Management of giant salivary sialolith: A case report

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Abstract
Sialolithiasis is the formation of calculi in the salivary gland or duct which results in the obstruction of salivary flow. Sialolith is a calcarceous substance, which may form in the gland or the duct of the major or minor salivary glands. Sialolithiasis is known to be one of the most common diseases of the salivary glands in middle aged individuals. The majority of sialoliths (80%) occur in the submandibular gland or its duct and are a common cause of acute and chronic infections of the gland. This case report describes a patient presenting with an unusually large sialolith in submandibular duct, treated surgically through intra oral approach with no subsequent post-operative complications. Also included is a brief literature review discussing the aetiopathogenesis, diagnosis, various diagnostic aids, and treatment modalities for management of salivary gland calculi depending on their site and size.

Keywords: Salivary gland; Giant Sialolith; Submandibular duct; Infection; sialolithiasis

Introduction
Sialolithiasis is the formation of calculi in the salivary gland or duct which results in the obstruction of salivary flow. Sialolith is said as a calcarceous substance, which may form in the gland or the duct of the major or minor salivary glands. Although any of the salivary gland or duct may be obstructed by the formation of sialolith, nearly about 80% of the sialoliths form in the submandibular gland. This is because a long, curved wharton’s duct has increased chance of entrapment of organic debris; the secretions of this gland is higher in calcium content and thick in consistency, and the position of the gland increases the chances for stagnation of the saliva. The factors like inflammation, local irritation or drugs can cause salivary stagnation leading to the buildup of an organic debris, which eventually will calcify. The size varies from less than 1 mm to a few cm in size, with most calculi being less than 10 mm in size(1).

Sialolithiasis affects approximately 12 in 1000 of the adult population with males being affected twice as much as females and children are rarely involved(2). Giant salivary gland stones measuring greater than 15 mm have rarely been reported in the literature(3). Sialoliths close to the hilum of the gland tend to become large
before they become symptomatic\(^3\). Intraductal sialoliths are more common, elongated in shape as compared to intraglandular stones, tend to be oval\(^4\).

There are currently two theories about the pathogenesis of salivary stones\(^5\). The first is that intracellular microcalculi develop in normal salivary tissue and are naturally voided through the duct system. If these became impacted during their discharge, they will act as a nidus and help in sialolith formation. An alternative explanation is that a mucous plug forms and calcifies after being exposed to a supersaturated solution\(^1\). Sialoliths may be ovoid or round, smooth or rough with a yellowish color. They are composed of calcium phosphate with small accounts of hydroxyapatite, magnesium, potassium and ammonia\(^3\).

The formation is supposed to occur in two phases: a central core phase and a layered periphery phase. The central core is formed by the precipitation of salts, which are bound by certain organic substances followed by second phase of the layered deposition of organic and non organic material\(^6\). Factors that tend to favour submandibular sialoliths are: longer caliber ducts and slower rates of salivary flow, salivary flow against gravity, increased alkaline nature of saliva and, high salivary mucin and calcium content\(^2\).

Diagnosis is usually done clinically by bimanual palpation in a posterior to anterior direction, which in turn reveals a palpable stone in a large number of cases, a uniformly firm gland suggestive of hypofunctional or non-functional gland\(^3\). Basic imaging like occlusal and panoramic radiographs, to detect deep submandibular or radiolucent or deep parotid stones sialography is useful but is contraindicated in acute conditions and in patients allergic to contrast material\(^4\). In such cases ultrasonographic examination is considered to be simple and non invasive modality of diagnosing the sialoliths.

Advanced diagnostic aids like Digital sialography and Subtraction sialography have increased sensitivity and specificity over conventional sialography techniques that were considered gold standard. Magnetic resonance sialography (MRS) is a new radiological technique indicated in cases of acute infection where other sialography techniques are contraindicated since it does not require cannulation of the duct. Diagnostic sialadenoscopy is a latest technique in which the complete ductal system can be explored\(^8\). The CBCT sialogram gives an exposure similar to fluoroscopic sialographic procedures, as performed in C-arm fluoroscopy X-ray units, but higher than an examination performed with plain or static images. It delivers a slightly higher radiation exposure and should therefore be reserved for more difficult cases\(^9\).

The present case is a giant sialolith reported in the right submandibular duct, measuring about 34mm x21mm and treated using transoral approach.

**Case Report**

A 65-year-old patient presented to the department of Oral and Maxillofacial Surgery, with a chief complaint of swelling in the anterior part of the right side of the floor of the mouth (Fig 1). The patient gave history of pain associated with peanut size swelling initially and slowly increased to present size, increase in size during chewing and associated with mild difficulty during intake of food. Patient also revealed a history of calcium supplements usage, secondary to femoral fracture since two years. Extra-oral examination revealed a palpable, firm swelling in the right submandibular region, non tender, non compressible and non reducible in nature. Intra oral examination revealed redness(inflammation) over right posterior region, mesial to right third molar (Fig 2) and bimanual palpation revealed a large, firm, non-tender swelling in the right anterior floor of mouth in the region of the submandibular duct. A paranasal sinus radiograph view showed the
mass to be radiopaque in the posterior region and extending back beyond the mandibular right first permanent molar (Fig 3). Additional investigations like ultrasonography and 3D CT scan were made to confirm (Fig 4,5,6) diagnosis which in turn revealed right submandibular duct sialolith measuring about 34mm x 21mm. No other relevant medical and dental history could be elucidated. A clinical diagnosis of submandibular sialolith was made and subsequently, a treatment plan of surgical excision of sialolith with transoral approach was decided.

Figure 1. Patient with swelling in the right sub mandibular region

Figure 2. Intra oral showing inflamed area due to penetration of sialolith

Figure 3. PNS radiograph showing radiopaque mass in the posterior region

Figure 4. Panoramic radiograph image showing sialolith extending to mandibular right first permanent molar area.

Figure 5. Ultrasonographic image of the sialolith in the submandibular duct.
Under general anesthesia, surgical technique was initiated by passing a suture around the duct posterior to calculi (stay suture) to prevent its dislodging or moving into less accessible region. The tongue was retracted towards the opposite side and the needle was passed well behind the calculus to pick up a deep bite of tissue over the duct. The floor of the mouth was lifted up with this suture. Starting beyond the papilla on the other side, the floor of the mouth was infiltrated with local anesthetic and adrenaline solution, to help in local hemostasis. A 4 mm incision anterior to the anticipated position of calculus was then made which bisected the angle between the lateral attachment of the tongue to the floor of the mouth and the plica sublingualis (fig 7). The wound was deepened gently with scissors keeping lateral to the sublingual veins and the duct was identified (fig 8). Once the duct was located, it was mobilized and the dissection was carried anteriorly towards the first molar region; taking into consideration that lingual nerve which passes under the duct is identified and protected (fig 9).

The duct was incised longitudinally over the sailolith and it was removed. The posterior stay suture round the duct was removed and saliva was expressed by pressure over the lower pole of gland. The remaining stay sutures were subsequently removed and the floor of the
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Figure 10. Excised sialolith measuring 34 x 21 mm.

Figure 11. 1 year post operative panoramic radiograph.

The mouth is closed with 3’0 silk black silk. It was measured to be 34mm x 21mm long along its greatest length (Fig 10). Due to the swelling in the right submandibular region and the size of the sialolith, the patient was reviewed alternate weeks post operatively to check salivary function of the gland. On review the right submandibular gland was palpable and clear saliva could be expressed from the duct on massage. These post operative sialochemistry revealed presence of calcium phosphate with small accounts of hydroxyapatite, magnesium, potassium and ammonia, reviews showed uneventful healing with absence of post-surgical complications till a follow-up period of one year (Fig 11).

Discussion

Salivary sialoliths are usually small and measure about 1mm to less than 1cm. Giant sialoliths measuring size more than 3cm are rare, with few reported cases\(^5\). Messerly\(^6\), Brusati and Fiamminghi\(^7\) removed a giant sialolith from the submandibular duct measuring greater than 30mm\(^4,6\). Thus, the present case, reports the history and successful management of such a rare giant sialolith of the submandibular duct.

In the present case, ultrasonography and 3D CT scan were used along with bimanual palpation and conventional radiographs to exactly locate the sialolith. Yoshimura Y\(^8\) et al. found that the detection rate of sialoliths using ultrasonographic images was higher when compared to sialography\(^6,8\). Sialoliths less than 3mm cannot be detected using ultrasonographic examination\(^8\).

The algorithm for the management of sialolithiasis depends upon the size and location of sialolith. In the management of small sialoliths, conservative methods such as proper hydration of the patient, application of moist warm heat and massaging the gland in conjunction with sialogogues can be done\(^8,13\). Since the sialolith in this case was larger measuring 34mm x 21mm, surgical excision using transoral approach was considered to completely expose and remove it. Zenk KJ et al\(^14\) suggested that transoral removal is the treatment of choice for submandibular sialoliths, which can be bimanually palpated and localized by ultrasonography. The submandibular sialoliths which are located close to the orifice of Warthin’s duct can be excised by Sialodochoplasty\(^15\). If the sialolith is too large or located in the proximal duct, piezoelectric extracorporeal shock wave lithotripsy (ESWL) or surgical removal of the stone or gland may be required.

Sialoendoscopy is a new treatment modality and minimally invasive technique for treating obstructions of the ductal system and for removal of large salivary stones\(^5,6,9,11\). Co2 laser technique have the advantage of less bleeding, minimum scarring with minimal post operative complications\(^17\).
Conclusion
Although various advanced diagnostic and treatment modalities have emerged in the management of sialoliths, the conventional techniques retain their popularity to date. A case of giant submandibular sialolith (34mm x 21mm) is reported which was diagnosed clinically and radiographically and treated with no postoperative complications.

References