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MULTIPLE SIALOLITHS IN THE WHARTON'S DUCT: A RARE CASE REPORT AND REVIEW OF LITERATURE

Maxillofacial Surgery

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ABSTRACT

A condition of the salivary glands that affects more than 50% of people is known as sialolithiasis. Submandibular gland and its duct houses majority of salivary calculi measuring less than 10mm. It favours men and is frequently observed in adulthood (1). 70% to 80% of cases feature solitary, only about 5% of patients have three or more stones(2). The case which we are presenting here had five Sialoliths involving Wharton's duct which were removed by intraoral approach with no post-operative complications.

KEYWORDS

Calculus, multiple sialoliths, submandibular gland, Wharton's duct

INTRODUCTION:

A calculus within the salivary gland or its excretory system that causes an obstructive phenomena is known as sialolithiasis. Although uncommon, bilateral involvement is nevertheless possible and primarily affects the submandibular glands. They can be of variable sizes ranging from a few millimetres to centimetres. Giant sialolithiasis is the term used to describe calculi larger than 1.5cm[3]. Salivary calculi are the second most common salivary gland disorder after mumps. It affects more than 1% of the population with male predilection over 30 years of age. All salivary glands can be concerned. However, they mainly occur within the submandibular glands or ducts with a frequency of 80%. This could be explained by its different salivary composition[3]

CASE REPORT:

A 25 year old male patient reported to the Department of Oral and Maxillofacial Surgery at Meghna Institute of Dental Sciences, Nizamabad with chief complaint of swelling and pain in the floor of the mouth on left side since 4 months. Detailed history revealed that it started as a small swelling which gradually increased over next three months and attained present size. Patient experienced pain which is sudden in onset, dull, intermittent type aggravated before meals, as well as pus discharge from the same region.

Intraoral examination revealed a well defined round swelling of approximately 4-5mm in size in the floor of the mouth in the lower left first molar region and three swellings of approximately 2-3mm in size in the lower left lateral incisor ,canine and premolar region . Overlying mucosa was normal in color except in the distal most part of swelling where there was break in continuity exposing the underlying yellowish mass. It was hard in consistency and tender on palpation [Figure 1]. A provisional diagnosis of multiple sialoliths in the left submandibular duct was given.

Figure 1: Intra oral photograph showing sialolith in the floor of the mouth on left side causing sialo-oral fistula

Figure 2: Mandibular occlusal radiograph revealing multiple radio-opacities(5 in number) in the floor of the mouth on left side

Radiographic evaluation on mandibular occlusal radiograph which revealed a large well defined oval homogenous radiopacity in the floor of the mouth on left side [Figure 2] in relation to lower left second molar. It was approximately 4-5mm in size and three homogenous radioopacity of approximately 2-3mm in the anterior region and one homogenous radioopacity of approximately 2-3mm in the mandibular third molar region on left side.

PROCEDURE:

Under aseptic conditions after painting with 5% betadine solution and draping the patient, local anaesthesia with 2% lignocaine with 1:80,000 concentration adrenaline was administered. A suture was placed behind the last calculus in order to avoid its posterior migration during surgery. A traction wire can also be used for this purpose. Excision of the calculus is achieved by placing an incision of the mucosa in the floor of the mouth parallel to the longitudinal axis of the Wharton's duct (Fig.3) Care should be taken not to extend the incision beyond first mandibular molar in order to avoid any damage to the lingual nerve. One of the sialoliths is large of around 1cm in diameter and the other four sialoliths are smaller of around 3mm in diameter (Fig 4). It should be noted that the literature is not uniform as to whether or not a marsupialization of the duct has better long-term results than healing by secondary intention[3]. A catheter is inserted into the sub-mandibular duct and is secured with sutures, in order to avoid stricture(Fig 5)

Figure 3: Intra-operative photograph showing excision of sialolith on left side

Figure 4: Excised sialoliths which are five in number

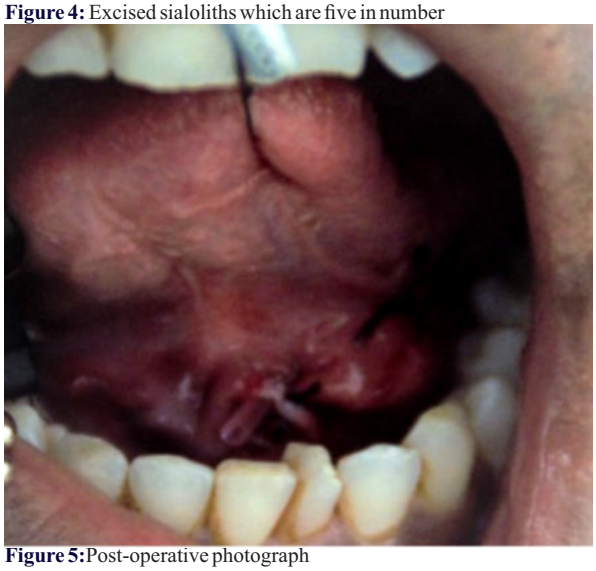

Figure 5: Post-operative photograph

DISCUSSION:

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 as 64%, 20 %, 16% in the Sub mandibular, Parotid ,Sublingual gland and duct respectively. Most of the sialoliths are of 5 mm in maximum diameter and all the stones over 10 mm should be reported as a sialolith of unusual size. Furthermore, they are classified as giant if the diameter exceeds 15 mm. One of the largest sialoliths of 72 mm size was reported by Rai and Burman[4]. Males are affected twice as much as females. While Seldin et al[5] and Lustmann et al[6] found the M:F ratio to be 1:1 in their studies.

Etiology

Though definite etiology is still ambiguous, sialoliths are thought to occur as a result of deposition of mineral salts around an initial nidus consisting of salivary mucin, bacterial colonies, exfoliated ductal epithelial cells, mucus plugs, foreign bodies or other cellular debris. Factors like stagnation of salivary flow due to flow against gravity, tortuous course of the duct, increased mucous content in saliva, dehydration, and change in salivary pH associated with oropharyngeal sepsis, impaired crystalloid solubility, high alkalinity, and increased calcium content, and physical trauma to salivary duct or gland, facial hair of the patient[8] may predispose to calculus formation[7].

Two stages of sialolith formation can be found in the literature: i) central core formation and ii) layered periphery formation. Firstly, mineral salts bound by certain organic substances precipitate to form the central core followed by deposition of organic and inorganic materials around the central core in layers.

Another theory contends that an unknown metabolic process can cause the precipitation of calcium and phosphate ions in saliva by raising salivary bicarbonate levels, which then change the solubility of calcium and phosphate[3]. A retrograde theory stated that any substance or bacteria of the oral cavity, that had migrated into the salivary ducts, can act as a nidus for further calcification[8]

Location:

Salivary calculi are commomly seen in the Wharton's duct.

Clinical features:

Depending on the location and the size of the stone, the presenting symptoms vary. Although most salivary stones are asymptomatic or cause minimal discomfort, while larger one may interfere with the flow of saliva and may cause pain and swelling. The symptoms include pain on sight of food or during eating and swelling of the involved gland caused by the accumulation of saliva due to blockage of lumen of the duct by a salivary calculus. Recurrent infections may occur due to the ascent of bacteria into the parenchyma of the gland[3]

Palpation:

The diagnosis of salivary calculi is based on the patient's history and clinical examination which are supplemented by radiographic findings. The patient may present with pain and swelling of the concerned gland at meal times and in response to other salivary stimuli, pus may be seen draining from the duct and signs of systemic infection. Extraorally, the submaxillary gland may be seen protruding beneath the angle of the mandible and the Parotid gland bulging just in front, of the ear. Intraorally, with Sub-lingual or Sub-mandibular gland involvement, the tongue and floor of the mouth on the affected side may be elevated and inflamed.

When the stone is not severely embedded in the substance of the gland, palpation of the stone is possible. The ideal way to complete this treatment is with simultaneous intraoral and extraoral manipulation (bimanual palpation). While one hand presses down on the floor of the mouth and the other feels the stone resisting this pressure. Sub-mandibular stones are frequently found when the floor of the mouth is palpated posterior to anterior. The glands are firm and tender on palpation[5]. A posterior to anterior bimanual palpation of the floor of the mouth reveals Sub-mandibular calculi in large number of cases.

Sialoliths are traditionally diagnosed by clinical features coupled with radiographs, though newer techniques are now available**.**

Investigations:

Radiograph:

When no stone can be seen or palpated, imaging studies can be a great help in diagnosing sialolithiasis.

Occlusal radiographs are extremely useful in showing radiopaque stones unless otherwise there are radiolucent stones. Submandibular gland calculi are mostly radiopaque (80% to 94.7%)[3]

According to Williams, standard extra-oral radiographs like **Orthopantamograph** are less diagnostic than intraoral radiographs or occlusal views. Stones in the superior gland or proximal Wharton's duct may get superimposed on the teeth or mandible, which can make visualization difficult in extra-oral radiographs. So, their identification will be difficult, especially when they are small[3].

Sialography

Sialography, which was previously used widely for detection of duct stricture, stenosis or sialoliths, is a method of injecting contrast material into Stenson's or Wharton's duct for visualization. Sialographic studies may reveal features of the involved salivary glands, having important diagnostic value. The anatomy of the duct can be displayed, revealing its form as narrow or large, the presence of

secondary branches leaving the main duct, and the presence of accessory glands or sialolithiasis, including their dimensions, number, and positions. Sialography is a very helpful tool in cases when signs of sialadenitis are related to a radiolucent or deeply impacted submandibular/ parotid stone. It appears as a filling defect

Sialendoscopy:
Introduction of sialendoscope has taken the diagnosis of these pathologies to the next level. Katz and Fritsch, used a flexible endoscope for the first time in the 1990s for evaluation of the salivary gland ducts. In addition to the radiolucent stones and those not shown in the imaging techniques, any possible duct stricture or other kinds of obstructions can also be found with this technique. In the case of a small sialolith and favorable conditions, a definitive sialolithotomy can be performed with sialendoscopy

Duct perforation is the most usual and common complication of the procedure which leads to fluid extravasation in the floor of the mouth. Temporary injury of the lingual nerve has been also reported in the literature[9]

Ultrasonography:
Katz et al. and various other authors have reported that ultrasonography is a very useful diagnostic tool for submandibular sialolithiasis with a sensitivity and specificity between 90% and 95% respectively[9]. However, a major drawback of this procedure is that smaller calculi (less than 2mm) remain undetected most of the times.

Newer techniques:
Computed tomography is very helpful in cases where other techniques provide ambiguous results, especially in parotid duct calculi. Usually, a CT scan is done both with or without contrast enhancement to differentiate between calculi and vascular structures[3]

Magnetic resonance imaging is another very helpful tool for the identification of smaller stones and to differentiate acute from chronic obstruction. Sialo-Magnetic Resonance Imaging is a non-invasive method of diagnosing sialoliths. It does not use any contrast vehicle and ionizing radiation overcoming, the major disadvantages of both computed tomography and contrast sialography. Acute inflammation is not a contradiction to this technique, which is a great advantage. The detailed morphology of the salivary ducts, up to second and third order branches can be seen with this technique, without the need for duct cannulation.[3]

Management:
Preservation of gland function in conjunction with low-level risk and discomfort for the patient should be the primary objective in the treatment of sialolithiasis. The treatment of choice varies according to the size, location and the number of stones[3]

Conservative management:
When stones are small , moist warm heat application with the administration of sialogogues and gland massage help in flushing the stone out of the duct. Small sialoliths can be removed through the duct orifice using bimanual palpation. The infection should be treated with antibiotics and these cases should be combined with simple sialolithotomy when required[3]

Sialendoscopy:
Sialendoscopy, which is a minimally invasive technique, was first introduced by Katz in 1991 and has since been developed by Marchael et al. and Nahlieli et al. Klein and Ardekian mentioned that sialoliths upto the range of 4-5mm of diameter are the ideal for sialendoscopic removal while it is a challenge for the surgeons to perform sialendoscopy of the sialoliths located deep within the hilum. There are speculations regarding the endoscopic approach since a general consensus is yet to arrive on either the maximum diameter of the stones that could be removed without fragmentation or whether impacted or hard stones could be managed effectively by endoscopy alone[10]. The general indication for the approach is the combination of sialendoscopy with extra-corporeal lithotripsy using Thullium LASER for the fragmentation of Lithiases between the range of 4 and 8mm.

Surgical techniques:
Upon failure of non-invasive techniques mainly due to the size of the stones and their location, the Sialoliths are treated by open surgical approaches. These include trans-oral duct incision, purely external approaches, or a combination of approaches. An Endoscopy assisted trans-oral removal technique for larger or impacted stones can also be of utility[3]

Although advances in sialoendoscopy and minimally invasive surgery have minimized the need for salivary gland excision, still there are some situations where excision is inevitable. This takes into account the inclusion of symptomatic or recurrent sialadenitis which is caused by multiple intraparenchymal stones or due to the presence of a very large stone, intraoperative complications of sialoendoscopy which requires gland removal, or inability in extracting the stone when minimally invasive procedures listed above are used and/or residual symptoms which are present despite the removal of stones such as recurrent inflammation. The excision of sub-mandibular gland for chronic sialadenitis is more frequent and is secondary to lithiasis.

The CO2 LASER enables bloodless surgery and a clear vision of the operation. Intervention can be carried out during acute stages without spreading of infection, and with minimal post-operative pain and edema.

A number of complications are associated with an open surgical approach. The primary concern is post-operative neurologic damage. A major cause of patient morbidity includes various other complications, including sialoceles, salivary fistula, facial scarring and Frey's Syndrome. There is a greater risk of facial palsy as in sialolith in parotid gland or in Stenon's duct, there are branches of the facial nerve that cross over the duct. There might be instances of non-closure of the duct because of an external incision, which might lead to salivary fistula.

Extracorporeal and Endoscopic Intracorporeal Shockwave Lithotripsy:
There have been alternative techniques that have been found out which includes the usage of Extracorporeal Shock Wave Lithotripsy and the most recent being using Endoscopic Intracorporeal Shock Wave Lithotripsy, where direct shockwaves are delivered to the surface of the stone lodgement inside the duct without damaging adjacent tissue (piezoelectric principle)[3]. A better option than surgical removal of the affected gland is Salivary Lithotripsy which is more useful and beneficial, as this prevents the risks of General Anaesthesia, damage of the facial nerve, surgical scar, Frey's syndrome, and minimum discomfort to the patient, but also with the preservation of the gland. The limitation for this technique is it requires additional equipment and it is more expensive.

CONCLUSION:
Sialoliths should be always considered in submandibular and facial pain particularly when it is related to mealtime. A careful history and correct imaging techniques are required to confirm the clinical diagnosis and to define the precise location of the calcification. Though advanced and more efficient methods are available occlusal radiographs are still useful in diagnosing sialoliths. Small stones can be treated conservatively but for larger stones, sialolithotomy combined with antibiotics is the treatment of choice if newer technologies are not available. According to literature, the success rate of the intraoral submandibular duct approach is between 85% and 100%[11].This submandibular duct technique is simple, adaptable to different anatomic situations and, can be performed under local anesthesia. Immediate and permanent post-surgery complications, such as lingual nerve injury, are exceptional.

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