SIALOLITH IN SUBMANDIBULAR GLAND- A REPORT OF THREE CASES AND REVIEW OF LITERATURE

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Abstract: Sialolithiasis (salivary stones) is the most common disorder of the salivary glands. The majority of sialoliths developing in the salivary glands are in submandibular gland or its duct (Wharton’s duct). There are many theories that have been proposed in the literature for the formation of salivary calculi. This article discusses review of literature, clinical and radiographic case report of three patients with sialolithiasis.

Keywords- Sialolithiasis, sialolith, submandibular gland, Wharton’s duct

Introduction
Sialolithiasis is a common disorder of salivary glands, it is a condition characterized by the obstruction of a salivary gland or its excretory duct due to formation of sialolith, further resulting in infection of salivary gland or duct. It is estimated that it affects 12 in 1000 individuals in the adult population.¹² The prevalence of sialolithiasis in children is very less. Males are affected twice as much as female patients.³ If the patient is giving the history of swelling in salivary gland region during mealtimes, then the diagnosis of ‘sialolithiasis’ should be considered. Most salivary calculi (80% to 95%) occur in submandibular gland, whereas 5% to 20% are found in the parotid gland.⁴ The reason for this could be high mineral content present in the saliva, more viscous type of salivary flow, longer and tortuous nature of salivary duct of the submandibular gland. The causative agents for sialoliths are unknown, but inflammation is the widely accepted causative condition.⁵ Commonly sialoliths found are less than 1 cm in size but in literature sialoliths of unusual shape and size are revealed.⁶ The aim of this article is to report three cases of submandibular sialolith and the discussion of its clinical aspect.
Case Reports

Case 1
A 25 year old male patient reported with the chief complaint of pain in the right submandibular region and difficulty to eat and swallow since 2 months. Patient gave the history of swelling in the same region on two occasions in past six months which subsided in some time. Medical history of the patient was not relevant.

Extraoral examination revealed diffuse swelling on the right submandibular region. (Fig 1) Bimanual palpation revealed presence of hard mass over the right submandibular gland. Intra oral examination showed a dilated duct of the right submandibular gland (Fig 2) with a normal duct of the left side.

Radiographic investigations were carried out. The occlusal radiograph of mandible revealed presence of two radiopaque mass seen on the lingual aspect of 45 to 47 on right side the one radiopacity measuring approx. 2x0.5 cm with laminated appearance; another radio-opacity seen with 48 region measured approximately 0.5x0.5 cm suggestive of calculus in the duct (Fig 3). The USG was done with high frequency transducer. The right submandibular Gland appears mildly enlarged. A 0.5 cm x 0.5 cm calculus with ductal dilatation in the right submandibular gland was seen (Fig 4).

Fig 1: Submandibular swelling right side  
Fig 2: Dilated duct, on right side

Fig 3: Radiopaque sialolith  
Fig 4: Ultrasonography of right submandibular region

Case 2
A male patient of 30 years reported with complaint of intermittent swelling in the lower left mandibular region since 4 months. Patient gave history of gradual increase in the size of swelling during meal times. The swelling used to reduce in size after 2 to 3 hours of meal. Patient also complains of decreased saliva in mouth. He had difficulty in swallowing the food. Extra oral examination revealed a diffuse swelling (Fig 5) in left submandibular region. Intraoral examination revealed no swelling, but the left submandibular duct was distinctly dilated. (Fig 6) Salivary flow was normal. The radiographic examination revealed presence of 5 cm by 0.5 cm calcified body in left submandibular region. The whole calcified structure was seen linearly in the length of the duct in the occlusal radiographic film.(Fig 7)
Case 3
A 27 year old male patient presented with a painless swelling in the right submandibular region (Fig 8). He stated that the occurrence of swelling was prominent during meal times. The medical history was not relevant. The swelling was diffused and non tender on palpation. The intraoral examination revealed no significant findings. (Fig 9) The radiographic examination was carried out. The occlusal radiograph of mandibular region showed presence of calcified structure in the region of right submandibular gland. The size of calcified body was 0.5 cm by 0.5 cm. (Fig 10)

Discussion
Sialolithiasis is a disease characterized by the development of salivary stones, known as calculi or sialoliths, in the salivary ducts or in the salivary gland themselves. Clinicians should evaluate the swellings in the submandibular region carefully, giving special attention to history of occurrence. More than 80% of salivary sialoliths occur in the submandibular gland or in its duct, 6-15% in the parotid gland or in its duct, and about 2% in the sublingual and minor salivary gland. Salivary calculi are generally small varying from 1 mm to 1 cm. They rarely measure more than 1.5 cm. Giant sialoliths are rare and defined as the size of 3.5 cm or larger. The largest sialolith reported in the literature was 70 mm in length in Wharton’s duct and was described as having a “hen’s egg” size. It is believed that a calculus may enlarge at the rate of approximately 1 to 1.5 mm per year.

It was reported that the most common symptoms of sialoliths are recurrent pain and swelling of the associated gland during meals, because the stone usually does not block the flow of saliva completely. Intraductal stones are more common when compared to intraglandular stones. Sialoliths are generally unilateral. Anatomic position of the gland, alkaline nature of saliva predispose to formation of sialolith. Generally these calcified structures are easily identified on radiographic investigations. 40% of parotid and 20% of submandibular stones are not radiopaque and sialography/diagnostic sialoendoscopy may be required to locate them.

Till date, many theories are proposed for the formation of sialoliths but exact aetiology and mechanism of formation or pathogenesis is not known. They are thought to occur as a result of deposition of calcium salts around an initial organic nidus consisting of altered salivary mucins, bacteria and desquamated epithelial cells, and as a result of flow stasis.
cells.[3] Traditional theories suggest that the formation occurs in two phases: a central core and a layered periphery.[14] A retrograde theory for sialolithiasis has also been proposed.[14] Salivary calculi are mainly made up of calcium phosphate with small amounts of carbonates in the form of hydroxyapatite, magnesium, potassium, and ammonia are components of sialoliths too.[12] Based on 120 submandibular gland sialendoscopy studies, Marchal et al., observed the presence of a sphincter system in the first 3 cm of the Wharton’s duct in 90% of their studied cases, and suggested that the variation of such sphincter-like mechanism within the salivary ducts could be responsible for easier retrograde migration of oral materials.[15]

Diagnosing a case of submandibular sialolithisis is very easy by using imaging studies. A mandibular occlusal radiograph is gold standard for identification and location of sialolith. The advanced diagnostic investigation for sialolith includes sialography, ultrasonography, computed tomography and scintigraphy. The investigation should be carried out according to size and location of the sialolith. The smaller sialoliths can be removed just bimanual palpation (milking out). Lithotripsy or surgical management become necessary if the size of sialolith is too large. Sialoendoscopy is a new way and minimally invasive technique for treating obstructions of the ductal system and can be used with the operation in the large salivary stones.[13] Long term obstruction in the absence of infection can lead to atrophy of the gland with resultant lack of secretory function and ultimately fibrosis.[10] Submandibular gland removal is indicated only when there is a stone of substantial mass (12mm or more) within the gland itself that is not surgically accessible intraorally and when there are small stones present in the vertical portion of Wharton’s duct from the comma area to the hilum.[10]

References