ISSN: 0975-3583,0976-2833 VOL12, ISSUE 03, 2021

LARGE ASYMPTOMATIC SIALOLITH OF WHARTON'S DUCT- A CASEREPORT

Dr. Gokul Venkateshwar
Professor, Dept of Oral and Maxillofacial Surgery
Dr. Ekta Keswani
Professor, Dept of Oral and Maxillofacial Surgery
Dr. Veenita Singh
Professor, Dept of Oral and Maxillofacial Surgery
Dr. Swapna Nayan
Professor, Dept of Oral and Maxillofacial Surgery
Dr. Tanvi Mahale
Resident, Dept of Oral and Maxillofacial Surgery
Dr. Nargis Rahman
Resident, Dept of Oral and Maxillofacial Surgery

ABSTRACT

Sialolith is relatively common (80%) in the submandibular salivary gland due to the viscous nature of its mucinous secretions, high calcium content, and its tortuous ducts. In the present case, the patient presented with a history of swelling below the tongue for the past 5 years. This case presents an asymptomatic sialolith discovered by an accidental finding by the dentist, however, they may be symptomatic in cases obstructing the salivary flow. The diagnosis of submandibular sialolith was made through clinical examination and intraoral occlusal radiograph. Our article presents a review on a giant submandibular sialolith covering the aetiology, diagnosis,various treatment modalities available for management of salivary gland calculi and a summarisation of various giant sialoliths reported till date , alongwith a case report of an intraductal stone removed intraorally.

KEY WORDS- Sialolithiasis, giant sialolith, sialolithotomyINTRODUCTION

Sialolithiasis is a benign (non-malignant) condition involving the formation of stones due to calcifications within the ducts of the major salivary glands of the head and neck. These include the parotid glands, submandibular glands, and sublingual glands. Sialolithiasis is the most common cause of salivary gland swelling with a reported incidence of 1 in 10000 to 1 in 30000.[1] Submandibular gland has the highest predilection for sialolithiasis with 80% occurrence rate, followed by the parotid (19%) and the sublingual (1%) glands.

Factors responsible for higher prevalence in the submandibular gland are-Anatomic factors-

- Length and tortuous course of the duct
- Located in dependent position
- Greater size and position of orifice
- Orifice smaller than lumen Pathologic factors-

ISSN: 0975-3583,0976-2833 VOL12, ISSUE 03, 2021

- High mucin content
- Greater alkalinity and organic matter
- Greater concentration of calcium and phosphate salts
- Low carbon dioxide content
- Rich in phosphatase enzyme

Sialolithiasis is usually seen between the age of 30 and 60 years. Males are affected twice as much as females[2].

It is uncommon in children as only 3% of all sialolithiasis cases has been reported in the pediatric population.

Individuals with sialolithiasis tend to be asymptomatic, however a small proportion can have an intermittent facial swelling associated with eating,which can be painful or painless.

Symptoms are usually unilateral in nature.

When the gland is bimanually palpated, saliva can be seen at the duct orifice, along with the presence of small stones. On palpation, a stone may be palpable in the duct and the gland may feel tender in the presence of infection.

In this article, we present a case of a large (20 mm) sialolith in the whartons duct of the left submandibular gland which was present superficially, treated with surgical excision and a review of existing literature of sialolith emphasizing on the theories of clinical features, etiology, differential diagnosis and treatment modalities for the same.

CASE REPORT

A 60-year-old male patient was referred to the corresponding author due to an incidental finding by his dentist.

No extraoral swelling was palpable. No relevant medical history was present.

An intraoral swelling on the floor of the mouth was seen. The swelling was present since 5 years but was not associated with pain or pus discharge.

Clinical examination revealed a superficial, 3 cm hard, non-tender swelling palpable near the lingual frenum with mild elevation of the tongue. There was no associated pus discharge or bleeding from the swelling.

Clinical examination revealed a well demarcated swelling at the anterolateral aspect of the floor of the mouth. The swelling was present on the left side of the floor of the mouth. On palpation,the swelling was hard, non-tender. No other clinical signs were noted. Extraoral examination was insignificant.

INVESTIGATIONS

1. OPG (Orthopantomogram)2.Occlusal view X ray 3.USG (Ultrasonography)

ISSN: 0975-3583,0976-2833 VOL12, ISSUE 03, 2021

2. OPG showed no findings
3. Occlusal view X ray revealed an oval radio opaque calculus on the left side measuring approximately 3 *2 cm as shown in fig 1.
4. USG report revealed cervical lymphadenopathy with few subcentimeter to centimetre sized non-necrotic lymph nodes, bilateral cervical levels largest measuring 1.9*0.6 cm in the left cervical level II. Partially necrotic lymph nodes measuring 0.7*0.5 cm in the left cervical IA and V.

Based on clinical and radiographic findings,we arrived at a preliminary diagnosis of submandibular gland sialolith.

Fig 1- Sialolith in the floor of the mouth

PROVISIONAL DIAGNOSIS

Sialolith in the left Wharton's duct. The various differential diagnosis of intraoral swelling near the floor of the mouth are given in table 1.1.

TREATMENT

The standard treatment protocol for the management of sialolith is based on its location and the symptoms associated with it. Moist warm heat application with administration of sialogogues and massaging of the gland helps in flushing the sialolith out of the duct.

Invasive management of sialolithiasis consists of extracorporeal lithotripsy, sialendoscopy and surgery [14].Regarding the surgical treatment, an ablative surgery is performed. Indeed, the treatment of choice for patients with sialoliths that are bimanually palpable and/or which are located within the prehilar region of the gland is a transoral surgery.

We performed the surgical procedure under general anaesthesia . After the intubation and

ISSN: 0975-3583,0976-2833 VOL12, ISSUE 03, 2021

scubbing, painting and draping under aseptic conditions, the surgical field was infiltrated with 1:200000 lignocaine with adrenaline.The duct, together with the oral mucosa, was incised from the ostium until the sialolith was visible (Fig 2,3). Following the incision the sialolith was visible to be present submucosal in the floor of the mouth (Fig 4).

The swelling/sialolith was palpated and a longitudinal incision along the whole length of the swelling was placed. After initial control of bleeding, blunt dissection was carried out to expose the whole length of the sialolith.The sialolith was then detached from the duct,without causing any damage to the duct. The sialolith was excised and the wound was closed in layers using 4 0 vicryl (Fig 5,6). Patient was extubated and the recovery was uneventful. Follow up was done at 1 month, 3 months, 6 months and 1 year.

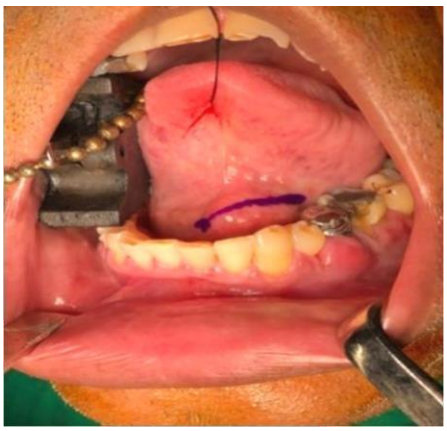

FIG 2: Clinical view of a longitudinal incision marked on the floor of the mouth

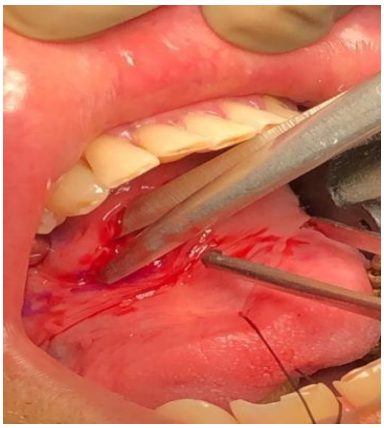

FIG.3: Flap reflected over the sialolith

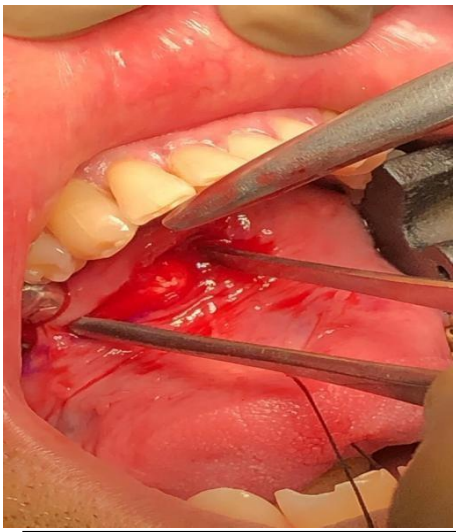

Fig 4-Sialolith exposed

Fig 5-sialolith removed

ISSN: 0975-3583,0976-2833 VOL12, ISSUE 03, 2021

Fig 6: closure done with 4 0vicryl

DISCUSSION

The sialolith/salivary gland stone is a calcified structure that forms inside a salivary gland or its duct, these include parotid gland,submandibular gland and sublingual glands. Basically, sialoliths are condensations of calcium salt; primarily calcium phosphate in the form of hydroxyapatite with small amounts of magnesium carbonate and ammonium. Wakely reported the distribution of sialoliths: 64% in submandibular gland and duct, 20 % in the parotid gland and duct and 16% in the sublingual gland and duct[2] .Salivary stones are known to be formed of an organic component (bacteria or desquamated cells) around which mineral salts have precipitated. Sialoliths are primarily composed of inorganic material [3,4].They usually contain calcium phosphates, either as carbonate apatite or hydroxyapatite, whitlockite and brushite [1,5,6]. Precipitation of calcium is usually explained by salivary stasis or decreased salivary flow [7]. Other inorganic components such as silicon, ferrum, brimstone, potassium and chloride can also be found but only in a small proportion [8]. Ammonium and magnesium can also be found in stones retrieved from an infected gland [6]. On the other hand, the organic material consists of neutral and acid glycoproteins, collagen, lipids, other proteins and carbohydrates such as mannose and glucose [23,9] .

Giant sialoliths are rare findings in clinical oral pathology with sizes ranging from 35 to 70 mm and all of them occurring in male patients. Although,giant sialoliths have been reported in the salivary glands, they have rarely been reported in the salivary ducts.The largestsialolith reported in literature was 70mm in length in wharton's duct and was described as having a 'hen's egg' size.The weight of the giant sialolith can vary from very light specimens of

ISSN: 0975-3583,0976-2833 VOL12, ISSUE 03, 2021

approximately 12 gms to heavy specimens of 93 gms[16].

Submandibular salivary gland saliva is known to be more viscous than parotid gland saliva due to a higher mucin concentration. Its pH is higher and contains twice as much calcium as parotid saliva [3,5,10]. The accumulation of calcium combined with the increase in pH decreases the solubility of calcium phosphate in saliva and thereby favours mineralization of a mucoid gel formed in the ductal system of a submandibular salivary gland.

The ability of a calculus to grow and become a giant sialolith depends mainly on the reaction of the affected duct. If the duct adjacent to the sialolith is able to dilate, allowing nearly normal secretion of saliva around the stone, it might be asymptomatic for a long period and eventually a giant calculus/sialolith will be formed [16] .

Etiological factors are divided into two groups-

1. Anatomical factors affecting saliva formation or flow (i.e., duct stenosis or inflammation/infection).
2. Composition factors (i.e., increased calcium content or altered enzyme function).

Research examining the geographic distribution of hard water and salivary calculi formation demonstrated no correlation with an increased incidence of salivary stone formation in areasof increased water hardness[11] .

Additional factors such as decreased fluid intake and pharmacologic side effects resulting in decreased salivary production (i.e., diuretic use); however, research in these areas remains limited. In recent years tobacco smoking has been discussed as a potential risk factor for the formation of salivary stones. Proposed mechanisms include the development of inflammation within the salivary ducts and decreased production of salivary amylase[1][12] .

In contrast to the small size calculi, 20 to 30 percent of which are radiolucent, giant silaoliths are mostly radiopaque and are easily depicted on panoramic radiographs, probably because their lithogenesis is long enough for calcification to be completed.

The treatment objective for giant sialoliths, as for the standard size stones is restoration of normal salivary secretion. A careful case history and correct imaging techniques are required to confirm the clinical diagnosis and to define the precise location of the calcification[17]. The giant sialolith should be removed in a minimally invasive manner via a transoral sialolithotomy to avoid the morbidity associated with sialadenectomy[16]. The cardinal rule

ISSN: 0975-3583,0976-2833 VOL12, ISSUE 03, 2021

when performing stone removal from submandibular gland is by cutting directly into the stone in longitudinal access of the duct while carefully dissecting the limgual nerve.There are newer methods of treatment for small sialoliths which include extra corporeal shock wave lithotripsy, endoscopic intracorporeal shock wave lithotripsy and laser lithotripsy,etc.

CONCLUSION

Giant submandibular gland stone are a rare entity. A careful history and correct imaging techniques are required to confirm the clinical diagnosis and to define the precise location of the calcification. Occlusal radiographs prove to be very useful in diagnosing sialoliths, in spite of more advanced and effective methods.There are various treatment protocols for the management of salivary stones, depending on the gland affected and stone location.

Asymptomatic giant sialolith of remarkable size may pose both diagnostic and therapeutic challenge for the clinician. Newer treatment modalities are effective alternatives to conventional surgical excision for smaller sialoliths.

REFERENCES

1. Huoh KC, Eisele DW. Etiologic factors in sialolithiasis. Otolaryngol Head NeckSurg. 2011 Dec;145(6):935-9. [PubMed]
2. Submandibular sialolithiasis: A series of three case reports with review of literature
3. Sandeep Pachisia, Gaurav Mandal, Sudipto Sahu, and Sucharu GhoshClin Pract. 2019 Jan 29; 9(1): 1119.
4. Kraaij S, Karagozoglu KH, Forouzanfar T, Veerman ECI, Brand HS. Salivary stones: symptoms, aetiology, biochemical composition and treatment. Br Dent J 2014;217:E23.
5. Harrill JA, King JS, Boyce WH. Structure and composition of salivary calculi. Laryngoscope 1959;69:481–492.
6. Teymoortash A, Buck P, Jepsen H, Werner JA. Sialolith crystals localized intraglandularly and in the Wharton's duct of the human submandibular gland: an X-ray diffraction analysis. Arch Oral Biol 2003;48:233–236
7. Grases F, Santiago C, Simonet BM, Costa-Bauzá A. Sialolithiasis: mechanism of calculi formation and etiologic factors. Clin Chim Acta 2003;334:131–136.
8. Marchal F, Kurt AM, Dulguerov P, Lehmann W. Retrograde theory in sialolithiasis formation. Arch Otolaryngol Head Neck Surg 2001;127:66–68
9. Su Y, Zhang K, Ke Z, Zheng G, Chu M, Liao G. Increased calcium and decreased magnesium and citrate concentrations of submandibular/sublingual saliva in

ISSN: 0975-3583,0976-2833 VOL12, ISSUE 03, 2021

sialolithiasis. Arch Oral Biol 2010;55:15–20.

10. Jayasree RS, Gupta AK, Vivek V, Nayar VU. Spectroscopic and thermal analysis of a submandibular sialolith of Wharton's duct resected using Nd:YAG laser. Lasers Med Sci 2008;23:125–131.
11. Tanaka N, Ichinose S, Adachi Y, Mimura M, Kimijima Y. Ultrastructural analysis of salivary calculus in combination with X-ray microanalysis. Med Electron Microsc 2003;36:120–126.
12. Koch M, Zenk J, Iro H. Algorithms for treatment of salivary gland obstructions. Otolaryngol. Clin. North Am. 2009 Dec;42(6):1173-92, Table of Contents. [PubMed]
13. Marchal F, Dulguerov P. Sialolithiasis management: the state of the art. Arch. Otolaryngol. Head Neck Surg. 2003 Sep;129(9):951-6. [PubMed]
14. Management of anterior submandibular sialolithiasis
15. LucasT. Duong1*, Thomas Kakiche2, François Ferré3, LaurentNawrocki4 and Ayman Bouattour5 J Oral Med Oral Surg 2019;25:16
16. Submandibular sialolithiasis: Report of six cases
17. Vikas Elias Kuruvila, N Bilahari,1 Beena Kumari,1 and Biju James
18. William.G.Shafer,Maynard K Hine,Barnet M Levy,With contributions from E Tomich,A Test book of Oral Pathology,4 th edition,chapter 10,Page No.561-562,W B Saunders Company
19. Giant Submandibular Sialolith of Remarkable Size in the Comma Area of Wharton’s Duct: A Case Report Manjunath Rai, BDS, MDS, MOSRCS (Edinburg, UK),* and Richi Burman, BDS, MDS,J Oral Maxillofac Surg 67:1329-1332, 2009
20. Sialolith-A rare case report
21. Ramesh M*, Ramesh K**, Paul G*** JPFA, Vo!' 24, September. 2010 99

ABREVIATIONS

OPG -OrthopantomographUSG-Ultrasonography mm-millimeters

SM-Submandibular
PAR-Parotid

PAREN-ParenchymalNR-not reported