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CASE REPORT

RARE CASE, SIALO-ORAL FISTULA WITH GIANT SIALOLITHIES OF THE WHARTHINS DUCT

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ABSTRACT

Sialolithiasis: is the most common disease of salivary glands. It is a condition characterized by the obstruction of a salivary gland or its excretory duct due to the formation of calcareous concretions, resulting in salivary ectasia and even determining the subsequent dilatation of the salivary gland. The sialolith usually measure from 1 mm to less than 10 mm. They rarely measure more than 15 mm. frequently and rare giant salivary gland calculi >15 mm. In this case male 42 years old have a giant stone in the right submandibular gland duct about 45mm which is represented one of the biggest submandibular duct stone reported in the literature with perforation of the wharthins duct, its diagnosed through the long history more than 20 years with pain, tenderness, dysphagia, difficult in specking. Clinical examination reveal extra oral bulginess of the submandibular gland with intra oral fistula in its duct, extra oral lateral radio graphical examination show large stone extended from the first right lower molar to the first right lower central, Its treated by very small trans-oral surgical incision under local anesthesia through the ductal oral fistula just extended anteriorly and posteriorly and the stone get out spontaneously by the action of the pressure of the discharge following the stone removal, No recurrence was seen on follow-up

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INTRODUCTION

Sialolithiasis is one of the most common diseases, its account more than 50% diseases of large salivary gland and is thus the most common cause of acute and chronic infection, Sialolithes of the salivary glands occur mostly in middle-aged patients and approximately 80% of all reported cases of sialoliths occur in the submandibular gland (Leung *et al.*, 1999), Giant sialoliths are classified as those exceeding 15 mm and 1 gram in weight, Although, large sialoliths have been described in the body of salivary glands, they are rarely found in the salivary ducts (Cawson and Odell, 1998). It's about 80% in the submandibular gland or its duct, 6% in the parotid gland or its duct and 2% in the sublingual gland or minor salivary gland (Steiner *et al.*, 1997). The stone its increase in size particularly when the patients have no painful symptoms (Zenk and Benzel, 1994). 40% of parotid and 20% of submandibular stones are not radiopaque and sialography may be required to locate them (Tepan and Rohiwal, 1985) Salivary calculi are usually unilateral and are not a cause of dry mouth (Williams, 1999). clinically they are round or ovoid, rough or smooth and of a yellowish color, They consist of mainly calcium phosphate with smaller amounts of carbonates in the form of hydroxyapatite, with smaller amounts of magnesium, potassium and ammonia (Bodner, 2002).

CASE REPORT

42-year-old male reported visit the privet maxillofacial clinic in Wasit/Iraq with the chief complain of pain and swelling in the right side in the floor of the mouth, detailed history revealed that it started as small intra oral swelling since 20 years back. Its associated with swelling in the right sub mandibular area post meal and subside gradually after meal time, it gradually increased over next years and then it burst leaving a yellowish white mass in the right floor of mouth which was noticed by patient. patient experienced pain, as well as pus discharge from that region.

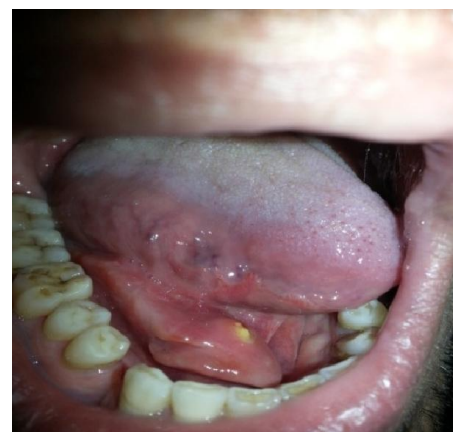


Figure 1.

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Figure 2.

Intraoral examination revealed a well defined oval swelling of approximately 20×10 mm in size in the floor of the mouth in relation to the right lateral canine premolars and first molar teeth. (Figure 1). 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. A provisional diagnosis of sialolith in the right submandibular gland duct was made.



Figure 3.



Figure 4.

Radiographic evaluation in lateral extra oral radiography (Figure 2) which revealed a large well defined oval homogenous radiopacity in the floor of the mouth on right side, where sialolith was enucleated by small trans-oral surgical incision under local anesthesia (Figure 3) through the ductal fistula just extended 2 mm anterior and posterior, the stone got out spontaneously by the action of the pressure of the discharge following the stone it was approximately 45×10 mm in size (Figure 4) No recurrence was seen on follow-up.

DISCUSSION

Sialolithiasis is a pathological condition caused by the obstruction of a salivary gland or its excretory duct by a calculus (Marchal *et al.*, 2001). Think to the stagnation of saliva rich in calcium, the sialolith is the result of the deposition of tricalcic phosphate salts around an initial nidus that consists of altered salivary mucin, desquamated epithelial cells and bacteria (Sidiqi and Sialolithiasis, 2002) The ability of a calculus to grow and become a giant sialolith depend mainly on the affected ducts reaction (Akin, 1991). the sialolith might increase in size, becoming a giant calculus, and remain asymptomatic for a long period. when the sialolith is located in a duct able to dilate and to allow a normal secretion of salivary flow around the stone (Batori *et al.*, 2005), Giant sialoliths are usually organized hard concretions, with a porous aspect, yellow color and hard texture. They usually have an oval or long shape and in radiographic images they can appear like teeth (Zenk, 1994)

Experimental studies have identified the increased salivary magnesium concentration as a key factor in determining the sialolithiasis (Steiner *et al.*, 1997). the deposition of salivary calculi is not associated with systemic diseases involving calcium metabolism (Marchal *et al.*, 2001), Gout is the only metabolic disease that predisposes, among others to salivary stone formation. However, the gout calculi consist, typically of urates and the urate sialoliths are ovoid or round, smooth or rough with a yellowish color, they consist of calcium phosphate with small accounts of hydroxyapatite, magnesium, potassium and ammonia (Siddiqui, 2002). bacterial infection are important factor involve into calculi's formation, toxins produced by bacteria, can produce a local environment with pH less than 5.5, that causes tissue damage, when tissue healing processes re-establish the 7.2 pH crystallization of salivary ions especially calcium phosphates occurs (Tepan and Rohiwal 1985). The submandibular gland is more susceptible to the development of the salivary calculi than parotid gland (Steiner *et al.*, 1997) because:

- The Warthon's duct is longer than /the Stensen's duct.
- The salivary flow is against gravity in the submandibular gland.
- The salivary submandibular pH is more alkaline and mucin proteins, calcium and phosphates are contained in greater amount than serous parotid saliva (Steiner *et al.*, 1997).

Regarding the diagnosis, in order to avoid that calculi remain unknown, is crucial to perform a careful history and to recognize clinical symptoms. Giant salivary gland duct calculi

Is characterized by salivary duct swelling, without any obvious reason or at meal times. The symptoms, referred by the patients during the meal times, are due to the higher stimulation of the salivary secretion and to the duct's obstruction that prevents its smooth flow. When affected by a salivary colic, the patient refers an acute pain, sense of swelling and pressure in the floor of the mouth (Batori *et al.*, 2005).

Conclusion

The diagnosis of giant sialolith in the Wharton's duct is further simplified when it presents as a hard mass in the floor of mouth specially when causing sialo-oral fistula. The diagnosis of sialolithiasis is based on several imaging techniques, ultrasonography represents an excellent first-level diagnostic technique because it reveals ductal and highly mineralized stones with a diameter of at least 1.5 mm with an accuracy of 99 % conventional intra-oral X-ray may be more useful than extra-oral radiography, particularly trans-occlusal radiography. In some cases the bimanual posterior-anterior palpation of the mouth's floor allows to determine the presence of a giant calculus, which appears like a palpable mass. The treatment of choice of small sialolith should be medical instead of surgical however, if the calculus is of a medium or large size, like the giant salivary gland calculi, a salivary colic may occur and the sialolith cannot be expelled spontaneously. The ultimate objective of giant sialoliths treatment is restoring a normal salivary flow. whenever the stone can be palpated intraoral, the best option is to remove it through an intraoral approach.

The clinicians should evaluate carefully the painful or painless swellings in submandibular area. this condition seems to be the most common disease in submandibular gland and Wharthon's duct due to the presence of gland Lithia's. Large submandibular sialoliths should be treated by appropriate approach to avoid possible severe post operative complications,the choice of a surgical approach to access the sialolith with submandibular gland preservation requires careful imaging evaluation and minimal invasive removal, transoral sialolithotomy. after surgical calculi removal the patients show asymptomatic and normally functioning glands in a short time,transoral sialolithotomy remains main stay of the treatment for giant sialolith in the duct of submandibular gland. Also, patients should be followed up regularly as recurrence has been reported in the literature.

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