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CASE REPORT

Parotid sialolithiasis: Review of literature and 3 case reports

[1]Dr Parul Tandon, [2]Dr Harish Saluja and [3]Dr Shivani Sachdeva

[1]Senior lecturer, [2]Reader, Department of oral & maxillofacial surgery, Rural dental college PIMS,Loni Maharashtra, INDIA

[3]Department of Periodontics Rural Dental College PIMS,Loni Maharashtra, INDIA

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Corresponding author

Dr Parul Tandon
Postal address-43/8,S.P. Marg, Civil Lines, Allahabad, Uttar Pradesh (211001), India
Email: parul.tandon12@gmail.com

ABSTRACT

Sialolithiasis the formation of salivary stones due to crystallization of minerals in saliva. It can cause blockage of salivary ducts and results in painful inflammation or sialadenitis of the salivary gland. The prevalence of sialoliths varies by location. Sialolith in the parotid glands is less common when compared with that of submandibular gland. However over a period of time we observed the greater incidence of parotid sialolithiasis in our institute. This article describes review of literature regarding salivary sialolithiasis and case reports of 3 patients presenting with parotid duct sialolith.

INTRODUCTION

Salivary gland lithiasis (Sialolithiasis) refers to a type of salivary disease involving the formation of calcareous concretions, or sialoliths, within a salivary gland or its excretory duct leading to obstruction of salivary flow.

Salivary stones are mainly composed of calcium oxalate and calcium phosphate associated with a thickened mucus and cellular debris[1]. The deposit of calcium and phosphate usually occurs in the skeleton.

When it occurs in an unorganized fashion in soft tissues, it is referred to as heterotropic calcification. Heterotropic calcification which results from deposition of calcium in normal tissue despite normal serum calcium and phosphate levels is known as idiopathic calcification. Sialolithiasis belongs to the category of idiopathic calcification[2].

Review of literature

Sialolithiasis is the most common disease of the major salivary glands after mumps and accounts for approximately 30% of all salivary disorders[3]. Zenk *et al* in 1994 stated that sialolithiasis accounts for more than 50% of diseases of the major salivary glands and is thus the most common cause of acute and chronic infections [4]. It is more frequent in adults than in children [5,6,7,8]. It was found that only 6.1% of 635 patients with sialolithiasis were younger than 20 years and among these there were two cases of a solitary parotid gland stone in a 4-year old girl and a 2-year old boy [8]. Kim *et al.* [9] in 2013 reported a case of parotid sialolithiasis in 2-years old patient and therefore it is the youngest age group reported in literature. It is estimated that it affects 12 in 1000 of the adult population[10], with a higher incidence in males aged

between 30-60 years[7,11,12,13,14,15]. Cawson *et al* 1998 stated that males are affected twice as much as females[16].

The most common localization is the submandibular gland where more than 80% of sialolith are found[10]. The predominance of submandibular stones can be explained by the following aspects. Anatomically, Warton's duct is longer and wider than Stenson'sduct, salivary flow is slow and against gravity, and chemically saliva is more alkaline and rich in calcium, phosphate and mucin proteins. The parotid gland is affected in 6% of the cases, 2% in the sublingual gland and other minor salivary glands in another 2% [17]. According to Lustman (1990), 5% to 20% of sialoliths are found in parotid gland[18]. When parotid glands are involved, calculi are most often located within the ductal system like in submandibular gland. Perrotta *et al*. in 1978 [19] describd 2 cases of bilateral multiple calculi located in the parotid parenchyma. In the same year Ayani *et al.* [20] reported a case of mono-lateral parotid calculi located in parenchyma. However in 1992 Seifert *et al.* reviewed 1200 cases of major salivary glands lithiasis with a ratio for parenchymal and ductal episodes of 1:35[21].

The exact etiology and pathogenesis of formation of salivary calculi is unknown. The etiological factors involved in the sialolith formation can be classified into 2 different groups: firstly-saliva retention due to morpho-anatomic factors (salivary duct stenosis, salivary duct diverticuli, etc), secondly-saliva composition factors (high supersaturation, crystallization inhibitor deficit, etc). A combination of these factors is speculated to be involved in the formation of calculi. Chronic infection of the parotid gland and secondary trauma to its duct underlies the origin of calculus, whereas the anatomical and chemical properties of the submandibular gland predispose it to calculus formation,which further induces stagnation of saliva and invasion of bacteria[11]. One study proposed the role of foods, bacteria or foreign bodies within the oral cavity in stone formation. These may ascend the duct system and become the nidus for calcification combined with attracted inflammatory cells in the formation of parotid stones. Another theory has proposed that an unknown metabolic phenomenon can increase the salivary bicarbonate content, which alters calcium phosphate solubility leads to precipitation of calcium and phosphate ions[7]. Salivary stagnation, increased alkalinity of saliva, infection or inflammation of the salivary duct or gland and physical trauma to salivary duct or gland may predispose to calculus formation[10].

It is reported that there is no co-relation between systemic abnormalities of calcium metabolism and stone formation[7]. Other than gout, electrolytes and parathyroid hormone studies in patients with sialolithiasis have not shown abnormalities, although unlike in sialolithiasis, stones in gout are made predominantly of uric acid[22]. The proposed association between hard water areas and salivary calculi has been shown to be incorrect[23]. In 1982, Isacson G *et al.* observed that there is no significant increase in the incidence of sialolithiasis in patients with nephrolithiasis[24]. Epinatianos A. *et al.* in 1991 observed the effect of hyper-calcemia on the major salivary glands of the rat and found that, although hyper-calcemia is an important etiologic factor for nephrocalcinosis and uroliths formation, its effect on the major salivary glands seems quite week, as in hyper-calcemia, the calcium metabolism is still ruled by the autonomic nervous system, thus preventing the formation of stones in salivary glands[25]. However, one study has suggested a link between sialolithiasis and nephrolithiasis, reporting an association in upto 10% of patients [18].

Careful attention should be paid to patient history and examination in order to avoid misdiagnosis. The differential diagnosis of parotid swelling or the radiopacities on radiographic examinations, could include viral sialadentis (mumps), acute bacterial sialadentis, lymphadenitis, dental abscess, human immunodeficiency virus sialopathy, Sjogren's syndrome, calcified lymph nodes, mainly after tuberculosis infection, phlebolithiasis, myositis ossificans and, finally, salivary gland tumors or metastasis[26]. Sialolithiasis typically causes pain and swelling of the involved salivary gland by obstructing the food related surge of salivary secretion. Calculi may cause stasis of saliva, leading to bacterial ascent into the parenchyma of the gland [7] and therefore infection, pain and swelling of the gland. Pain & swelling of the concerned gland at mealtimes & in response to other salivary stimuli are especially important.

Bimanual intraoral palpation is useful in detecting stones. The parotid stones can be revealed around the orifice of Stenson's duct or along its course. Deeper parotid stones are often not palpable. Imaging modalities are very useful in diagnosing sialolithiasis. Conventional intraoral X-ray may be useful but has limited application, since 40% of parotid and 20% of submandibular stones are radiolucent[27]. For parotid stones, the masseter muscle and the buccinators are additional obstacles to visualizing the calculi in Stenson's duct. In such cases, sialography is an

adequate replacement modality which makes it possible to visualize the whole duct system. However, sialography is not viable in the setting of acute infection or in patients allergic to contrast medium. Nor it is indicated when the stone is located in the distal part of the duct, as the injected contrast medium could push the stone proximally, hence making its removal difficult[7]. Ultrasonography with high frequency linear transducer (10-13MHz) is first choice investigation for diagnosis of salivary stones. Submandibular and parotid calculi diagnosis can be readily performed with ultrasonography, for almost all intra parenchymal stones. Its sensibility decreases for ductal and in particular, for papillary stones[28]. CT scan and magnetic resonance imaging (MRI), although more complex and expensive than sialography, are currently emerging as the diagnostic tools of choice due to their advantages of accuracy and lower invasiveness. CT scan enables the detection of recently calcified salivary stones, which conventional radiography cannot achieve, although thick radiological slices could occult the stones. However, CT scan is neither capable of localizing the stone precisely within the duct, nor of visualizing the ducts and their anomalies[29]. On the other hand, MRI allows consistent and accurate visualization of salivary stones and the duct system. Sialo-MRI is a diagnostic, non-invasive method recently introduced with promising results. This technique produces sialographic images but without contrast medium injection. An important advantage of sialo-MRI is the fact that the structural anatomy of the salivary glands remains unchanged with this technique which allows an exact delimitation of the glandular acini and duct[28].

The advent of salivary gland endoscopy has been a major advance, not only in terms of providing an accurate means of diagnosing and locating intra-ductal obstruction, but permitting minimally invasive surgery that can successfully relieve blockages not amenable to intraoral approaches. It permits both diagnostic assessments a definitive treatment in the same operative session[30].

Therapeutic strategy depends upon the size, location, and number of stones. Patients presenting with sialolithiasis may benefit from a trial of conservative management, especially if the stone is small[7]. The patient must be well hydrated and the clinician must apply moist warm heat and gland massage, while sialogogues are used to promote saliva production and flush the stone out of the duct. Most stones will respond to such a regimen, combined with simple sialolithotomy when required [31,32]. In the case of parotid stones, the possibility of intraoral approach is limited to those that lie anterior to the curvature of Stenson's duct above the masseter muscle.

The facial nerve can be compromised depending on the location of the calculi. Extra-oral approach is no longer performed due to the risk of surgical scars and a lower effectiveness compared to intraoral approach[33]. Parotid stone management is more problematic as only a small segment of Stenson's duct is approachable through an intra-oral incision. In addition, opening of Stenson's duct can be complicated by subsequent stenosis of the duct.

Alternative methods of treatment have emerged such as the use of extracorporeal shock wave lithotripsy (ESWL) and more recently the use of endoscopic intracorporeal shockwave lithotripsy (EISWL), in which shockwaves are delivered directly to the surface of the stone lodged within the duct without damaging adjacent tissue (piezoelectric principle)[34]. Both extra and intra-corporeal lithotripsy are gaining increasing importance in the treatment of salivary stone disease [28]. Nahlieli & Baruchin (1997) reported the usage of a rigid mini-endoscope into the ductal system of the major salivary glands[35]. Katz in 1991[36] first introduced a flexible mini-endoscope for the same purpose. Extra-corporeal shock wave lithotripsy (ESWL) is proposed as a minimally invasive alternatives to surgery for treatment of salivary stones. ESWL has been available since 1980 for the treatment of urinary stones; in the mid 1980's the technology was applied to gallstones[37]. Extra-corporeal shock wave lithotripsy uses shock waves generated outside the body to pulverize or crush the stone inside the body. The purpose of this treatment is to disintegrate the salivary stone into concentrations smaller than 2mm to permit spontaneous or induced salivation to flush out the sandy material. More recently the use of endoscopic intra-corporeal shockwave lithotripsy (EISWL), in which shock waves are delivered directly to the surface of the stone lodged within the duct without damaging adjacent tissue (piezo-electric principle)[34].

Parotid gland removal should only be carried out for cases of sialolithiasis resistant to minimally invasive techniques. If the salivary stone is in the duct orifice, surgery to remove the salivary stone is performed using duct dilation, incision, or exposure. If it is in the duct proximal part or intra-glandular portion, salivary gland extirpation is considered[38]. In particular, if the salivary stone is in the parotid gland, parotid gland extirpation not for the superficial layer but for the entire layer is done to prevent relapse [39]. However, salivary gland extirpation may damage a nerve or cause aesthetic dissatisfaction[33].

Temporal nerve damage (16-38%), and (9%) permanent paralysis of facial nerves has been reported[40]. Frey's syndrome was rare[41]. Other side effects include sialocele, fistulous opening, infection, and hematoma[42].

We present case reports of 3 patients who reported to us with swelling in the parotid region.

Case 1- A 40 years old man reported with a chief complaint of painful swelling in the left parotid region and low grade fever since 15 days. Patient was taking medications on closed prescription. Pain was severe and intermittent in nature that aggravates on mealtimes. Extra-oral examination revealed swelling in the left parotid area with indurated overlying skin that was warm and tender to touch. Intra-orally, palpation revealed a circumscribed mobile mass approximately 1cm x 0.5cm in diameter opposite the 2nd upper molar tooth. The mass was hard in consistency and tender on palapation. The salivary ductal orifice was red and edematous with purulent discharge from the orifice. OPG was performed which showed an oval sialolith located near the orifice of left stenson's duct. (fig. 1-3).

1. Preoperative swelling over the left cheek region.

2. Preoperative- Inflamed left stenson's duct orifice

3. OPG showing radiopaque mass(left side) near crown of 1st & 2nd maxillary molars

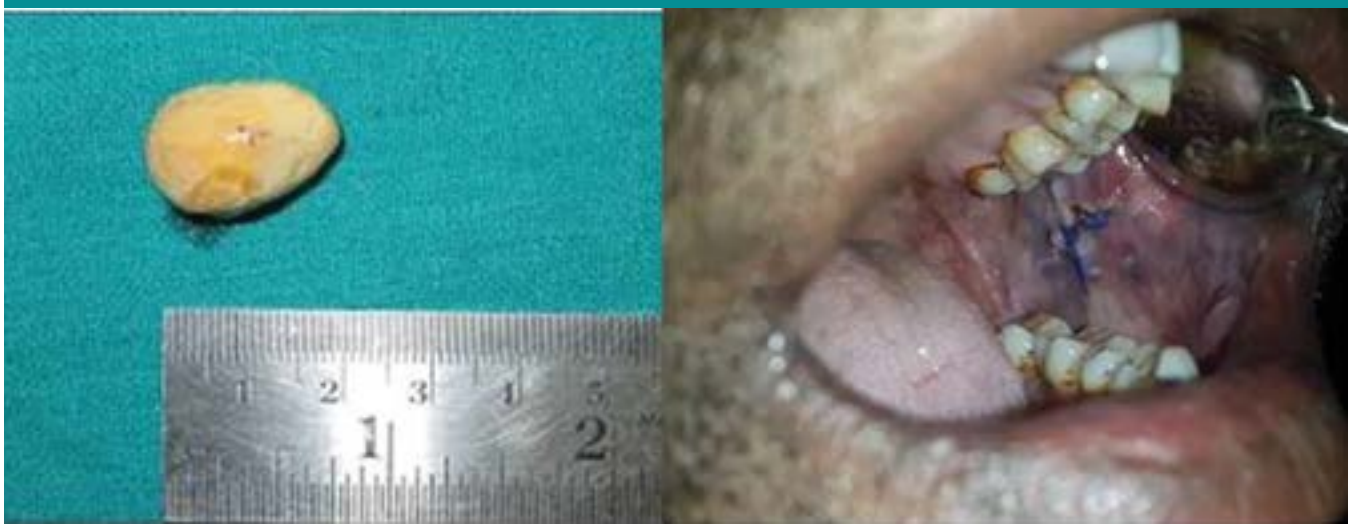
Patient was admitted and medicinal treatment was initiated with analgesic-anti-inflammatory drugs and intravenous antibiotics. On the 3rd day, swelling & fever began to subside. As the calculi was located near the duct orifice, we planned sialolithotomy by an intraoral approach under local anaesthesia. A horizontal incision was made just beneath the parotid papilla followed by the blunt dissection around the duct up to the stone's

location. The ductal layer was sutured to the oral mucosa to maintain ductal patency (Fig 4, 5). The patient was kept on parenteral antibiotics for 4 days followed by oral antibiotics for 3 additional days.

Case 2-A 35 years female patient reported with a chief complaint of pain in the right parotid region since 7 days and low grade fever. On extra-oral examination, face was bilaterally symmetrical. However, right parotid region was tender on palpation. On intraoral examination, parotid papilla was inflamed and a solitary hard mass was palpated near the duct orifice.

Case 3- A 42 years old male reported with pain and swelling in the left parotid region since 10 days. He visited to a local doctor where he was informed about the drainage of pus intra-orally and so prescribed oral antibiotics and analgesics but he was not relieved. On extra-oral examination, there was slight swelling in left parotid region and skin was warm and tender to touch. On intra-oral examination, a solid mobile mass was palpated near the parotid duct orifice which was tender. There was no pus discharge at the duct opening on pressing which could be because patient was on antibiotics. Both the case 2 & 3 were managed the same way as case 1.

4. Intraoperative



5. Postoperative: follow up after 1 week

REFERENCES

Hiraide, F. and Nomura,Y. 1980. The fine surface structure and composition of salivary calculi. *Laryngoscope* **90**:152-8.

White, S. C. and Pharoah, M. J. 2004. Oral radiology principles and interpretation. Chapter 27. In: Soft Tissue Calcification and Ossification. Mosby, Missouri: pp. 597-614.

El Deeb, M., Holte, N. and Gorlin, R. J. 1981. Submandibularslivary gland sialoliths perforated through the oral floor. *OralSurg Oral Med Oral Pathol.* **51**:134-9.

Zenk, J., Benzel, W. and Iro, H. 1994. New modalities in the management of human sialolithiasis. *Minimal Invas Ther Allied Technol.* **3**(5):275-84.

Antoniadis, D., Mendonidou, I., Papanayotou, P. and Trigonidis, G. 1989. Clinical study of sialolithiasis. Findings from 100 cases. *Hell Stomatol Chron.* **33**:245-51.

Laforgia, P. D., GFFavia, N., Chiaravalle, M. G. and Lacaita, A. 1989. Laforgia. Clinico-statistical, morphologic and microstructural analysis of 400 cases of sialolithiasis. *Minerva Stomatol.* **38**:1329-36.

Williams, M. F. 1999. Sialolithiasis. *Otolaryngol Clin North Am.* **32**:819-34.

Zenk, J., Constantinidis, J., Kydles, S., Horning, J. and Iro, H. 1999. Clinical and diagnostic finding in sialolithiasis. *HNO.* **47**:963-9.

Kim Hoon Do, Song Woo Sun, Kim Yeong Jin and Kim Won Duck. 2013. Parotidsialolithiasis in a two-year-old boy. *Korean J Pediatr.* **56** (10):451-455.

Leung, A. K., Choi, M. C. and Wagner, G. A., 1999. Multiple-sialoliths and a sialolith of unusual size in the submandibular duct: A case report. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod.* **87**(3):331-3.

Grases, F., Santiago, C. and Simonet, B. M. 2003 A Costa-Bauza. Sialolithiasis: mechanism of calculi formation and etiologic factors. *ClinChimActa.* **334**:131-6.

Chu, D. W., Chow, T. L., and Lim, B. H. 2003. SPY Kwok. Endoscopic management of submandibular sialolithiasis. *SurgEndosc.* **17**:876-9.

Austin, T., Davis, J. and Chan, T. 2004. Sialolithiasis of submnadibulargland. *J Emerg Med.* **26**:221-3.

Antoniadis, D. 1989. Mendonidou, PPapanayotou, GTrigonidis. Clinical study of sialolithiasis. Findings from 100 cases. *HellStomatolChron.* **33**:245-51.

Laforgia, P. D., Favia, G. F., Chiaravalle, N., Lacaita, M. G. and Laforgia, A. 1989. Clinico-statistical and microstructural analysis of 400 cases of sialolithiasis. *MinervaStomatol.* **38**:1329-36.

Cawson, R. A. 1998. EWOdell. Essentials of oral pathology and oral medicine. 6th edition, Churchill Livingstone: Edinburgh. pp239-40.

El Deeb, M., Holte, N. and Gorlin, R. J. 1981. Submandibular salivary gland sialoliths perforated through the oral floor. *Oral Surg Oral Med Oral Pathol.* **51**:134-9.

Lustmann, J., Regev, E. and Melamed, Y. 1990. Sialolithiasis: a survey on 245 patients and a review of the literature. *Int J Oral Maxillofac Surg.***19**:135-8.

Perrotta, R. J., Williams, J. R. and Selfe, R. W. 1978. Simultaneous bilateral parotid and submandibular gland calculi. *Arch Otolaryngol.* **104**:469-70.

Ayani, N. 1978. Multiple salivary calculi of the parotid gland. *Otolaryngology* (Tokyo) **50**:59-62.

Seifert, G. Miehlke, A., Haubrich, J. R. and Chilla. 1986. Diseases of the salivary glands: pathology, diagnosis, treatment, facial nerve surgery. Stuttgart, Germany: George Thieme Verlag. pp85-90.

Work, W. P. and Hecht, D. W. 1980. Inflammatory diseases of the major salivary glands. *In Papperalla* MM, Shumrick D F (eds): Otolaryngology, Philadelphia,WB Saunders, **3**:2235-2243.

Sherman, J. A. and McGurk, M. 2000. Lack of correlation between water hardness and salivary calculi in England. *Br J Oral Maxillofac Surg.* **38**:50-53.

Isacson G. and Lundquist, P. G. 1982. Salivary calculi as an aetiological factor in chronic sialo-adenitis of the submandibular gland. *ClinOtolaryngol.* **7**:231-6.

Epivatianos, A. and Tsoungus. M. 1991. The effect of the hypercalcemia on the major salivary glands of the rat. *Stomatologia Athenai.* **47**:306-13.

Gadodia, A., Bhalla, A. S., Sharma, R., Thakar, A. and Prashad, R. 2011. Bilateral parotid swelling: a radiological review. *Dentomaxillofac Radiol.* **40**:403-14.

Bodner, L. 1999. Parotidsialolithiasis. *J Laryngol Otol.* **113**: 266-7.

Andretta, M., Tregnaghi, A., Prosenikliev, V. and Staffieri. A. 2005. Current opinions in sialolithiasis diagnosis and treatment. *Acta Otorhinolaryngol Ital.* **25**:145-149.

Avrahami, E., Englender, M., Chen, E, Shabtay, D., Katz, R. and Harell, M. 1996. CT of submandibular gland sialolithiasis. *Neuroradiology.* **38**:287-90.

Geisthoff, U. W. 2007. Techniques for multiple modal salivary gland stone therapy. *Oper Tech Otolaryngol.* **18**:332-40.

Jr Pollack, C. V., Jr Severance, H.W. 1990. Sialolithiasis: case studies and review, *J Emergency Medicine.* **8**:561-565.

Pietz, D. M. and Bach, D. E. 1997 Submandibular sialolithasis. *General Dentistry.* **35**:494-496.

Dulguerov, P., Marchal, F. and Lehmann, W. 1999. Postparotidectomy facial nerve paralysis:possible etiologic factors and results with routine facial nerve monitoring. *Laryngoscope.* **109**:754-62.

Iro, H. Th Schneider, H. and Fodra, C. *et al*. 1992. Shockwave lithotripsy of salivary duct stones. *Lancet.* **339**:1333-1336.

Nahlieli, O. and Baruchin, A. M. 1997. Sialoendoscopy: Threeyears experience as a diagnostic and treatment modality. *J Oral Maxillofac Surg*. **55**:912.

Katz, P. H. 1991. Endoscopy of the salivary glands. *Ann Radiol.* **34**:310,1991.

DeBurgh Norman and J. E., McGurk, M. 1995. Extracorporeal piezoelectric shockwave lithotripsy of salivary duct stones. In: Color Atlas and Text Books of salivary duct and Lacrimal glands 1stEdn, Mosby-Wolfe, London, **1995**:263-266.

Hald, J. and Andreassen, U. K. 1994. Submandibular gland excision: short and long-term complications. *ORL J Otorhinolaryngol Relat Spec.* **56**:87-91.

Beahrs, O. H., 1963. The facial nerve in parotid surgery. *Surg Clin North Am* **43**:973-7.

Bates, D., O'Brien, C. J., Tikaram, K. and Painter, D. M. 1998. Parotid and submandibualrsialadenitis treated by salivary gland excision. *N Z J Surg.* **68**:120-4.

Moody, A. B., Avery, C. M., Taylor, J. and Langdon, J. D., 1999. Acomarision of one hundred and fifty consequetiveparotidectomies for tumors and inflammatory disease. *Int J Oral Maxillofac Surg.* **28**:211-5.

Amin, M. A., Bailey, B. M. and Patel, S. R. 2001. Clinical and radiological evience to support superficial parotidectomy as the treatment of choice for chronic parotid sialadenitis: a retrospective study. *Br J Oral Maxillofac Surg.* **39**:348-52.