CASE REPORT

INTRAGLANDULAR SIALOLITH OF SUBMANDIBULAR SALIVARY GLAND: A CASE REPORT

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ABSTRACT: Sialoliths are calcified structures that develop within the ductal system and is the most common disease affecting the salivary glands which lead to recurrent painful swelling of the involved gland which often increases while eating. Submandibular salivary gland is the commonest site. Their findings are usually incidental until an acute phase is encountered. Here we present a case report of a 56 year old man who presented with pain and swelling in the lower jaw.

KEYWORDS: Submandibular gland, Sialolithiasis, Sialadenitis.

INTRODUCTION: The term 'sialolith' is derived from the Greek words, sialon meaning saliva and lithos meaning stone. Salivary gland lithiasis (sialolithiasis) is the most common disease of major salivary glands after mumps and accounts for approximately 30% of all salivary disorders with a higher incidence in males aged between 30 and 60 years. The most common localization is submandibular gland where 92% of calculi are found, the duct being more frequently affected than the parenchyma.

CASE REPORT: A 56 year old male patient came to the department of Oral Medicine and Radiology with a chief complaint of pain and swelling in the right lower jaw region since 10 days. Swelling increased while eating. Patient gave a history of cardiac disorder for which he is under medication since 10 yrs. On extra oral examination, a swelling of size 4×5 centimeters cm was seen extending anteroposteriorly from the right lower border of mandible till the right angle of mandible. The swelling was hard in consistency, surface was smooth and tender on palpation. Right and left submandibular lymph nodes were palpable. Right submandibular lymph nodes were enlarged, soft, mobile and tender. (Image 1).

On examination of submandibular, salivary glands edema and inflammation of soft tissue surrounding the duct was noted. Swelling increased in size when he was given drops of lemon. On bimanual palpation, soft tissue surrounding duct was tender and enlarged. Based on history and clinical findings, sialadenitis of the right submandibular gland was suspected.

A lateral oblique radiograph was taken which showed a homogenous oval shaped radio opacity of size 1.2×2 centimeters with well-defined border present 0.5 centimeters anterior to the right angle of mandible and inferior to the right inferior alveolar canal. (Image 2).

For further confirmation of the diagnosis, an ultrasound of the right submandibular gland was performed which revealed enlarged right submandibular gland measuring approximately 3.8× 2.5 centimeters with inhomogenous echo texture and mild increased vascularity and a calculus measuring 13 millimeters within the gland. Enlarged level 3 lymph nodes measuring 2.4 centimeters were also noted. (Image 3).

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Therefore based on the radio graphical and ultrasound findings, a final diagnosis of intraglandular sialolith of right submandibular gland was established.

DISCUSSION: Sialolithiasis is the most common disease of salivary glands and is the most common cause of acute and chronic infections. The size of salivary calculus varies from 1mm to a few cm in diameter. They are usually unilateral in occurrence and round to oblong, have an irregular (majority) or smooth surface, vary in size from a small grain to the size of a peach pit, and are usually yellow.¹

Most of the salivary calculi are less than 10mm in size; whereas, only 7.6% are larger than 15 mm. Careful history and examination are important in the diagnosis of sialolithiasis. Sialolithiasis typically causes pain and swelling of the involved salivary gland by obstructing the food related surge of salivary secretion. Pain and swelling of the concerned gland at meal times and in response to other salivary stimuli are especially important. Complete obstruction causes constant pain and swelling, pus may be seen draining from the duct and signs of systemic infection may be present.² Bimanual palpation of the floor of the mouth, in a posterior to anterior direction, reveals a palpable stone in a large number of cases of submandibular calculi formation. Bimanual palpation of the gland itself can be useful, as a uniformly firm and hard gland suggests a hypo-functional or non-functional gland.³

Stone formation is not associated with systemic abnormalities of calcium metabolism.³ Electrolytes and parathyroid hormone studies in patients with sialolithiasis have not shown abnormalities.⁴ Gout is the only systemic illness known to predispose to salivary stone formation,⁴ although in gout the stones are made predominantly of uric acid.³

The exact etiology and pathogenesis of salivary calculi is largely unknown. Genesis of calculi lies in the relative stagnation of calcium rich saliva. Calculi may cause stasis of saliva, leading to bacterial ascent into the parenchyma of the gland³ and therefore infection, pain and swelling of the gland. 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.^{5,6} Calculi generally consist of a mixture of different calcium phosphates (mainly hydroxy-apatite and carbonate-apatite) together with an organic matrix.¹ Long term obstruction in the absence of infection can lead to atrophy of the gland with resultant lack of secretory function and ultimately fibrosis.³

The etiologic factors involved in the sialolith formation can be classified into two different groups: on one hand, saliva retention due to morpho-anatomic factors (salivary duct stenosis, salivary duct diverticuli, etc.) and on the other, saliva composition factors (high super saturation, crystallization inhibitor deficit, etc.)⁷

Traditional theories suggest that the formation occurs in two phases: a central core and a layered periphery.⁸ The central core is formed by the precipitation of salts, which are bound by certain organic substances. The second phase consists of the layered deposition of organic and non-organic material.⁹ Submandibular stones are thought to form around a nidus of mucous² whereas parotid stones are thought to form most often around a nidus of inflammatory cells or a foreign body.^{2,10,11}

Another theory has proposed that an unknown metabolic phenomenon can increase the saliva bicarbonate content, which alters calcium phosphate solubility and leads to precipitation of calcium and phosphate ions.^{3,12}

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A retrograde theory for sialolithiasis has also been proposed.⁷ Aliments, substances or bacteria within the oral cavity might migrate into the salivary ducts and become the nidus for further calcification.

Submandibular sialolithiasis is more common as its saliva is (i) more alkaline, (ii) has an increased concentration of calcium and phosphate, and (iii) has a higher mucous content than saliva of the parotid and sublingual glands. In addition, the submandibular duct is longer and the gland has an antigravity flow.^{13,14}

Ultrasonographic (US) examination is considered as a first choice, simple and non-invasive modality to evaluate sialoliths especially during acute infection. Submandibular and parotid calculi diagnosis can be readily performed with US, for almost all intra-parenchymal stones. US examination is considered less accurate in comparison to computerized tomography (CT) in distinguishing multiple stones. It has also been reported that sialoliths smaller than 3mm may not be detected during US examination, as they will not produce acoustic shadows.¹⁵

Digital sialography and subtraction sialography have increased the sensitivity and specificity of conventional sialography techniques that were considered gold standard.¹⁵

CT sialography has been used to delineate the ductal system of submandibular gland; this technique demonstrates the soft tissue of gland and ductal system with 3D reconstruction that avoids superimposition of anatomic structures.¹⁵

Magnetic resonance sialography (MRS) is a new technique that is considered an excellent radiological modality for the diagnosis of sialolithiasis. MRS may be indicated in cases of acute infection where other sialography are contraindicated since MRS does not require cannulation of the duct.¹⁵

Scintigraphy uses technetium-99m pertechnetate which helps in determining function of gland.³

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.¹⁶

The algorithm for the treatment of sialolithiasis depends upon the size and location of sialolith. Patients presenting with sialolithiasis may benefit from a trial of conservative management, especially if the stone is small.³ 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 anti-staphylococcal antibiotic prescribed. Most stones will respond to such a regimen, combined with simple sialolithotomy when required.^{2,10}

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. Once open, the stone can be identified, milked forward, grasped and removed. The gland is then milked to remove any other debris in the more posterior portion of the duct.³ If the gland has been damaged by recurrent infection and fibrosis, or calculi have formed within the gland, it may require removal.

Alternative methods of treatment have emerged such as the use of extracorporeal shock wave lithotripsy (ESWL) and more recently the use of endoscopic intracorporeal shockwave lithotripsy (EISWL), in which shockwaves are delivered directly to the surface of the stone lodged within the duct without damaging adjacent tissue (piezoelectric principle).¹⁷ Both extra and intracorporeal lithotripsy are gaining increasing importance in the treatment of salivary stone disease.¹⁸

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CONCLUSION: Even with the advent of various advanced diagnostic and treatment modalities in the management of sialoliths, the conventional techniques retain their popularity to date.

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IMAGE 1

IMAGE 2

IMAGE 3

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