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Case Report

A rare case of large submandibular sialolith with spontaneous extrusion

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Abstract Sialolithiasis is one of the commonest disease of salivary glands. Submandibular salivary gland or its duct is a major site of sialolithiasis and it is always leads to sialadenitis. However large sialolith formation and self-extrusion is a rare condition. A case of submandibular sialolithiasis is reported where patient presents with recurrent submandibular swelling and subsequently end up with a spontaneous extrusion. The mechanisms and management of the salivary gland calculi are also discussed.

Keywords: Extrusion, sialolithiasis, submandibular salivary gland.

Introduction

Sialolithiasis is the commonest benign disease of salivary glands characterized by calcified concrement formed within salivary glands. It is usually occurs in its duct causing obstruction of salivary secretion and outflow. This condition accounts for approximately one third of the salivary gland pathology (Williams, 1999). It is the most common cause of acute and chronic sialadenitis. Leung *et al*. (1999) estimated that it affects 12 in 1000 of the adult population in Ethiopia. This is probably due to the dryness because of inadequate water supply. Sialolithiasis appears in the third to sixth decades of life, commonest between fourth and fifth decade of life and it is almost equally distributed among genders according to the most recent study (Kraaij *et al*., 2014).

Submandibular salivary gland has the highest occurrence rate, where more than 80% of sialolithiasis, occurs in this gland, followed by 19% in parotid and 1% in the sublingual gland or minor salivary glands (Batori *et al*., 2005). Swelling is the commonest symptom in the submandibular sialolith (more than 80%) followed by pain (less than 50%) and 3% of patients are asymptomatic. The duration of symptoms varies from days to years depends on its onset, size and site of sialolith.

Case summary

A 50-year-old gentleman presented with acute onset of right submandibular swelling for 2 days associated with intermittent throbbing pain. Both swelling and pain were episodic and aggravated by eating. He had a similar episode last 2 years where he experienced extrusion of a small stone over the floor of mouth followed by long duration of remission. For current episode, an Orthopantomogram (OPG) was done showed a right submandibular calculi (Fig. 1). In view of tolerable symptoms, he was treated conservatively with antibiotic and analgesic. He claimed that he practiced regularly massaged submandibular gland especially when it started to cause pain during eating.

Following ORL clinic presentation at 3 weeks, he extruded a large calculi in floor of mouth and the symptoms resolved (Fig. 2). Neck examination revealed a localized tender right submandibular swelling measuring 3 cm x 2 cm (previously measured 5 cm x 4 cm). On intraoral examination, the right Wharton's duct looked dilated and the submandibular gland was ballotable. He was treated for acute sialedenitis. There was no recurrence upon follow up.

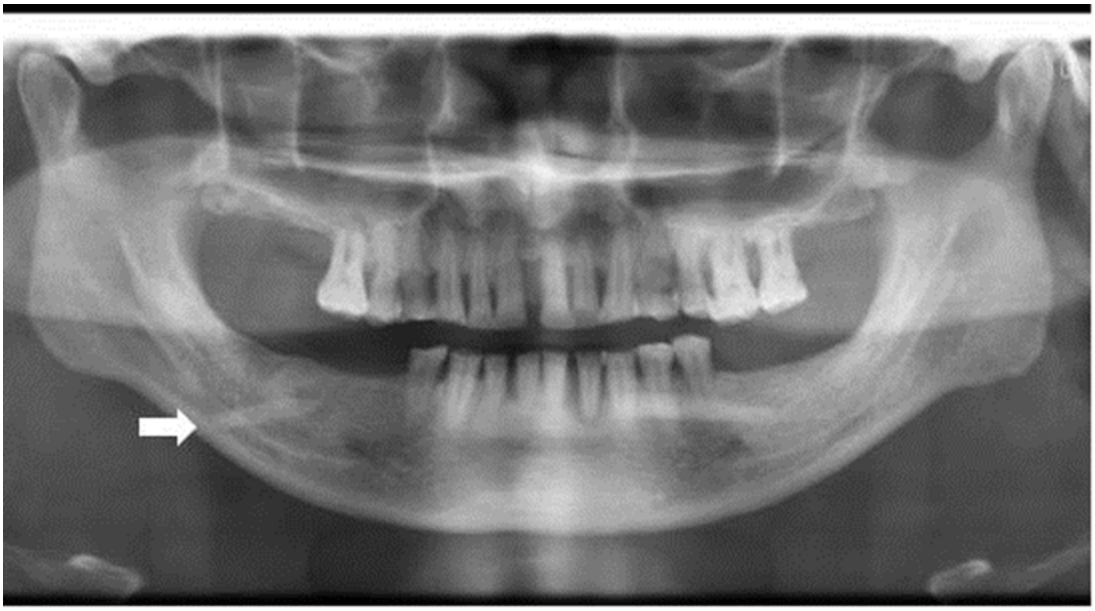

Fig. 1 OPG reveals a well-defined radiopacity (white arrow) over right submandibular region, corresponds to submandibular calculi.

Fig. 2 Submandibular calculi measuring 1.7 cm in length.

Discussion

Sialolithiasis is common and represent more than 50% of large salivary gland diseases. The aetiology of submandibular sialolithiasis is explained by 3 hypotheses. Firstly is the anatomical variation of salivary ducts that predisposes to the accumulation and stasis of saliva. About 80-90% of submandibular calculi located in Wharton's duct (57% in the hilum and 34% in the distal duct), only 10% found in the parenchyma. Wharton's duct runs parallel to floor of mouth and course upward, ascending between mylohyoid and hyoglossus muscles. Its angulation, large diameter with narrow orifice (0.1-0.5 mm) can favor an antigravity salivary flow (Haring, 1991). Also, the submandibular gland saliva is alkaline with a high mucin facilitates salivary flow stasis whereas parotid gland saliva is serous in nature (Haring, 1991).

Secondly, sialomicroliths is identified in almost all normal submandibular glands

but only 10–20% of the normal parotid glands (Harrison *et al*., 1997). These microscopic concretion contains calcium, phosphorus, organic secretory material and necrotic cells. Submandibular sialolithiasis has higher calcium and phosphate concentration which is corresponds to sialomicrolith. The mass of sialomicroliths become impacted causes stagnation of salivary flow. Micro and macro obstruction results in sialadenitis.

Thirdly, there is different biochemical composition of saliva. Sialoliths are mainly composed of inorganic material (hydroxyapatite, octacalcium phosphate, whitlockite and brushite). Submandibular sialolithiasis contains 82% inorganic and less organic materials (collagen, glycoproteins, amino acids, carbohydrates, lipids and phospholipids) (Zenk *et al*., 1994). Lack of organic properties, there is lack of initiation of mineralization of calculi. Hydroxyapatite and whitlockite are often found in submandibular calculi. Parotid calculi has more balance composition, it composed of 49% inorganic and 51% organic material (Zenk *et al*., 1994). Other minor inorganic components are potassium, sodium, ferum, silicon, magnesium, brimstone, ammonium and chloride. Besides that, the higher pH and viscosity of submandibular saliva secondary to high mucous concentration predispose to sialolithiasis. Taking accounts of different pathogenesis, submandibular calculi is formed around a nidus of mucous whereas parotid calculi often form around a nidus of inflammatory cells or a foreign body.

There are many other predisposing factors such as dehydration, inflammation of the gland, gout, trauma, stricture of duct, smoking, sjögren's syndrome and drugs (diuretics, anti-cholinergic medications) (Huoh and Eisele, 2011). The factors correspond to our patient are site of the sialolith and smoking. Clinical examination must be completed by an intraoral bimanual palpation and gentle probing. The challenge is to detect the location, size of sialolith, number of stones, unilateral or bilateral glands involvement. OPG is helpful in majority of diagnosis as 80% of submandibular and 60% of parotid calculi are radiopaque. Ultrasonography has a sensitivity of 99%, and it can detect calculi with a diameter of 2 mm or more. Sialography is required for suspected case of radiolucent sialoliths with recurrent and persistent symptoms or deep seated sialolith. However, it is contraindicated for iodine allergy and acute infection. In general, most of the sialoliths are usually of 5 mm in size and any calculi over 10 mm in maximum diameter is considered as unusual size (Isacsson *et al*., 1984).

Our patient extruded a large sialolith measuring 17 mm; this is an unusual condition with spontaneous extrusion. This is probably facilitated by frequent massage by the patient. Computed tomography scan able to detect any size of sialolith and magnectic resonance imaging sometimes is used for better soft tissue delineation in cases of sialadenitis.

Conservative management includes hydration, warm moist compression, gland massage, sialogogues, and salivary duct irrigation. Antibiotic therapy is applied in case of presence of both local and systemic infection. Most of cases benefit from the conservative treatment just like our patient. Larger sialolith is more difficult to be dissolved by salivary enzyme but there is a chance of spontaneous extrusion.

Sialoliths which located near to the orifice of the duct or within the prehilar region better is for transoral removal. Sialolithotomy under local anesthesia is indicated for small and assessable sialolith. Diagnostic and therapeutic sialoendoscopy helps salivary duct catheterization and removal of calculi. Extracorporeal shock-wave lithotripsy is described as invasive treatment for big calculi, practically this is not routinely done. Submandibulectomy is indicated following failure of conservative treatment or in cases of proximal sialolith localization (Seward, 1968). If our patient has persistent or worsening of symptoms and have not extruded the sialolith, right submandibulectomy would be the surgical choice.

Conclusion

The site and size of sialolith and the severity of symptoms determines the choice of treatment. The sialolith can be slowly

increased in size, it can be recurrent or it can causes local or systemic infection. It also can be dissolved or extruded spontaneously. The aim of both conservative and surgical treatment is to restore salivary secretion and to eliminate the sialolith.

References

Batori M, Mariotta G, Chatelou H, Casella G, Casella MC (2005). Diagnostic and surgical management of submandibular gland sialolithiasis: report of a stone of unusual size. *Eur Rev Med Pharmaco Sci*, **9**(1): 67-68.

Haring JI (1991). Diagnosing salivary stones. *J Am Dent Assoc*, **122**(5): 75-76.

Harrison JD, Epivatianos A, Bhatia SN (1997). Role of microliths in the aetiology of chronic submandibular sialadenitis: a clinicopathological investigation of 154 cases. *Histopathology*, **31**(3): 237-251.

Huoh KC, Eisele DW (2011). Etiologic factors in sialolithiasis. *Otolaryngol Head Neck Surg*, **145**(6): 935-939.

Isacsson G, Isberg A, Haverling M, Lundquist PG (1984). Salivary calculi and chronic sialoadenitis of the submandibular gland: A radiographic and histologic study. *Oral Surg Oral Med Oral Pathol*, **58**(5): 622-627.

Kraaij S, Karagozoglu KH, Forouzanfar T, Veerman ECI, Brand HS (2014). Salivary stones: symptoms, aetiology, biochemical, composition and treatment. *Br Dent J*, **217**: E23.

Leung AK, Choi MC, Wagner GA (1999). Multiple sialolths and a sialolith of unusual size in the submandibular duct. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod*, **87**(3): 331-333.

Seward GR (1968). Anatomic surgery for salivary calculi. IV. Calculi in the intraglandular part of the submandibular duct. *Oral Surg Oral Med Oral Pathol*, **25**(5): 670-678.

Williams MF (1999). Sialolithiasis. *Otolaryngol Clin North Am*, **32**(5): 819-834.

Zenk J, Benzel W, Iro H (1994). New modalities in the management of human sialolithiasis. *Minim Invasive Ther*, **3**(5): 275-284.