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Submandibular Megalith With Erosion of the Floor of Mouth
A Rare Case Report

Authors: Santosh Patil*, Suchita Sharma**, Leela Krishna Prasad***
* Assistant Professor, **Intern, ***Reader

Institution: Dept of oral medicine and radiology, Jodhpur Dental College, Jodhpur National University, Jhanwar road, Naranadi, Jodhpur (Raj).342001. India.

Corresponding Author:

Dr. Santosh Patil,
Dept of oral medicine and radiology, Jodhpur Dental College, Jodhpur National University,
Jhanwar road, Naranadi, Jodhpur (Raj).342001. India.
E-mail – drpsantosh@yahoo.com

Abstract: Sialolithiasis accounts for the most etiology of salivary gland obstruction which leads to recurrent painful swelling of the involved gland which often exacerbates while eating. Stones may be encountered in any of the salivary glands but most frequently in the submandibular gland and its duct. Very few cases of giant submandibular sialoliths with intraoral perforations have been reported in literature. We report the case of a fifty year male patient who had a giant submandibular sialolith which led to erosion of the floor of the mouth. Orthopantomograph was used to confirm the clinical diagnosis. The sialolith was removed with intraoral approach and no postoperative complications were noted. The article also reviews the various available diagnostic modalities and treatment options.

Introduction: Sialolithiasis is the most common disease of salivary glands[1] and is considered as one of the major causes of salivary gland dysfunction. The majority of the studies in the literature have found a male predominance, whereas Alcure ML et al. found females being affected more often in their study. The disease most commonly affects middle aged patients, the age range being 42-58.4 years.[2] The submandibular glands is most commonly involved followed by the parotid, sublingual and minor salivary glands.[3] Intraductal stones are more common when compared to intraglandular stones.[4] The submandibular gland is most frequently involved because of its anatomic location, long, tortuous duct with a narrow orifice compared to the main portion of duct. Along with these factors, alkaline saliva rich in mucin also contributes to the stone formation.[5]

Case History: A 50 year old male patient reported with a chief complaint of a mass in left side of the floor of the mouth for one month duration. There was no significant history of associated pain or any other

complaints. The patient's medical and dental history was also not significant. Clinical examination revealed a well defined hard mass in the left anterior region perforating the floor of the mouth. The mass was a yellowish-white, non tender, indentation being noted on the ventral surface of the tongue opposing the mass (Figure-1). Orthopantomograph (OPG) revealed a well defined radioopaque mass extending from the left mandibular canine distally and apically beyond the first molar (Figure-2). On the basis of clinical and radiological findings, a diagnosis of left submandibular duct sialolith was made. The sialolith was excised surgically with intraoral approach under local anesthesia, the stone measured 38 millimetres (mm) and was yellowish-white in color. No postoperative complications were noted. **Enlarged Pictures At The End of Manuscript.**

Figure 1. Sialolith eroding the floor of mouth.

Figure 2. OPG showing well radioopaque mass.

Discussion: Most cases of submandibular sialoliths are asymptomatic. Pain and swelling may be the cardinal signs and symptoms[6] which are more pronounced on anticipation of food due to the obstruction of salivary flow.[1] Hypotheses regarding the pathogenesis suggest that, there is an initial organic nidus which progressively grows by the deposition of inorganic and organic substances or that intracellular microcalculi are excreted in the canal and act as a nidus for further calcification. In some cases, the existence of mucosal plugs acting as a nidus in the ductal system was reported. A possibility of debris, bacteria or substances migrating in the salivary ducts from oral cavity has also been suggested.[7]

Few is any systemic disorders are associated with sialolith formation, with the exception of gout which may predispose to sialolith formation.[8] In the literature, sialoliths measuring more than 15 millimetres (mm) were considered rare,[9] various reports of submandibular sialoliths measuring between 23mm and 31mm have been reported.[4,8] The sialolith observed in our case was quite large, measuring approximately 38mm and was associated with erosion of the floor of the mouth, making this reported case extremely rare.

The key step in diagnosis of sialolithiasis is the elucidation of a thorough history and careful clinical examination. Various clinical and imaging methods are available for diagnosing sialoliths, the clinical scenario with which the patient presents to the clinician defines the algorithm for salivary gland imaging. Occlusal and panoramic views are the most common radiographic techniques used to diagnose sialolith.[7] All salivary stones can not be visualized through conventional radiograph because a few of them are hypominelarized[2] and are superimposed by other radiodense tissues. In these cases other advanced imaging modalities should be considered. Ultrasonographic (US) examination is considered as a simple and non invasive modality to evaluate sialoliths especially during the acute infection. Yoshimura Y et al. found in their study that the detection rate of sialoliths using ultrasonagraphic images was higher when compared to sialography.[10] US examination is considered less accurate in comparison to computed tomography(CT) in distinguishing multiple stones.[11] It has also been reported that sialoliths smaller than 3 mm may not be detected during US examination, as they will not produce acoustic shadows.[12] Digital sialography and subtraction sialography have increased the sensitivity and specificity of conventional sialographic technique

which are considered the gold standard.[13] The major advantage of these newer techniques is the production of an image without the superimposition of overlying anatomical structures. The disadvantage being the need to use contrast agents[12] that simulates conventional sialography. These agents may expose the patient to radiation hazards, cause pain associated with the procedure, perforate the duct's wall and may be contraindicated during acute infections.[13]

CT sialography has been used to delineate the ductal system of the submandibular gland, this technique demonstrates the soft tissue of gland and ductal system with 3D reconstruction that avoids superimposition of anatomic structures. This technique has similar disadvantages that are seen with other sialographic techniques.[12] Magnetic resonance sialography (MRS) is a new technique that is considered an excellent radiological modality for the diagnosis of sialolithiasis.[13] MRS may be indicated in cases of acute infection where other sialographic techniques are contraindicated since MRS does not require cannulation of the duct.[11] Other advantages of this technique are the low radiation doses and the lack of pain associated with procedure.[13,14] The disadvantages of this technique include claustrophobia, cost factors, artifacts and contraindication in patients with cardiac pacemakers. Diagnostic sialadenoscopy is a newer technique in which the complete ductal system can be explored. It provides direct and reliable diagnostic information of ductal pathologies. The need for technical perfection is the only limitation of this technique.

The algorithm for the treatment of sialolithiasis depends upon the location and size of the sialolith. In case 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 may be considered.[13] Small stones can also be milked out through ductal orifices by bimanual palpation. Zudin KJ et al. found that transoral removal is the treatment of choice for submandibular sialoliths which can be bimanually palpated and localized by ultrasonography.[15] Sialodochoplasty can be performed to remove the submandibular sialoliths which are located close to the orifice of Warthin's duct. To remove the stones distal to the punctum, a transverse incision can be made distally on the stone taking care not to injure the lingual nerve injury,[9] as performed in our case. Batori M et al. reported a case of a 13 mm submandibular sialolith which was treated by complete surgical removal of the gland.[5] In the management of large sialoliths which are located in the close proximal duct, extracorporeal shock wave lithotripsy (ESWL) can be considered. Endoscopic intracorporeal shock wave lithotripsy (EISWL) is also gaining importance because of less damage to the adjacent tissues during the procedure.[9] Sialadenoscopy, which is a non invasive technique can be use to manage large sialoliths as well as ductal obliteration. CO2 laser, because of its advantages of minimal bleeding, less scarring, clear vision and minimal post operative complications, is gaining its popularity in the treatment of sialolithiasis.

Conclusion: Although various advanced diagnostic and treatment modalities have emerged in the management of sialoliths, the conventional techniques retain their popularity to date. We have reported a case of giant submandibular sialolith with erosion of floor of mouth which was diagnosed clinically and radiographically and treated with no postoperative complications.

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