B- Measurement of sailolith.

Case report 3:

A 47 year old male patient, with primary complain of severe pain and a swelling in the left submandibular area since 1 week. The pain was aggrevated while eating food. Patient gives the history of ibuprofen 400 mg for pain relief. He gives past history of submandibular duct stone 15 years back and under local anesthesia in a private clinic stone was removed.

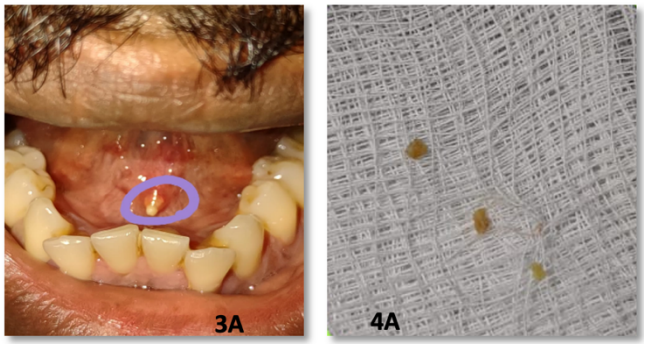

Figure 3A (**Case 3**) - Intraoral view showing part of the submandibular salivary stone perforating the floor of the mouth, Fig 3B- Multiple Sailoliths.

During palpation of the submandibular gland, an effort was made to manipulate the intraoral site by causing pressure on the submandibular region. First stone measuring 2.5mm which was projecting from the duct orifice was removed by Adson forcep, further milking of the submandibular salivary gland was done to release engorged saliva by giving patient a few drops of lemon juice, two small size stones measuring 2mm and 1.5 mm were expelled from the orifice of the wharton's duct. The patient was recalled after one week for follow up reveals normal saliva flow from the duct.

Case report 4

27 year old healthy male patients reported to the clinic with dull pain in lower left submandibular area, on examination revealed yellowish hard mass projecting from the submandibular duct orifice. As the patient was not willing for the treatment, the patient was planned next day for further treatment. Next day the reported self-exfoliation of salivary stone early in the morning.

Figure 4. (Case 4) - Intraoral view showing submandibular salivary stone perforating the floor of the mouth.

Discussion

The main goal in treating sialolithiasis should be preservation of gland function together with minimal danger and pain for the patient. The treatment chosen varies depending on the size, position and number of stones.

Saliva flows against gravity, high calcium and mucin content, and has a more alkaline pH in the submandibular gland, which may explain why calculi occur there more frequently than in the parotid gland[17]. The wider diameter of Wharton's duct and its longer, tortuous duct, the fact that salivary secretions from the submandibular gland flow against gravity and are more alkaline than those from the parotid gland, the fact that submandibular saliva appears to be more mucinous than that from the parotid gland, and the fact that submandibular saliva has a

higher concentration of calcium and phosphate are some of the possible causes of salivary gland calculi[18]. It is believed that salivary stasis or a reduction in salivary flow may contributes in calcium precipitation[19].

The exact etiology of salivary calculi is not completely understood. Numerous theories have been put out in an attempt to understand the exact etiology of salivary stones. Typically, it is believed that they develop when tricalcic phosphate salts are deposited around a nidus composed of bacteria, desquamated epithelial cells, and altered salivary mucin[10]. It is hypothesized that bacterial toxins create a local environment with a pH lower than 5.5, causing tissue injury. When a pH of 7.2 is restored during the healing process, salivary ions, particularly calcium phosphates, crystallize, causing calculus to develop[10]. Another theory suggests that an unknown metabolic process raises the salivary bicarbonate level, changing the solubility of calcium phosphate and causing precipitation of calcium and phosphate ions[3]. According to the retrograde theory, materials or bacteria from the oral cavity move into the salivary ducts and serve as the nidus for further calcification[3].

Sialolithiasis can be seen in any age group. The fourth and fifth decades of life are when salivary stones are most prevalent. Patients with submandibular stones tend to be slightly younger than those with parotid stones in terms of average age[20, 21].

The age in the cases reviewed from the published literature ranged from 22 to 70 years, majority of the patients are over the age of 40 years. The incidence is higher in men (n = 20) compared to women (n = 9), with a male to female ratio of 3.2:1, among the 30 case reports summarized in Table 2. Between the left and right sides of the mouth cavity, salivary stones are evenly distributed. Left side was involved in 17 cases, whereas 11 cases were seen on right side. So it would seem that the sialolith has a preference towards the left side. But our case series shows equal distribution.

Single salivary stone is found in the affected salivary gland in 70–80% of patients, two salivary stones are found in 20% of patients, and three or more salivary stones are found in about 5% of patients[22]. 80–90% of submandibular stones are found in the duct, with 34% of them in the distal duct and 57% in the hilum. The gland itself contains 10% of the submandibular stones[23]. Stones which perforate the floor of mouth are rare. They are also called as perforating stone (sialolith) and also being referred as self-exfoliating stone in the literature. Perforation of the stone through the floor of mouth by the submandibular duct stone is rare.

Sialolithiasis is characterized by episodes of pain and swelling during meals that may last for a few hours and are followed by lengthy remissions (weeks or months)[3]. In the cases that were reviewed, pain and swelling were the most prevalent symptoms. A salivary stone produces pain and swelling in the affected salivary gland by obstructing the salivary secretion during consumption of eating food. The degree of obstruction determines how severe the symptoms are. In few cases patients were asymptomatic. Saliva may leak through or around the sialolith in cases of partial occlusion of the duct[24]. In these situations, a salivary stone may not cause any symptoms and may only be discovered by chance on a dental panoramic radiograph.

It is not clear what causes salivary gland stones, so there's no way to prevent them, they're not generally linked with any other health problems. The only known systemic condition that increases the chance of sialolith development is gout. Gout patients' salivary stones are mainly composed of uric acid[25]. It has been hypothesized that those who live in places with hard water are more likely to develop salivary calculi. The prevalence of salivary stones and the calcium concentration of the water in various parts, however, are unrelated[26].

The majority of patients with suspected submandibular duct stone will visit the clinic when the condition is acute, so bimanual palpation of the floor of the mouth should be part of the physical examination for submandibular gland[27]. Panoramic and occlusal views are the most often used radiographic modalities to identify submandibular salivary stones [13, 28]. The fact that bigger salivary stones are mostly radiopaque and can be detected on panoramic radiographs is likely due to the length of their lithogenesis, enabling full calcification[15].

For detection of small salivary stones, investigations like sialography, ultrasonography, and computed tomography may be required. Stones having a diameter of 2 mm or larger can be detected using ultrasonography[20]. This method also has the added benefit of being

particularly effective during acute attack of sialendenitis[27]. Any size of sialolith can be detected with computer tomography (CT) and cone beam computer tomography (CBCT), but they have the drawback of having a high radiation dosage[29]. A more recent diagnostic technique, magnetic resonance sialography, allows for examination of the ducts without the need of radiation or dye injection, although it has drawbacks related to cost and applicability in patients who are claustrophobic[30].

The goal of treatment for submandibular duct stone is to reestablish normal salivary production. The salivary stone's size and location will determine the way it is managed. It is possible to milk and manipulate a small stone that is piercing the floor of the mouth through the duct opening[31]. The use of sialagogues, irrigation, and gland massage serve as non-invasive conservative treatments for sialolithiasis. When the stones are small and in the duct, this procedure has the best success rate. Antibiotics ought to be provided when an infection appears to be present[3].

A reasonably straightforward intraoral technique performed under local anesthesia can remove almost all intraductal submandibular stones. This includes stones in the submandibular region close to the duct's orifice. The distal portion of the duct, where nearly half of the submandibular calculi are located, can be easily released surgically through an incision in the floor of the mouth[3]. Following stone removal, it is advised to massage the salivary glands many times per day, along with a sour diet and sialagogues to increase saliva production[32]. Sutures should not be used in the incised duct since this may increase the chance of scarring[29]. Surgery, sialoendoscopy, or extracorporeal shock-wave lithotripsy are among invasive treatments for sialolithiasis. The secretion rate of the treated gland is comparable to that of the contralateral gland in 75% of instances following transoral surgical excision of submandibular stones[33]. Salivary gland function can be reestablished in response to factors such glandular infection, salivary stone diameter, and patient age[34].

Recurrence of salivary stone is rather uncommon, and is estimated to occur in 1–10% of the patients[35]. In a survey conducted by Lustmann on 245 individuals with sialolithiasis over a 20-year period (1968–188), he observed a recurrence rate of 8.9% during a follow-up of 10 years[36]. To avoid recurrence, it is advised to consume a diet high in proteins and liquids, including acidic foods and drinks[10]. Nemade SV [37] presented a case of recurrence of perforating submandibular duct stone perforating the floor of the mouth on the left side similar to our 3rd case where the patient had similar stone formation 15 years back.

Spontaneous expulsion of submandibular duct stone has been documented in the literature. The opening of the Wharton's duct is smaller than the lumen, providing a sphincter that will prevent the passage of the stone, making spontaneous passage of a sub-mandibular duct stone rare. Sutay et.al, [38] reported a case where a submandibular duct stone perforating the floor of the mouth was planned for surgical removal, after 3 days the stone was self-exfoliated on itself without any surgical intervention which was similar to one of our case (4th case report). This is can be explained by the fact that the usual diameter of submandibular duct is in the range between 0.2 mm and 2.2 mm and submandibular gland duct has the ability to dilate up to large extent to accommodate large stone.

Through our case series we would like to highlight that it is possible to remove the stone through the forceps without the need of further intervention, as the stone in the wharton's duct make it sufficiently dilated hence can be conveniently removed, which are in similar with case reports of Park SY et.al.[39] in one case of their case series had inserted a polyethylene tube after removal of the salivary stone from the orifice of the wharton's duct with a belief that there would be a chances of stenosis of the duct, but frequent drop out of the polyethelene tube had occurred. In remaining two cases polyethylene tube insertion and suturing was not done, they observed good healing and normal saliva flow.

In our case series: the submandibular duct stone was located anteriorly in the floor of the mouth and was removed by the addson's forceps after increasing the salivation with the help of lemon drops, along with milking of the submandibular duct without requiring of additional surgical procedures. This was because the submandibular duct was already dilated due to the presence of the salivary stone in the duct. Post removal there was normal salivation, hence no further intervention like suturing the orifice of

the duct to the mucosa or inserting a catheter or any other intervention was not required.

Hence, smaller stones can be treated conservatively, Conventional or conservative treatment comprises frequent consumption of water and sour juice, as well as routine milking of the submandibular gland[63].

	Case I	Case II	Case III	Case IV
Gender	Female	Male	Male	Male
Age	37	24	47	27
Side	Right	Right	Left	Left
Size	9 X 4 mm	12 X 3 mm	-2.5 mm - 2 mm - 1.5 mm	___

Table 1. Details of Perforating Stone in our case series.

Conclusions

Sialoliths should always be taken into account in submandibular and face pain, especially if it coincides with the meal. To confirm the clinical diagnosis and identify the exact location of the calcification, appropriate imaging procedures must be used in conjunction with a thorough history. Despite the availability of more sophisticated and effective techniques, occlusal radiographs are still helpful in the diagnosis of sialoliths. Smaller stones can be treated conservatively, but if better technologies are not yet available, sialolithotomy combined with antibiotics is the preferred treatment for bigger stones. In recent years good results have been seen without the insertion of tube to maintain the patency of the duct and without suturing the duct to the surrounding mucosa in the floor of the mouth.

Declaration of Interest

The authors report no conflict of interest.

Author	Side	Sialolith size	Age/ Sex	Removal method
Siddiqui S.J[3]	R	30mm	52/F	Stone was removed under local anesthetic with sharp dissection.
Kurtoğlu G[7]	R	2.5×0.8×0.6	52/ F	Removed without using any anesthetic agents with the aid of forceps.
Oteri et al.[10]	L	1.5 mm	51/F	Under local anesthesia, an incision along the mucosa of the floor
Sutay S [37]	R	37 X 7 mm	22/F	Extruded spontaneously on itself
Yaman F [40]	L	2x3x3 cm 2x2x1 cm	22/M	Stone was removed by the curette and forceps.
Leite TC [41]	R	35×7 mm	54/F	The sialolith was surgically excised under local anesthesia
Abdullah [42]	L	3.6 cm	37/M	Left submandibular intraoral stone removal with marsuplization of the duct
Pachisia S[43]	R	1×3cm	75/M	Sialolithotomy was done under LA and sutures were placed
Shahoon H[44]	L	83X12 mm	30/M	Light pressure at distal ligature, sialolith was expelled through the incision.
Akinyamju[45]	L	4.4 x1.8cm	54/M	The sialolith was removed non-surgically
Saluja H[46]	R	2 × 1 cm	65/M	sialolith was remove by blunt dissection under local anesthesia
Vasanthika T[47]	R	4.5 cm x 3.5 cm	56/ M	Under general anaesthesia. Calculus was removed from the duct without making any incisions over the floor of the mouth
Alhamdani[48]	L	18 mm	35/M	Manipulation & Milking of the submandibular gland was performed
Singh AM[49]	R	42 mm	40/ M	The stone extraction with marsupialization of duct was done under local anaesthesia
Erdogan[50]	R	25x7 mm	28/M	- Under local anesthesia, an incision was made over the canal overlying the sialolith, sialolith was removed. - Intravenous catheter stent placement was performed into the duct to ensure normal salivary flow
Rao K[51]	L	2.2 X 0.5 X 0.4 cm.	37/M	Milking of the submandibular duct was performed bimanually and the sialolith was expelled.
Gill MP[52]	L	10 × 5 mm.	37/M	Spontaneous expulsion of Sialolith on itself
Rodrigues GHC[53]	L	45 mm	48/F	During examination of the submandibular gland, a calculus removed without anesthesia or surgical excision
Singhal A [54] Case 1& 2	L L	15mm	55/M 40/M	Self-exfoliation Self-exfoliation
Gehani RE[55] Case 1& 2	L L	_____	41/M 32/ F	The stone was removed via a transoral approach with sharp dissection under local anesthesia
Bhovi TV [56]	R	35 mm	50/F	Under local anesthesia, a sialolith was via intraoral sialolithotomy
Ansari K[57]	L	1.5x 1.0cm	55/M	Under local anesthesia a incision was given over the duct Blunt dissection was done and sialolith was removed
Thiyagarajan V[58]	L	1 x 3.5 cm	48/F	Transoral sialolithotomy was done to remove the submandibular duct stone under local anaesthesia.
Adhikari AD[59]	L	21.3, 9.02, 3.8 mm	56/M	Transoral approach under local anaesthesia
Sathe NU[60]	R	8x2.5cm	70/M	Removed using Tilley's forceps and gentle manipulation under LA
Oliveira [61]	L	13 mm & 16 mm	50/M	A surgical removal via intraoral incision under local anesthesia, through sialolith anchorage by suture thread
Kiran DN[62]	R	40 X 20 mm	65/ M	Intraoral incision was made directly over the sialolith and blunt dissection was performed.

Table 2. Comparative table of various cases of operated submandibular duct calculi in the published literature.

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