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Posterior submandibular sialolith retrieval-A case report and review of literature

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Abstract
Sialolithiasis is a common salivary gland disorder, of which submandibular salivary gland shows most common predilection. It is characterized by the presence of a calculus within the salivary gland parenchyma or excretory duct. Treatment mainly depends on the location and size of the sialolith. Usually large masses (more than 4mm) are easy to retrieve from the duct. But as the size of the calculi decreases it may slip off posteriorly which make the retrieval difficult without endoscopic assistance. Here we present a case report of two sialolith occluding the submandibular duct, of which the posterior one was very small. Both masses were retrieved locally without endoscopic assistance.

Keywords: abscess, sialolith, submandibular duct, retrieval

Introduction
Salivary calculi are the second most common salivary gland disorder after mumps. It affects more than 1% of the population[1]. Sialolithiasis is the formation of calcific masses either in the glandular parenchyma or ductal system or both. It can occur in any of the minor or major salivary gland. Presence of a calculus within the salivary gland or its excretory system may lead to obstruction of salivary gland which causes pathognomonic pain during meals [1, 2]. The size of calculi is variable, ranging from a few millimeters to several centimeters. When size of calculi exceeds 1.5 cm, it is called as "Giant sialolithiasis" [3]. Rai and Burman [4] is credited to report one of the largest sialolith which was about 72mm in dimension. Since such giant salivary stones cannot be accommodated in ductal system they are are usually embedded inside parenchymal tissue [3].

Case Report
A 23-year-old male patient reported with swelling and pain in relation to the left side of floor of the mouth during meals. The swelling was indurated and was tender on palpation with pus discharge from the sub mandibular duct. Doubtful of sialolithiasis, an occlusal radiograph was taken which showed two radiopaque masses medial to 45, 46 (Figure 2). The diagnosis was confirmed by cone beam computed tomography (CBCT) (Figure 3, 4). CBCT revealed a two calcified masses noted in the left submandibular region. The largest and anterior one was 8.19 mm in greatest mesio-distal dimension whereas the posterior one was 3.96 mm in mesio-distal dimension.
Under local anesthesia, the posterior end of induration was located and placed a stay suture posterior to the calculi to prevent the slippage of stone posteriorly. We then placed an incision over the stones to expose the calculi and facilitate its removal. The anterior stone was easily retrieved. But due to small size of the posterior stone, it was difficult to remove. The wound was closed without tension with 4-0 vicryl, maintaining the patency of sub mandibular.

Fig 1: Indurated sub mandibular duct

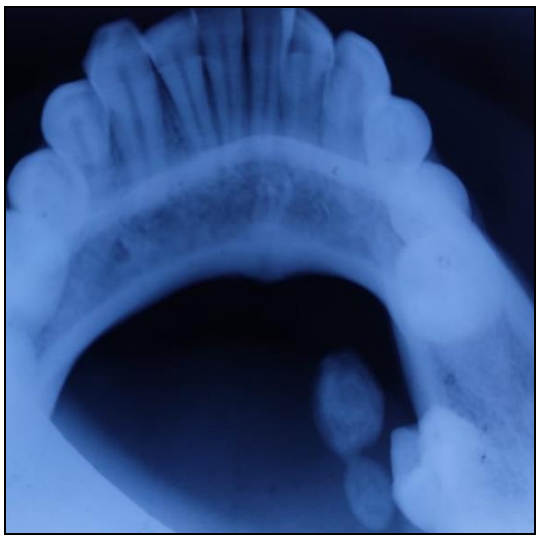

Fig 2: Occlusal radiograp

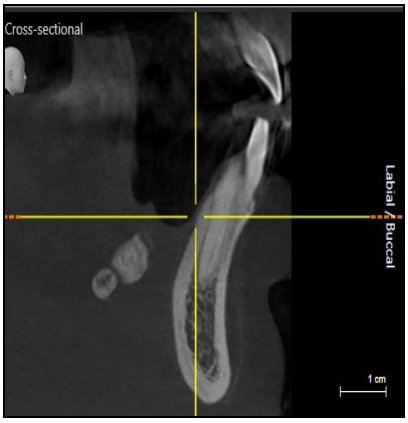

Fig 3: CBCT- Sagittal view

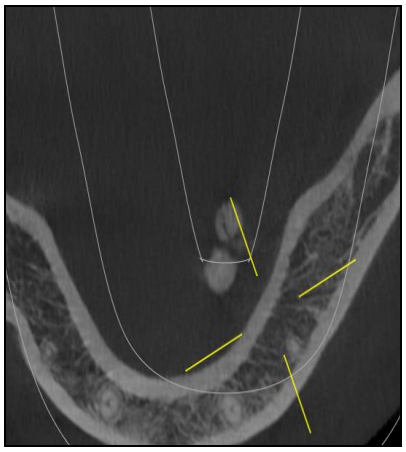

Fig 4: CBCT-Axial view

Fig 5: Anterior sialolith exposed

Fig 6: Posterior sialolith

Fig 7: Retrieved Calculi

Discussion

Prevalence

Even though majority of the cases (75%) of sialolithiasis occur on one side of the gland, there are reported cases of bilateral involvement (3% occur bilaterally. Submandibular gland is the most commonly affected gland (80% to 92% of cases) followed by the parotid gland (6% to 20%) and the sublingual and minor salivary glands (1% to 2% of cases).

Etiopathogenesis

The exact etiology and pathogenesis of salivary calculi remain unknown. Several hypotheses were found in the literature ranging from anatomical variations of the salivary ducts, agglomeration of sialomicroliths to an altered biochemical composition of saliva [5, 6]. Sialomicroliths are defined as a microscopic calcifications in a salivary gland. It consists of calcium phosphate crystals, as well as organic secretory material and necrotic cell residues [7]. There are reports suggesting that intracellular micro calculi formation acts as a nidus for the stone formation. Microlith obstruct the salivary flow which in turn elevate bacterial load, which causes obstruction the ductal system. Increase in calcium and phosphate salts in the saliva is another theory that attributes to the development of calculi [8], which, in turn, form a deposition along with desquamated cells, salivary mucus, and bacteria. Factors like stagnation of salivary flow, dehydration, and change in salivary pH, increased calcium content, and physical trauma to salivary duct or gland may predispose to calculus formation. Other potential causes for sialolithiasis are infections, salivary dysfunction, ductal anomalies, foreign bodies, and ductal epithelium metaplasia. The abnormal enlargement of calculi depends mainly on how the affected duct reacts to the mass. Sialolithiasis most commonly occur in the submandibular gland, due to the anatomy of this duct being long and its flow of saliva against gravity. Also the wharton's duct rests at the lower level of the oral cavity, and this location allows for retrograde infection of the gland by oral flora [9, 10]. The type of salivary secretions from the submandibular gland are also more mucoid in nature as opposed to the more serous secretions from the parotid gland which makes the gland more vulnerable to sialolith formation.

Composition and shape

Salivary stones consist of organic component (bacteria or desquamated cells) around which mineral salts are

precipitated. The main composition of sialoliths are mainly inorganic material [2, 4]. Mineral salts usually contain calcium phosphates, either as hydroxyapatite, whitlockite or brushite. Precipitation of calcium is usually explained by salivary stasis or decreased salivary flow [11]. Traces of inorganic components such as silicon, ferrum, brimstone, potassium and chloride can also be found but only in a small proportion [12]. When the gland is infected, organic material like neutral and acid glycoproteins, collagen, lipids and are found [13]. The shape of calculus depends on its origin. It will be elongated if it is developed in the ductal system and oval or round if it originates from the hilus or gland.

Pre disposing Factors

High saliva viscosity, higher salivary protein content, higher salivary calcium concentrations than unaffected patients, reduced concentrations of crystallization inhibitors citrate, magnesium and phytate are predisposing factors for the formation of sialothiasis [12]. Gout appears to be systemic factor associated with a predisposition to sialolithiasis formation due to the abnormal uric acid level [14]. Use of diuretics was proven to predispose the formation of salivary stones as one of its known effects consists of decreasing the salivary flow [15]. But surprisingly even though there is hypo function of salivary gland with decrease in salivary flow in Sjögren's syndrome, no increased incidence of salivary stones were found among these patients [16].

Clinical Features

There will bea subsequent increase in intraglandular pressure due to the complete obstruction of the salivary flow caused by a sialolith. This leads to an accumulation of saliva. At mealtimes, the obstruction of the excretory duct causes a salivary retention syndrome (also known as the mealtime syndrome [17], swelling of the concerned gland and eventual pain. Eventhough salivary retention is a feature it's not specific to sialolithiasis. Down the lane, the raised intraglandular pressure will lead to the formation of connective tissue and atrophy of acinar cells [11]. Incomplete obstruction will only cause a decrease of the salivary flow, thus explaining why some salivary stones can be symptomless.

Diagnosis

A congestive ostium of Wharton's duct compared to its counterpart is often found while examination. Bimanual palpation of the mouthfloor has to be performed in a posterior to anterior direction along the trajectory of the submandibular duct in order to search for any inflammation or induration. Also the salivary flow from the duct should be monitored.

To confirm a suspected diagnosis, several imaging tools are available. One third of the reported sialoliths are not radiopaque enough to detect in the conventional radiographs. But occlusal radiographs are one of the conventional diagnostic imaging modality [18]. Ultrasonography can be used to detect the presence of salivary stones and to assess the state of the glandand thus USG is a part of the first-line examinations. CBCT is another promising first line examination modality. Less irradiation and fewer artefacts and precision in locating the sialolith make it noteworthy [1].

Sialography determines the exact size and location of stones within the salivary gland ducts. However, if active infection is suspected, sialography is contraindicated due to the risk of exacerbating the extent of infection.

Sialendoscopy is indicated when the computed tomography does not show a calculus and when a minimally invasive technique should be favoured [19, 20, 21]. It allows a good visualization of the salivary duct system and its flushing system enhances the image quality and provide a therapeutic effect [22, 23].

MR sialography is another noninvasive technique with rapid acquisition of images that demonstrate the main duct and intraglandular branches up to second order branching in the salivary glands. MR sialography can visualize parotid ducts wall damage and abnormality in the gland parenchyma [24]. In addition, MR sialography is less bias when examine the duct system; and it allows detailed observation of the ducts system [25].

Management

Non-invasive conservative management of salivary stones consists of gland massage after each meal, daily intake of adequate water and administration of sialogogues.

Invasive management of sialolithiasis consists of lithotripsy, sialendoscopy and surgery. Extracorporeal shock wave lithotripsy (ESWL) is a minimally invasive approach that uses high energy shock waves generated outside the body to pulverize or crush the stones inside the body. In intracorporeal lithotripsy a lithotripsy probe is inserted into the salivary duct under endoscopic guidance, which introduces shock waves in the form of laser beams, pneumatic devices and electro-hydraulic or electrokinetic probes. The goal of both techniques is to reduce the size of the sialoliths making them more easily cleared from the salivary duct system spontaneously after sialogogue induced salivation, or during endoscopic procedures.

Surgically an incision can be made in the mouthfloor's mucosa following the longitudinal axis of the submandibular duct. A traction wire or a stay suture can also be placed behind the calculus in order to avoid its posterior migration during surgery. The incision should not exceed the first mandibular molar in order to avoid any damage to the lingual nerve. The stone can then be released from the canal using metzenbaum chisel or a curette. An antispasmodic can be given prior to surgery to increase the diameter of the ostium promoting canal drainage.

An anteriorly placed and palpable sialolith can be removed locally, whereas a posterior and palpable stone should be performed under general anesthesia [26, 27, 28, 29, 30]. Calculi smaller than 4 mm may be removed using a basket probe, while larger stones will require endocanal fragmentation (endoscopic laser) or external fragmentation (extracorporeal lithotripsy). Stones less than 4 mm diameter needs sialendoscopy regardless of their position under local or general anesthesia.

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