Multiple sialolithiasis of submandibular gland: a case report

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Abstract

Objective: Sialolithiasis is a common disease of the salivary glands. Sialolithiasis occurs mainly in the submandibular gland (80–90%) and to a lesser extent in the parotid gland (5–20%). Sialoliths may be single or multiple. Multiple sialolith in the submandibular gland are rare. Seventy to 80% of cases feature solitary stones; only about 5% of patients have three or more stones. Patients commonly experience pain and/or swelling.

Methods: A 51-year-old male came to Department of Oral and Maxillofacial Surgery with a swelling and pain at the right submandibular. The panoramic radiograph showed a radiopacity mass with well-defined in right submandibular. The ultrasonographic interpretation showed hypochoic inhomogeneous mass with multiple

calcification. The submandibular gland removal was performed by extraoral approach.

Result: Multiple sialolith in the submandibular gland are rare. A surgical approach for removal of sialolith in the submandibular gland could be intraoral or extraoral. In this case report, we described five stones in the submandibular gland, which were removed by extraoral approach.

Conclusion: There are various methods available for the management of salivary stones. In this case report, removal of the submandibular gland with the stone should still be preferred as the gold standard of treatment and could avoid recurrence of this medical condition.

Keywords: Sialolithiasis, Multiple, Submandibular gland

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Introduction

Sialolithiasis is a most common salivary gland disease. Submandibular gland disease are most prevalent in mid-age with incidence of 12 in 1000 of adult population. This disease affects men twice more than women.^{1,2} The sialolithiasis generally occurs in the submandibular glands with prevalence of more than 80%, about 6% in the parotid gland and 2% in the sublingual gland or minor salivary glands. Sialolith can be either single or multiple stones. The prevalence of multiple sialolith in submandibular gland are rare.² Approximately 70–80% of cases are solitary stone, only about 5% of cases that have three or more stones in the salivary glands.¹ Salivary gland stones are generally unilateral and does not cause dry mouth. Clinically, salivary stones are round or oval, rough or smooth and yellowish.²

Sialolith stone comprises of less amount of calcium phosphate compare to carbonate in the form of hydroxyapatite, which is also lesser than magnesium, potassium and ammonia.¹⁻³ The composition of submandibular glands stone consists of 82% inorganic and 18% organic materials, and the composition of parotid gland stones consist of 49% inorganic and 51% organic materials. The organic material is composed of a various carbohydrates and amino acids. But bacteria elements were not found in the core of sialolith.²

The pathogenesis and etiology of salivary stones is poorly understood.^{2,4} Other than the composition of the saliva, some anatomical factors of submandibular glands and ducts are important in the pathogenesis of stone formation. The submandibular salivary alkaline pH can also support the formation of salivary stones.⁴

Case Report

In October 10th, 2014, a 51 years old male patient came to Oral and Maxillofacial Surgery polyclinic with chief complaint of lump and pain on his right lower jaw. The lump had occurred since three years before, first occurred with a size of table tennis ball, the size was often shrinking. The complaint was accompanied with pain during meal time or while chewing. There was history of toothache a month prior to hospital admission. There was no history of weight loss. History of other lump in other part of the body was denied. History of the same complaint in family was also denied. To treat his condition, the patient came to Rajawali hospital. Where he was given analgesic and antibiotic. Due to no improvement, the patient was referred to Hasan Sadikin Hospital.

In general physical examination, there was no abnormalities found. Respiratory rate was

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Figure 1 Clinical photograph of the patient, A. Right side view, B. Frontal view, C. Left side view

Figure 2 Intra oral photograph

A

B

Figure 3 A. Panoramic Radiograph, B. Ultrasonography

18 times/minutes, blood pressure check was 130/90 mmHg and pulse was 82 times/minutes.

Alocal extra oral examination presented a symmetrical face [figure 1B, 1C](#), with lump in right lower jaw, the color of the skin was similar to surrounding tissue [figure 1A](#). Through palpation on extra oral, the lump had a hard consistency, non-febrile temperature, uneven surface, fixated, and no tenderness, with 4x3x3 cm in size [figure 1A](#).

Intraoral examination found gangrene radices of teeth 46, 38 and 14. No visible abnormalities were found in buccal mucosa, gingiva, tongue and floor of the mouth [figure 2](#). There was no enlargement of lymph nodes found on submandibular glands and neck examination.

In laboratory tests, hematology and clinical chemistry was within normal limits. In radiology, the panoramic view showed a radiopaque mass with firm boundaries in the right submandibular region [figure 3A](#). On ultrasonography (USG) of the neck, there was hypoechoic and inhomogeneous mass accompanied by multiple calcifications with 4.0x3.46x3.33 cm size in the medial region of the parotid gland [figure 3B](#).

On October 23rd, 2014, surgical treatment was conducted to remove the right submandibular gland. Incision was done on the right submandibular region and continued with removal of right submandibular gland and retrieval of 5 sialolith stones. The flap closure was done by suturing the muscle and the skin layer-by-layer and then followed with drain installation and cleaning the surgery area. A press bandage was applied on the post-sutured site [figure 4](#).

Patient was discharged on the second day after surgery [figure 5A](#) then he was scheduled to control at the polyclinic of oral and maxillofacial surgery on November 7th 2014 [figure 5B](#) bringing the result of anatomic pathology tissue examination. Microscopically tissue sample was found to be coated. The subepithelium showed stromal fibro-collagen connective tissue, powdered massive inflammatory cells: lymphocytes, eosinophil, and plasma cells. Salivary gland presence among the cells with tubular shape coated with thorax

A

B

C

D

Figure 4 A. Incision site preparation, B. Removal of right submandibular gland and retrieval of sialolith stones, C. Flap closure with suturing and drain application, D. The excised submandibular gland and 5 sialolith stones

Figure 5 A. Post-operative of the second day, B. Control after 2 weeks

epithelium, the cells' nucleus were within normal limits. The ducts of salivary gland were dilated. A subcapsular view presence hyperplastic lymphoid follicles with dilating blood vessels accompanied with bleeding area. Also present are mature fat cells, with nucleus on the edge of the cells was within normal limit. There was no sign of malignancy. So histologically it was to be concluded as chronic nonspecific sialadenitis sialolithiasis at submandibular region.

Discussion

Sialolithiasis is a common salivary gland disease.^{1,2,5} The sialolithiasis generally occurs in the submandibular glands with prevalence of more than 80%, and 6% in the parotid gland and 2% in the sublingual gland or minor salivary glands.²

Clinical manifestations of submandibular stones will appear more clearly at mealtime, during the time of maximum saliva production, causing a sudden swelling and is usually very painful.^{2,5} A clinician should record the symptoms like pain, lumps, decreased salivary flow, and malaise.⁶⁻⁸ Swelling and pain that recurs in the gland can be associated to food or when seeing foods. The swelling reduced gradually, but will be repeated every time when the salivary flow is stimulated.^{1,2,5,6} This process may continue until the complete obstruction, infection, or a combination of both. Obstruction, with or without infection, may lead to atrophy of the involved glandular secretion cells.⁸ Glands infection manifests with swelling in the floor of the mouth, erythema and associated cervical lymphadenopathy.⁵ The period and duration of the swelling are very helpful in diagnosis. Clinical examination and palpation of the gland can help determine the lump in the gland, fixation, ulceration, or local nerve involvement and the intraoral examination can assess the decrease or increase of the salivary flow.^{1,6}

Radiographs often show radiopaque stones. To examine the submandibular gland, the panoramic

or occlusal photograph can be useful to see the gland ducts. Dental panoramic photograph show both areas of submandibular glands and parotid glands, but the definitive diagnosis usually requires more specific examination.⁶

Ultrasonography is a simple method that can indicate a sialolith with high accuracy. But sialolith that are smaller than 3 mm cannot be detected with ultrasound.¹

There are several techniques used for sialolith stone removal, which depends on the duration of symptoms, number of recurrent episodes, the size of the stone, and perhaps most importantly the location of the stone.^{3,5,9} Small sialolith, located near the peripheral ducts can be removed by manipulation (called flushing gland). Large sialolith are removed by surgery. Sometimes, the stone that is not affected can be extracted through the duct intubation using a soft plastic catheter and the application of suction to the tube. Multiple stones or stones in the salivary gland requires the removal of the gland itself.³

Almost half of the submandibular gland stones is located in the distal third of the submandibular duct and a simple surgery through an incision can be done at the floor of the mouth, which is simple and usually not associated with complications. If the salivary stone is located anterior enough, the stone can be massaged and manipulated through the duct hole with probe lacrimal guidance and a dilator to open the duct. Once open, the stone can be identified and removed. The glands are then massaged to remove other debris in the posterior part of the duct. The opening of the duct is required to remove the stone. It involves a transoral approach where the incision is made directly into the stone location. Through this approach, a more posterior stone/stones can be removed by cutting directly on the longitudinal axis of the duct. The wound closure was not done for the purpose of drainage. If the gland has been damaged by recurrent infection and fibrosis or if stone has been formed in the salivary gland, then it requires the removal of the salivary gland itself.^{2,6} Complications that may occur are scar tissue, facial nerve paralysis, or paresthesia of the lingual.⁶

Conclusion

Sialolithiasis is a common salivary gland disease. However, the prevalence of multiple sialolith in the submandibular gland is rare. Clinical manifestations of submandibular stones can be clearly noticed at the meal time, when the saliva production increases, which causes a sudden swelling and is usually very painful. The swelling will reduce gradually, but it will be repeated every time when the salivary flow is stimulated. There are several

methods of management that are used in the removal of the salivary gland stones. Handling of the submandibular gland stones depends on the duration of symptoms, number of recurrent episodes, stone size and location of the stone. In this case, the removal of the submandibular gland is still the best option in the management of multiple sialolith and can avoid possible recurrences.

Conflict of Interest

The authors report no conflict of interest.

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