Case study

Submandibular Sialolithiasis: Multiverse Presentation Of Four Cases

Abstract
Sialolithiasis is caused by the development of a calculus in the salivary gland or duct, often observed in the oral region. This disease is mostly seen in adults or young adults and seldom develops in children. Of all the cases of sialolithiasis, only 3% are seen in the pediatric population. The clinical presentation typically consists of a painful swelling of the involved salivary gland at meal times. The clinical signs often lead to an easy diagnosis. Most commonly affected isthe submandibular gland 92%, followed bythe parotid 6% and sublingual and minor salivary glands 2%. Here, we reportthe4 cases of sialolithiasis of the submandibular gland with multiverse presentation.

INTRODUCTION

Salivaryglanddiseases constitute a heterogeneous group of lesions of great morphological variations like neoplastic and nonneoplastic. Among Non-neoplastic lesions,sialolith is 2nd most common. Sialoliths are calcified structures that develop within glandular tissue parenchyma of the major and minor salivary glands and in ducts. These are usually round or oval in shape of variable sizes leading to partial or complete obstruction of the salivary duct. They may exhibit recurrence.[1] 80-90% of the salivary gland and duct calculi are found in the submandibular gland, 5-10% in the parotid gland, and approximately 0-5% in the sublingual and other minor salivary glands.[3]Salivary stones are mainly composed of calcium phosphate with smaller amounts of carbonates in the form of hydroxyapatite, smaller amounts of magnesium, potassium and traces of ammonium along with organic material mainly composed of carbohydrates and amino acids.[7]

The incidence of sialolithiasis peaks in the third to sixth decade of life. Salivary calculi in the pediatric population comprise only 3% ofcases and are rarely bilateral. Most calculi are relatively smaller (<1 cm, 93.1%) in children and located in the distal duct (62%).[2]

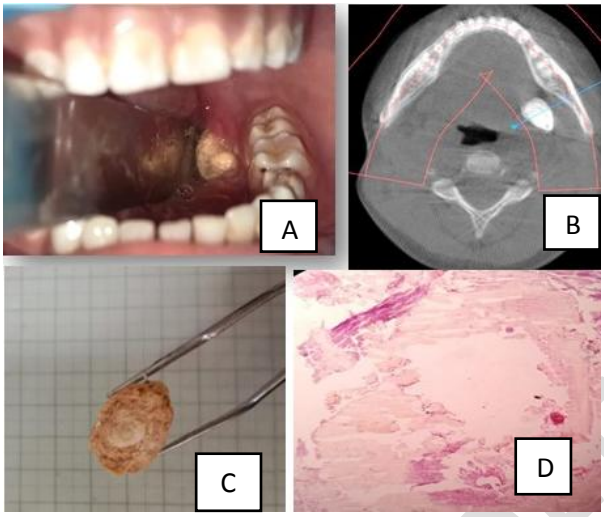
Submandibular gland sialolithiasis is more common because of the anatomical factors associated with the formation of sialoliths in this gland. The Wharton's duct of the submandibular gland is the longest duct among all salivary gland ducts with the path of the duct going in an upward direction (antigravity flow). Also, the main portion of duct is wider than its orifice. Along with these anatomical factors,the submandibular gland saliva is alkaline in nature and rich in mucin, which can promote the formation of a sialolith.[4]

The aim of this article is to report a series of 4cases of sialolithiasis of the submandibular gland of multiverse presentation.Two caseswereof giant sialolith in which case one was sialolith in the posterior region of the oral cavity in a 12 year old child which was unusual considering the age of the patient, another case was 72 year old female having sialolith in the left submandibular salivary duct. 3rd case was 62 year old female having sialolith at the orifice of the submandibular salivary duct. Another was a case of Wharton's duct sialolith which recurred within 2 months after surgical removal.

Case presentation

Case 1

A 12-year-old female child reported withchief complaints of pain in the lower left molar tooth, and swelling in the sublingual posterior region from the past 7 months. She complained of mild pain which increased during meal times with dryness of the mouth for 2 months. She had already taken antibiotic therapy.There was no improvement in the intensity of pain and size of the swelling. Extra oral examination revealed a swelling in the leftsubmandibular region around 1.5 × 2 cm in size, which was soft on palpation and did not appear to be fixed to any underlying structures. Intraoral examination revealed a yellowish-white calcified mass in the left posterior sublingual region with fair oral hygiene.Surgical excision was done under local anesthesia and the specimen was sentfor histopathological investigations.



(FIGURE 1A)Intraoral examination: yellowish-white calcified mass in left posterior sublingual region and fair oral hygiene.

(FIGURE 1B) The CBCT showed well-defined radiopacity which was 20×16.5 mm ze.

FIG: 1C)Gross examination yellowish white calcified mass approx. 16x 20 mm size

FIG:1D)Histopathological features 1.Numerous concentric areas of calcification of varying densities & small cavitational areas. 2.Scattered necrotic tissue areas surrounded by eosinophilic amorphous material.

Case 2

A 72-year-old female reported with chief complaints of pain in the lower left region of jaw, and swelling in the submandibular region from the past 4 months. She complained of mild pain which was increased during meal times since 15 days. On Intraoral examination revealed a swelling in lower left front region of floor of mouth, that was tender on palpation and did not appear to be fixed to any underlying structures. Her medical historywas insignificant.Surgical excision was done under local anesthesia and the specimen was sentfor histopathological investigations.

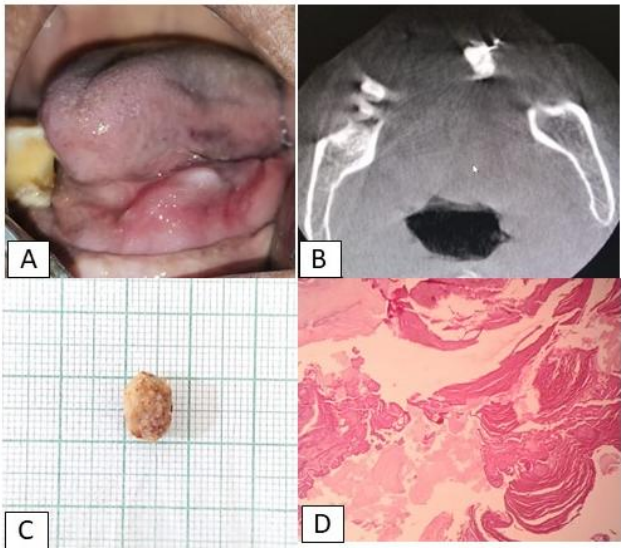


FIGURE2A) Intraoral examination:a swelling in lower left front region of floor of mouth.

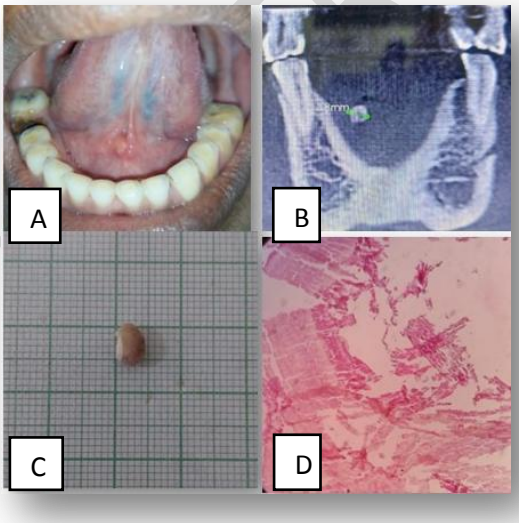
FIGURE2B) CBCT showed well-defined radiopacity which was 20×17 mm size.

FIGURE2C) Gross examination yellowish white calcified mass approx. 17x 20 mm size

FIGURE2D)) **Histopathological features**1.Numerous concentric areas of calcification of varying densities & small cavitational areas. 2.Scattered necrotic tissue areas surrounded by eosinophilic amorphous

Case 3

A 62-year-old female visited the hospital with the chief complaint of swellingunderneath the tongue. The swelling was painful on palpation. The medical history was unremarkable. On clinical examination, a yellowish mass about 3×3 mm in size was observed near the submandibular duct orifice on the right side of the oral cavity. On mandibular occlusal radiograph,a radiopaque mass was seen in the right anterolingual region. Surgical sialolithotomy was planned because removal by manual manipulation of the sialolith seems impossible. Under local anesthesia, the sialolith was removed through a minimal incision in the mucosa of the duct in which it was located, and suturing of the incision site was not performed.



3A) Intraoral photograph. Swelling on the right side of the mouth floor.
3B) Occlusal radiograph.Aradiopaque mass was observed on the right side of the mandible.
3C) salivary stone retrieved approx. 7x4 mm in size
3D) Decalcified H&Estained sections shows multiple small scattered necrotic tissues areas surrounded by eosinophilic amorphous material.

FIGURE 3(A-D)

Case 4

A 31 year old female reported for the management of pain in the floor of the mouth which aggravated during food intake. Intraoral palpation revealed that the presence of hard structure within the whartons duct closer to orifice. There was no purulent discharge from theduct. There was no lymphadenopathy.The occlusal radiograph (Figure 3B) confirmed the presence of small calculus in right submandibular duct.

After 2 months again she came with the same complaints of pain in the floor of mouth at the same site and also aggravated during meal time. her medical history was insignificant.

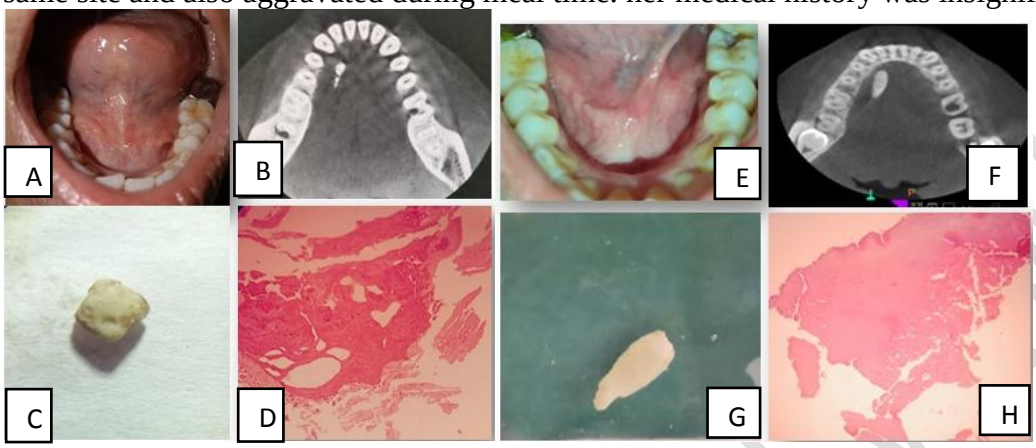


FIGURE4(A-H)

Ist occurrenceof sialolith

4A) Intraoral photograph. swelling on the right side of the mouth floor.

4B) Occlusal radiograph.A radiopaque mass on the right side of the mandible.

4C)salivary stone retrieved approx. 10x6 mm in size

4D) Decalcified H&E stained section showed multiple small scattered necrotic tissues areas surrounded by eosinophilic amorphous material.

Recurrence after 2 months

4E)After 2 month followup

4F) Occlusal radiograph on 2nd recurrence

4G) Salivary stoneretrived6x4 mm in size

4H) Decalcified H&E stained section showed multiple small scattered necrotic tissue areas surrounded by eosinophilic amorphous material.

DISCUSSION

Sialoliths are calcified accumulations that occur in the glandular tissue parenchyma of the major and minor salivary glands and in ducts. The central part of a sialolith consists of materials such as bacteria, foreign substances, and exfoliated epithelial cells on which a calcium salt is deposited concentrically to form a lamellar structure.[11]

The etiology of sialolith formation is still unknown. However, there are several factors that contribute to it. Inflammation, irregularities in the duct system, local irritants, and anti-cholinergic medications may cause the pooling of saliva within the duct which is thought to promote stone formation. Clinically, it presents as an acute, painful, and intermittent swelling of the gland, especially during a meal, when the saliva flow is increased. The degree of symptoms is dependent on the extent of salivary ductal obstruction and the presence of secondary infection.[3] The lith may totally or, partially block the flow of saliva, causing salivary pooling within the duct and gland body. The enlargement of the gland consequently

causes pain. The involved gland is usually enlarged and tender, pus may be seen draining from the duct and signs of systemic infection may be present. Stasis of the saliva may lead to infection, fibrosis, and glandular atrophy. Sialolithiasis is considered the most frequent cause of acute and chronic sialoadenitis.Sialolithiasis must be differentiated from other diseases that cause swelling in salivary glands, such as acute or chronic bacterial sialadenitis, viral sialadenitis, radiation, and systemic diseases such as, sarcoidosis, and sjögren's syndrome. Moreover, complications of sialolithiasis include: secondary infections, abscess, salivary duct stenoses, mucocele, Kuttner's tumor and glandular parenchyma atrophy in chronic states.[11]

Sialoliths occur mainly in middle-aged adults, and are rare in pediatric patients. The prevalence of sialolithiasis in children is low because the condition requires a considerable amount of time to develop,and cross-sections of the salivary glands are very small, making invasion by foreign substances difficult.[1] In addition, concentrations of calcium and phosphorus in the saliva increase with age, facilitating sialolith formation in adults.[6] In children, saliva flow is rapid; thus, most sialoliths are located distally.Conflicting information on the incidence rates in male and female has been presented. Shinohara et al.Reported that sialolithiasis occurs more frequently in girls than in boys, with a rate of 1:1.6 in children younger than 10 years. However, Nahlieli et al.Reported a higher incidence of sialolithiasis in boys than in girls. Commonly, a sialolith measures from 1 to less than 10 mm, with a mean size reported as 6-9 mm. Giant sialoliths are rare and classified as those measuring >15 mm in one dimension.[5] Out offour cases,case 1 and case 2representeda giant sialolith as size of sialolithapprox. 18x20 mm.

Basic imaging methods of sialolithiasis are X-ray images, X-ray sialography, ultrasonography (US), computed tomography (CT), and magnetic resonance imaging (MRI). Sialoendoscopy, which is also a therapeutic method, is becoming increasingly popular.A major role in sialolithiasis diagnostics is played by unenhanced computed tomography, ultrasonography and digital subtraction sialography. Also,MRI sialography is gaining on popularity.Intraoral and extraoralX-ray images allow for opaque calculi visualisation . Approximately 80–90% of the stones are opaque on a standard X-ray. However, up to 20% of the calculi cannot be revealed with a review X-ray. It was observed that the stone of the submandibular gland produces opacity only in 80–90% of the cases, while the salivary duct stone of the parotid gland only in 60%. [9]

Sialolith structure can be seen histopathologically in concentric and irregular patterns, with high and low mineralization. In H&E staining, it appears as alternating eosinophilic and basophilic zones or globular calcified zones, formed by organic and inorganic materials. Basophilic zone indicates a highly mineralized area. Whereas the eosinophilic zone indicates less mineralization. Frequently, the core is predominated by organic materials while inorganic materials form the concentric pattern. In many studies, the core is globular and highly mineralized or only composed of mineral/inorganic materials.[10]

In the reported cases,(FIG:1D)Histopathological features**:** 1.Numerous concentric areas of calcification of varying densities & small cavitational areas. 2.Scattered necrotic tissue areas surrounded by eosinophilic amorphous material.(FIGURE. 2D, 3D, 4D &4H)

The management of sialoliths is decided based on the topographyof the gland, the size and number of stones, the functionalstate of the gland, the degree of superinfection, and the surgeon'stechnical skill. Selection of treatment mode also depends on the preservationof gland function, discomfort to the patient and low levelof complications.[7]

The Conservative approach, includes analgesia, hydration, local heattherapy, and milking of the gland to expel the stone and maintain salivary flow. Discontinuation of anticholinergic

medications can alsobe recommended and in case of gland superinfection antibioticscovering oral flora are suggested. Gland massage after everymeal with daily intake of at least 1.5 L of water and sialagoguesadministration are also conservative methods of management.[11]
The recurrence rate of sialoliths is 1-10% .Notably, a study of patients with persistent, residual or recurrent sialolithiasis, Koch et al. Reported a range of 2–11 stones in 16 patients, with a single patient reported to have 11 stones. These patients had failed initial treatment with extended transoral duct surgery, 49% due to recurrent disease, and were referred to a tertiary referral center for salivary gland disease. One of the case similarly failed initial therapy with a ductal incision and had a symptomatic recurrence. This suggests that certain patients are predisposed to the formation of a greater number of stones, though the risk factors remain unclear, and that gland excision and ductal clearance early on may preclude continued symptoms. Invasive management of sialolithiasis consists of extracorporeal lithotripsy, sial endoscopy and surgery.

CONCLUSION

Sialolithiasis is not commonly observed in children but should be considered in the differential diagnosis in patients who present with submandibular swelling and pain. Establishing a diagnosis of sialolithiasis requires a thorough history and physical examination along with routine radiographs.
The analysis of the biochemical composition of saliva may play a role in recurrence which needs to be further investigated. Further research may help to prevent recurrence.

REFERENCES

1. Cornell mccullomIII, DDS Cameron Y.S. Lee, DMD David I. Blaustein, DDS, phdd. Sialolithiasis in an 8-year-old child: case report (Pediatr Dent 13:231-33, 1991)
2. Su-Young Park1 , Sang-Ho Lee1,2, Nan-Young Lee1,2, and Myeong-Kwan Jih1,2*Sialolithiasis in children: Three case reports Oral Biol Res 2019;43(4):340-348
3. Tulasi Lakshmi D, Firoz Babu P, Negi LS and Nayyar AS. Recurrent Sialadenitis with Sialolithiasis of Submandibular Gland: A Case Report. J Dent App. 2016; 3(4): 358-360
4. Sandeep Pachisia et al, Submandibular sialolithiasis: A series of three case reports with review of literature Clinics and Practice 2019; 9:1119 doi:10.4081/cp.2019.1119
5. Thong H, Othman I, Ramli R, et al. (November 10, 2021) Painless Giant Submandibular Gland Sialolith: A Case Report. Cureus 13(11): e19429. DOI 10.7759/cureus.1942
6. Constantino Ledesma-Montes, DDS, msc, phd,*Giant Sialolith: Case Report and Review of the Literature, J Oral MaxillofacSurg 65:128-130, 2007
7. Shamita Tiwari., et al. "Multiple Recurrent Obstructive Sialolithiasis of Left Wharton's Duct - A Case Report". Acta Scientific Otolaryngology 3.12 (2021): 26-30
8. Morcos Nakhla a et al, The gland that kept on giving: A case report of over 40 sialoliths from a single submandibular gland https://doi.org/10.1016/j.xocr.2022.100464
9. Iwona Rzymska-Grala1 et al. Salivary gland calculi – contemporary methods of imaging © Pol J Radiol, 2010; 75(3): 25-37
10. Kezia Rachellea Mustakim1,*et al Histopathology and ultrastructural findings of pediatric sialolithiasis: a brief communication https://doi.org/10.5125/jkaoms.2022.48.2.125

11. Kraaij S., *et al*. “Salivary stones: symptoms, aetiology, biochemical composition and treatment”. *British Dental Journal* 217(2014): E23.