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Case Report

Intraoral Surgical Management of the Multiple Giant Submandibular Sialolithiasis

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ABSTRACT

Sialolithiasis is considered one of the most common diseases of the salivary gland, mainly affects submandibular glands. In the current case, a 57-year-old male complaining of a tender solid mass in the right sublingual area was cadidate to undergo surgical treatment. The patient suffered swelling and pain for 11 months. Imaging revealed multiple unilateral salivary stones in the submandibular glands. The sialoliths were removed through intraoral approach under local anesthesia. Examination revealed four sialoliths, one of them sized larger than 37 mm in length. Beside those giant sialolithes, what makes this case is unique is the patient's endocrine disorder, the secondary hyperparathyroidism. So it would be plausible to consider checking possible endocrine and electrolyte imbalance in such cases alongside correct selection of surgical treatment plane.

INTRODUCTION

Accounting for 30% of the salivary disorders, sialolithiasis is considered one of the most common diseases of the salivary gland (1). Pain and swelling are most common symptoms associated with sialolithiasis, also signs like restricted mouth opening, reduced salivary flow, and purulent discharge can be seen. (2-4). 80% of submandibular stones are radiopaque; hence sometimes techniques like sialography, computed tomography scan and ultrasound may be neccessary to locate them (5). Commonly Sialoliths are between 5 and 10 mm in size and giant sialoliths measuring more than 35 mm are rare, however case of a sialolith in the submandibular duct up to 72 mm in length published in the literature (6).

In this case report, radiological findings, the clinical, paraclinical and surgical treatment of an large sialolith located in submandibular gland duct is presented. This case described here is of interest because the large sized multiple salivary stones being concurrent with endocrine disorder hyperparathyroidism.

METHODS

In April 2016, an 57-year-old man was admitted to the Tabriz dentistry school, Department of Oral and Maxillofacial Surgery complaining of persistent purulent discharge and and a tender solid mass in the right submandibular and sublingual area. The patient suffered swelling and pain for 11 months. The patient had no record of previous episodes of sialolithiasis. Computed tomography (CT) scan and orthopantomograph images revealed multiple unilateral salivary stones in the submandibular glands (Figure 1-3). Three stones was located in near the glandular hilum, and one was located in the distal part of Wharton's duct. The patient had no history of smoking, alcohol consumption or drug abuse.

An informed consent was obtained, the sialoliths were removed through intraoral approach under local anesthesia. An upward, and medial pressure was applied to the approximate site of sialolithes submandibulary so that the sialolith moved close to the incision. The incision was performed along the warthon duct over the sialolithes and the dissection was

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carefully preformed to to expose them from the floor of the mouth (Figure 4).

Examination revealed four hard, yellow, elongated stone sized 37 mm long and 14 mm wide, 28 mm long and 13 mm wide, 9 mm long and 8 mm wide and finally 6 mm long and 5 mm wide (Figure 5). Due to sufficient drainage no catheter was required.

An antibiotics (cloxacillin, 2000 mg/day), and a nonsteroidal anti-inflammatory drug (NSAIDs) plus acetaminophen (diclofenac sodium, 150 mg/day plus acetaminophen 2000 mg/day) were prescribed postoperatively for 5 days. Follow up reassessment took place on after one week and one month postoperatively (Figure 6).

Clinical examinations showed normal function of The submandibular gland. Beside those giant sialolithes, what makes this case is unique is the patient's endocrine disorder. Patient systemic evaluation reveal that patient suffers from secondary hyperparathyroidism (Table 1). Inspite of Hypocalcemia, all hematological kidney function tests were normal.

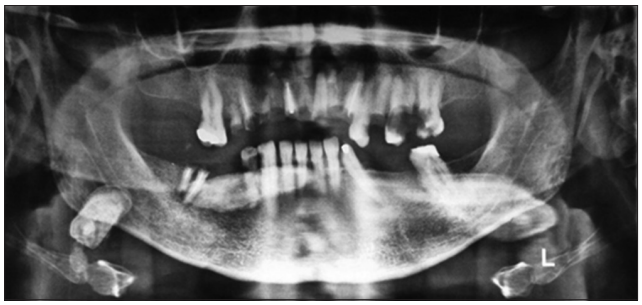

Figure 1. Orthopantomograph images revealed multiple unilateral salivary stones in the submandibular glands

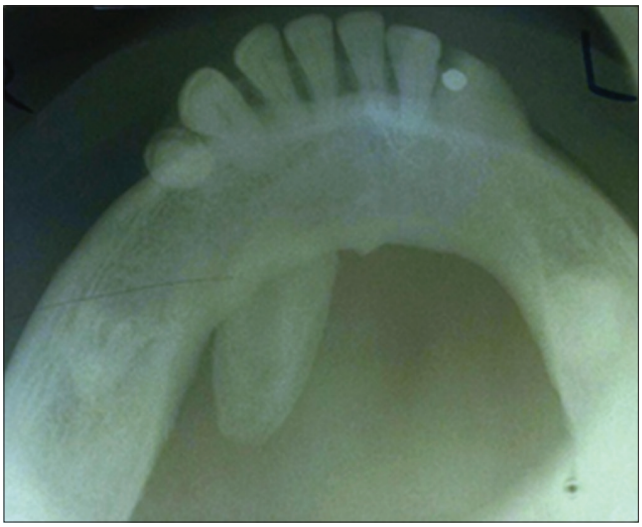

Figure 2. Occlusal view of most proximally positioned sialoliths

DISCUSSION

Sialolithiasis is one of most common occurring disease of the salivary glands, obstructing salivary secretion by a calculus. This can lead to pain and inflammation and, in some cases, infection of the affected gland. The most common symptom is swelling. Pain, fever and pus secretion are other frequent symptoms (7). As the patient grows older the rate of medication consumption increases causing reduced secretory activity, glycoprotein synthesis impairment, electrolyte concentrations alterations, and salivary glands cell membranes structural deterioration are seen(8).The size of sialolithes varies from small particles, to large concrement

Table 1. Hematologic laboratory test results showed high blood level of PTH and hypercalcemia

Test	Results	Units	Reference range
W.B.C	6200	/cumm	4000-10000
R.B.C	4.31	Mil/L	4-5.5
Hemoglobin	13.7	g/dl	12-16
Hematocrit	39.9	%	40-54
Blood urea	30	Mg/dl	15-50
Creatinine	1	Mg/dl	0.3-1.1
Sodium	137.4	mEq/l	135-148
Potassium	4.05	mEq/l	3.5-5.5
Calcium	8.4	Mg/dl	8.5-11
Phosphore	3.9	Mg/dl	2.5-5
P.T.H	132.8	Pg/ml	15-68.3

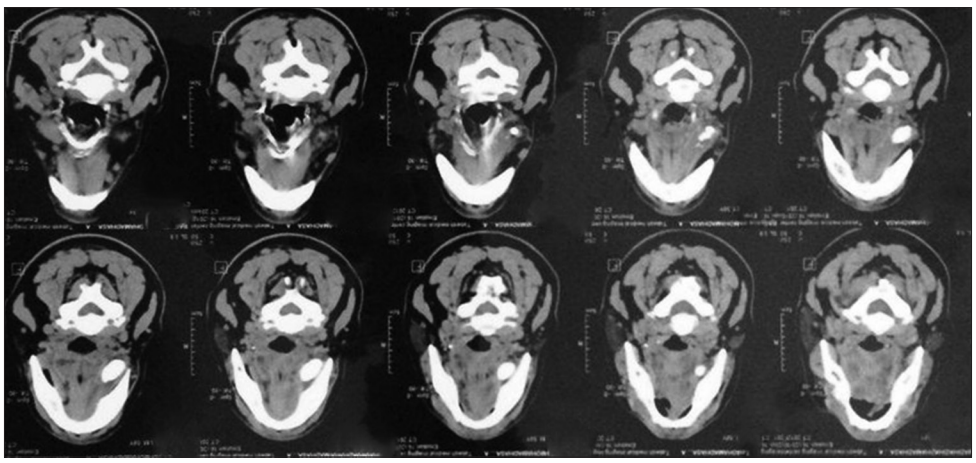

Figure 3. Sagittal view of spiral CT of sialolithiasis

Figure 4. Transoral approach to remove sialolithiasis

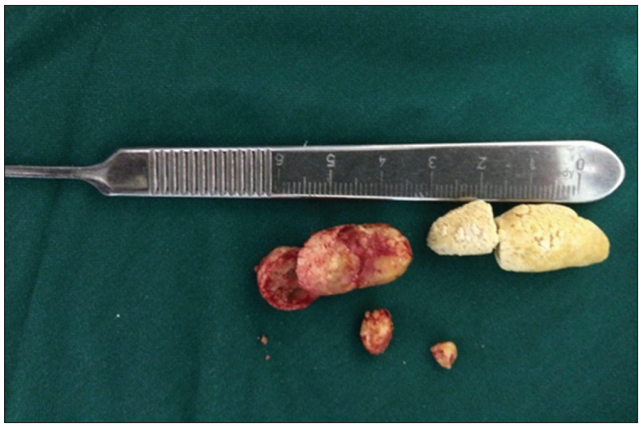

Figure 5. Surgically removed sialoliths

Figure 6. One week later, view of surgery site in first session of follow up

formations. The salivary calculi are averagely sized between 3.3 to 17.9 mm (9). Usual locations of salivary calculi are the hilus and in the ducts of glands, while in 9 to 17% of all cases, sialoliths occur on the parenchyma(1, 10).

Regardless of its size or location, the ability to palpate the sialolith, is considered to be the most important factor in the successful transoral removal of the stone(11). In most cases, intraoral approach to removal of the obstructing stone is the treatment of choice. In certain instances, as the current case this method can be applied for sialoliths located in the hilus of the submandibular gland. In some cases, when the sialolith is small and located near the orifice, a lacrimal probe can be used to widen the orifice. Sialoadenectomy or partial submandibulectomy is crucial to treat Intraglandular sialoliths (7). In the present case, the patient presented with typical symptoms, and the clinical and radiographic examination revealed typical but somehow oddly sized sialolithes. Howere more investigations revealed the presence of secondary hyperparathyroidism as endocrine disfunction.

The diagnosis was made and confirmed by clinical and radiographic examination, and removal of sialotlith through trans oral approach was the selected treatment.

The mechanisms behind sialolith formation remain unclear. It was suggested that factors such as altered pH of saliva, abnormality in Ca2+ metabolism, and reduced salivary flow rate may contribute to sialolith formation (12). High Ca2+ content of the saliva and the the alkaline pH can play role in high incidence of sialolith formation in the Wharton's duct. Also a long ascending pathway of the Wharton's duct alongside with gravity may contribute to reduced salivary flow rate and leading to sialolith formation (3).

Commonly secondary HPT is the result of chronic renal failure (CRF). Also malabsorption,osteomalacia and rickets are considered other causes of secondary HPT. Phosphate abnormal renal tubular absorption leads to reduced phosphate excretion and hyperphosphatemia. Also Impaired renal conversion of 25- hydroxycholecalciferal to 1,25 dihydroxycholecalciferol results in a decrease in absorption of calcium from intestine. Reduced vitamin D production and elevated serum phosphate levels result in hypocalcemia. prolonged hypocalcemia causes parathyroid chief cell hyperplasia occurs and elevated parathyroid hormone (PTH) secretion (13). However in this case the kidney functions normally and paraclinical and hematological biochemistry test related to kidney function was normal. So in this case sHPT develops as a compensatory mechanism to low calcium intake, increased excretion,or reduced intestinal calcium absorption (14) After all,Williams suggested that stone formation is not associated with systemic abnormalities of calcium metabolism and in patients with sialolithiasis, parathyroid hormone and electrolytes studies have not demonstrated abnormalities (15).

Current case demonstrates that correct selection of surgical treatment plane is crucial to properly treat sialolithiasis and retrive salivar gland fuction with minimal post surgical complications. Also unique aspect of this case is endocrine disorder being concurrent with multiple significantly large sialolithiasis. However a single case of study neither confirm nor refute the role of endocrine disorder in stone formation. However maybe it is wise to consider checking possible endocrine and electrolyte imbalance in order to improve our current understanding of sialolithiasis formation and managing in a proper way beside surgical treatmet.

CONFLICT OF INTREST

Non to declare.

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