CASE REPORT

Unveiling What Lies Beneath! Intraglandular Sialolith: A Clinical Dilemma

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ABSTRACT

Sialolithiasis is the most common disease affecting the salivary glands and accounts for 30% of all salivary gland disorders, affecting 0.01 to 1% of the population. Submandibular gland being the most common site, with the incidence of intraglandular sialolith much rarer as compared to the intraductal ones and they may go undetected for years. Their findings are usually incidental, unless an acute phase is encountered. Here, we present a case report of 26-year-old male patient in whom the intraglandular (submandibular) sialolith was encountered incidentally on the radiographs.

Keywords: Sialolithiasis, Intraglandular sialolith, Submandibular gland.

INTRODUCTION

The term sialolithiasis derived from the Greek words, sialon meaning saliva and lithos meaning stone. It is the most common disease of salivary gland and a major cause of salivary gland dysfunctions. It often gives rise to obstruction of salivary gland duct with a swelling of gland which usually appears at meal times.1 The salivary stasis produced due to obstruction predisposes to secondary retrograde infection.2 Here, we report a case of intraglandular sialolith in submandibular gland in a 26-year-old male patient which was incidentally discovered on an orthopantomograph.

CASE REPORT

A 26-year-old male patient reported to the department of oral medicine and radiology with a chief complaint of inability to close the mouth following a road traffic accident 10 days back. Patient gave a history of suffering from tuberculosis one year back for which he was treated. On extraoral examination, a diffuse swelling was present involving lower one-third of left side of face extending anteroposteriorly 1 cm distal to the corner of mouth to angle of the mandible and superoinferiorly from ala tragus line to the lower border of mandible. The swelling was soft in consistency, tender on palpation and a step deformity was present in relation to the right angle of the mandible and left parasymphyseal region. The left and right submandibular lymph nodes were palpable, enlarged, mobile, tender and soft in consistency (Fig. 1). Reduced mouth opening and occlusal derangement were noticed and mobility of the segment in relation to 33 was present. Based on history and clinical findings, fracture of left parasymphyseal and right angle of the mandible were suspected.

An orthopantomogram was taken which revealed a fracture line in the left parasymphyseal and right angle of the mandible. It also revealed a well-defined oval-shaped radiopacity with a central radiodense core surrounded by alternating radiodense and radiolucent layers giving a concentric ring appearance in the region of left angle of mandible measuring about 3 × 2 cm in dimension. Adjacent to this, another radiopacity with a radiolucent rim was seen, measuring 1 cm in diameter (Fig. 2).

Fig. 1: Facial profile of patient showing swelling of left side of face
Due to the incidental finding of this unusual radiopacity, the patient was further subjected for other investigations. PA skull view showed well-defined radiopacity overlying the left mandibular molar region (Fig. 3).

Due to previous history of tuberculosis, patient was advised further investigations. Routine hemogram revealed increased ESR and Mantoux test were positive. FNAC for the left submandibular lymph node was performed to rule out tubercular lymphadenitis which came negative. Thus, based on the medical history, various radiographic and hematological investigation performed, we came to a diagnosis of a calcified submandibular glands.

For further confirmation of the diagnosis, an ultrasound of left submandibular gland was performed, which revealed the gland to be normal in shape and size with presence of two calculi within the gland of size 25 mm and 5 mm approximately (Fig. 4). Three-dimensional and coronal computed tomography (CT) scans showed a large multilayered calcified lesion in left submandibular region measuring $28 \times 16$ cm in largest dimension and two small satellites. The central layer appears to be more dense and calcified with laminations suggestive of a stone in left submandibular gland. Multiple undisplaced fractures were also seen on left and right side (Fig. 5).

Therefore, based on the ultrasound and CT findings, a final diagnosis of intraglandular sialolith of left submandibular salivary gland was established.

**DISCUSSION**

Sialolithiasis is the most common disease of salivary glands. It is estimated that it affects 12 in 1000 of adult population. It accounts for more than 50% of diseases of salivary gland and thus is most common cause of acute and chronic infections. The size of salivary calculi varies from 1 mm to a few cm in diameter. Most of the salivary calculi are less than 10 mm in size; whereas, only 7.6% are larger than 15 mm.

Sialolithiasis is more common in submandibular gland (80%) as compared to parotid (19%) followed by sublingual (1%) glands. This may be attributed to the increase alkalinity of its saliva, greater concentration of calcium and phosphate and high mucus content. In addition, the submandibular duct is long and transverse upward and forward from the gland to the floor of mouth thus, forming an acute angle on its path which facilitates stasis of saliva. Also, the main portion of duct is wider than the orifice and there is decrease in wall thickness.
It is believed that salivary calculi develop as a result of deposition of mineral salts around the nidus of bacteria, mucus or desquamated cells. The salivary stagnation, increased alkalinity of saliva, infection or inflammation of salivary duct or gland and physical trauma to salivary gland and duct may predispose to formation of sialolith. Stasis, which is associated with dehydration, debilitation, ineffective treatment after surgery, allergic status and neural origin starts the accumulation of ductal debris which in turn permits precipitation of mucoid element and salts in order to form the organic matrix, in an environment of decreased pH as a result of ascending infection. As this chain reaction continues, the mucoid material gains laminar structure by aggregation of carbohydrates, amino acids, desquamated epithelial cells, bacteria and salts upon nidus. Thus, the internal structure of sialolith is laminated.

The composition of salivary stones is predominantly calcium phosphates and calcium carbonates in form of hydroxyapatite with small amounts of magnesium, potassium and phosphate. The organic matrix is composed of various carbohydrates and amino acids. Parotid stones tend to be less dense and less calcium-rich most likely this is related to the serous nature of the secretions from parotid parenchyma, in contrast to the more mucinous secretions from the submandibular gland.

The clinical presentation of sialolith varies from a painful swelling (59%) to a painless swelling (29%) or just pain (12%). Patient may present with a history of recurrent pain and swelling during meal time. The patients may also have repeated infection and abscess formation. Long standing obstruction by sialolith may cause severe damage to the acini of the gland, resulting in permanent decrease or even absence of salivary secretion. This reduced or absent salivary secretion may give rise to recurrent infection.

A careful history and examination are important in the diagnosis of sialolithiasis. Bimanual palpation of the floor of the mouth helps in detecting submandibular calculi in most of the cases and if the gland is found to be uniformly firm and hard, it suggests a hypofunctional or nonfunctional gland.

Imaging studies are very useful for diagnosing sialolithiasis. Occlusal radiographs are useful in showing radiopaque stones. It is very uncommon for patients to have a combination of radiopaque and radiolucent stones. The submandibular gland calculi are radiopaque anywhere between 80 and 94.7%. Extraoral radiographs, such as orthopantomograms and posteroanterior views are also helpful. Sialography is considered as gold standard for diagnosing sialolith and is useful in patients showing signs of sialadenitis related to radiolucent stones or deep submandibular stones. Sialograms are reported to be 100% effective in detecting ductal and intraglandular calculi.

Ultrasounds are an inexpensive, noninvasive and widely available method for detecting salivary calculi. On ultrasound, the stones appear as hyperechoic foci casting posterior acoustic shadowing. Those smaller than 2 mm may not cast acoustic shadows. Ultrasound can also assess the complications of calculi, e.g. sialadenitis, sialocele, abscess and gland atrophy.

CT scan is an expensive, albeit highly diagnostic modality for detection of sialoliths. Most stones contain enough calcium to be visible on CT scans.

Sialo-MRI is a diagnostic, noninvasive method recently introduced with promising results, in the evaluation of salivary gland disease. This technique produces sialographic images but without contrast medium injection and without the disadvantage of ionizing radiation (CT and contrast sialography). 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. The major disadvantages of the sialo-MRI examination are the relatively long time required for the scanning examination (45 mins), the high costs, lack of compliance in the claustrophobic patient and the presence of artifacts in patients with dental bridges or a metallic prosthesis.

Scintigraphy uses technetium-99m pertechnetate which is selectively concentrated and secreted in salivary gland. It also helps in determining the function of gland. But in obstructive conditions, there may be marked decrease in uptake of pertechnetate that does not correlate well with functional recovery after stone removal. Sialoendoscopy has become one of the important tools in assessment of salivary gland.

Patients presenting with sialolithiasis may benefit from a trial of conservative management, especially if the stone is small. 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. With gland swelling and sialolithiasis, infection should be assumed and a penicillinase resistant antistaphylococcal antibiotic prescribed. Most stones will respond to such a regimen, combined with simple sialolithotomy when required. If the stone is sufficiently forward, it can be milked and manipulated through the duct orifice. This can be done with the aid of lacrimal probes and dilators to open the duct. Deeper submandibular stones are removed through sialoadenectomy. Endoscopy is a minimally invasive technique for removal of calculi from salivary glands as well as an excellent diagnostic procedure, as miniaturised endoscopes conforming to the physiological widths of the ducts are used to directly view and then deliver shockwaves to the stones.

Shockwave lithotripsy using piezoelectric lithotripter has also been advocated as the primary treatment for submandibular gland sialolith, but it requires advanced armamentarium. Laser sialolithotomy has also been used as alternative method for salivary stone management. In case of presence of intraglandular sialolith and destruction of gland, the removal of entire gland is recommended.

CONCLUSION

Calcifications found in routine orthopantomograph may turn out to be an interesting diagnosis. Although most of the radiopacities superimposing angle of mandible are usually
considered lymph node calcifications but intraglandular submandibular salivary gland sialolithiasis, though rare, must be considered in differential diagnosis as in most cases it is associated with minimum or no symptoms.

REFERENCES