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REVIEW ARTICLE

GIANT SIALOLITH IN SUBMANDIBULAR REGION- A CASE REPORT WITH REVIEW OF LITERATURE

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ABSTRACT

Sialolithiasis is one of the most common pathology of salivary gland in middle-aged patients . Sialoliths are localized in submandibular glands in nearly 80% of the reported cases and they are classified as 'giant' in case any dimension exceeds 15 mm. Giant sialolith in submandibular gland is a rare disorder with prevalence of 0.1% in population. The aim of this article is to report the case of a patient suffering from sialolithiasis caused by giant salivary calculi in Wharton's duct and also review of literature on sialolithiasis. The diagnostic approach was done by occlusal radiography, orthopantamography and ultrasonogram and the excision was performed under general anesthesia to remove the atrophied submandibular gland and a giant sialolith of 28mm length localized in the Wharton's duct.

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INTRODUCTION

Sialolithiasis is one of the most common diseases of salivary glands. Sialolith causes swelling, pain, obstruction of the salivary flow and recurrent infections of the associated gland. It is estimated that it affects 12 in 1000 of the adult population (Alyas, 2005). It is more prevalent in middle ages between 30 and 60 years with male predominance. Sialoliths most commonly occur in the submandibular gland (80–95%), whereas 5 – 20% are seen in the parotid gland, 0-5% in sublingual gland or minor salivary glands (Baurmash, 2004). Intraglandular localization of salivary calculi is rarer when compared with intraductal localization. Sialolith formation is more likely to occur in the submandibular gland due to its anatomic position requiring the salivary flow against gravity, longer and more tortuous duct and production of alkaline saliva rich in mucin (Austin, 2004). Most submandibular calculi are detected as radiopaque formations in plain radiographs and as radiolucent filling defects in sialography. Approximately 20% of sialoliths are not radiopaque, and sialography or sialendoscopy may be required to diagnose them. Salivary calculi are usually unilateral and do not cause dry mouth.

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According to the literature, sialoliths measuring >15 mm in any dimension or weighing 1 g are named as 'giant' (Bodner, 2002 and Soares, 2009). Giant sialolith of the submandibular gland is rarely reported. A case of giant sialolith of submandibular gland, which is 28 mm in length, is presented in this case report.

CASE REPORT

A 35-year-old female patient (Fig -1) reported our department with the complaints of recurrent pain, swelling and redness in the left submandibular area for past one year. H/O swelling for past one year, increased in size during meal time, and reduced in size immediately after meals. H/O continuous pain for past 4 days. On extra oral examination, facial asymmetry evident with the presence of a single diffuse swelling in left lower third of the face and extends to the neck in submandibular area (Fig-2) roughly measuring 4x5cm. Swelling and hyperemia on the left submandibular region was observed. Bimanual palpation revealed a hard and tender mass (Fig-3), normal salivary flow was present and the orifice of Wharton duct is hyperemic, swollen and ulcerated, and the floor of the mouth is elevated in the left. A preliminary diagnosis of submandibular sialolithiasis was made, mandibular occlusal radiograph (Fig-4) showed conical shaped radiopaque mass extending in the left molar region in wharton's duct, orthopantamogram (Fig-5) also showed conical radiopaque mass in left second premolar

and first molar region and Ultrasonogram (Fig-6 a & b) was performed which showed presence of calculi of size 2.7 x 1.1 x 1.2cm seen in the left sublingual region with mass effect on left sublingual gland, the left submandibular gland is atrophied and right submandibular gland is normal in size and echo texture.

Fig. 1. Patient profile

Fig. 2. Submental view

Fig. 3. Intra Oral

Fig. 4. Mandibular Occlusal Radiograph

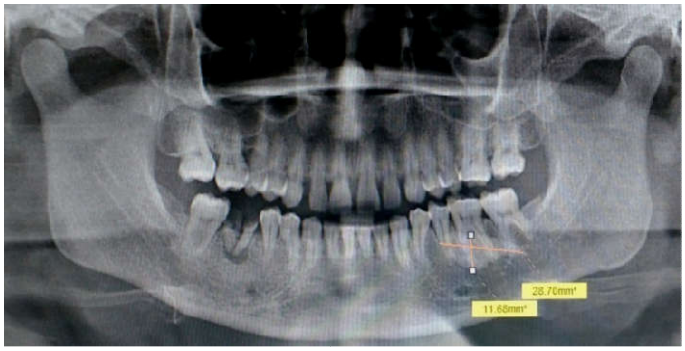

Fig. 5. Orthopantamogram : presence of conical radiopaque mass in left second premolar and first molar region

Fig. 6a. Ultrasonogram : 2D high resolution ultrasound performed using linear transducer. Presence of calculi of size 2.7x1.1x1.2cm seen in the left sublingual region with mass effect on left sublingual gland

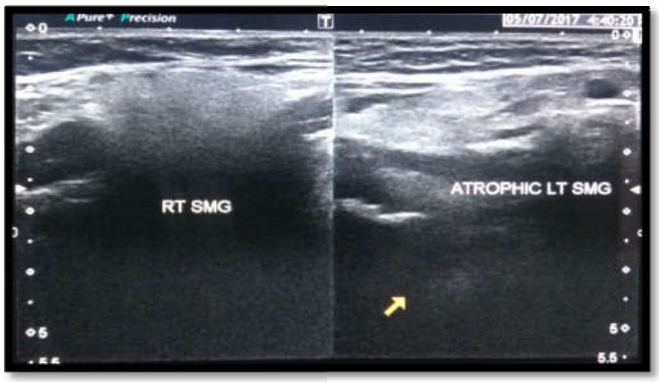

Fig. 6b. The left submandibular gland is atrophied and right submandibular gland is normal in size and echotexture.

Fig.7. Excised specimen

Few sub centimeter hypoechoic lymphnodes are seen in the bilateral; cervical levels 1b and 2 (3-5mm short axis). Oral antibiotics and non-steroid anti-inflammatory drugs for 2 weeks were prescribed to treat the acute inflammation of the gland and a follow-up visit was planned. Two weeks later, it was seen that the acute inflammation of the gland had subsided and resection of submandibular gland was planned with the informed consent of the patient. Excised specimen was sent for histopathological examination. Pathologic examination confirmed the diagnosis of chronic sialadenitis and a giant sialolith of 28 mm in size (Fig-7) The postoperative period was uneventful and the patient recovered without any complications.

DISCUSSION

Sialolithiasis is a condition where a calcified mass or sialolith forms within a salivary gland or duct. Although the exact etiology is unknown, sialoliths are thought to occur as a result of deposition of mineral salts around an initial nidus consisting of salivary mucin, bacteria or desquamated epithelial cells (Austin, 2004). Stagnation of salivary flow, high alkalinity and increased calcium content predispose to sialolith formation. Salivary calculi are more often found in submandibular gland than parotid gland due to some factors such as the direction of salivary flow against gravity, a longer and more tortuous structure of Wharton duct and the higher calcium and mucin content of saliva produced in the submandibular gland (Bodner, 2002). The deposition of salivary calculi is not associated with systemic diseases involving calcium metabolism. Gout is the only metabolic disease that predisposes, among others, to salivary stone formation. However, the gout calculi consist, typically, of urates. Sialoliths are ovoid or round, smooth or rough with a yellowish colour. They consist of calcium phosphate with small accounts of hydroxyapatite, magnesium, potassium and ammonia. Bacterial infections are important factors involved into calculi's formation. Toxins produced by bacteria can produce a local environment with pH of less than 5.5 which results in tissue damage (Work, 1980 and Escudier, 1998). Most calculi occur in Wharton duct, whereas the rest are found at the ductal hilum or within the gland. Sialoliths of submandibular gland are rarely radiolucent. They were reported to be found more frequently in the left submandibular gland as in our case (Goudot, 1986). Sialoliths usually remain within the gland and enlarge but they rarely migrate to other locations. Sialolithiasis of submandibular gland usually has an asymptomatic course. The symptoms begin whenever the lumen of Wharton duct is obstructed by a salivary calculus with resultant accumulation of saliva causing pain and swelling of the involved gland (Goudot, 1989 and Iqbal, 2012). The stasis of saliva can lead to bacterial ascent into the parenchyma of the gland and therefore recurrent infections. Long-term obstruction of salivary flow and recurrent infections can lead to atrophy of the gland with resultant loss of secretory function and ultimately fibrosis.

Conventional intra-oral X-ray may be more useful than extra-oral radiography, particularly ooclusal radiography. Ultrasonography are used to visualize the images of with sialolith size larger than 2 mm (Iwona Rzymska-Grala, 2010). Sialography is an adequate technique to detect salivary gland calculi that allows the visualization of the whole duct system. However, sialography is not indicated in the case of acute infections or patients sensible to contrast medium and also when a radiopaque calculus is observed in the distal portion of the duct , because the injected contrast medium could move the calculus to the proximal portion i.e., closer to the gland and thus complicating its removal. Scintigraphy could be performed in the event of a suspected sialolithiasis, when sialography is not indicated and in patients with no permeable glandular ducts. Computerized tomography and nuclear magnetic resonance have changed the imaging techniques of detection of sialoliths. CT-Scan has become the examination of choice to detect gland salivary sialolith, because not invasive like sialography. Sialoendoscopy is a new diagnostic means of directly visualising intra-ductal stones that has bridged the diagnostic gap between the clinical suspicion of salivary obstruction and the limitations of conventional radiology (Cappaccio, 2007). The primary objective of treatment for giant sialoliths is the restoration of normal salivary secretion and prevent the damage to the gland. The treatment options for submandibular sialoliths are intraoral removal of stone, interventional sialendescopy and resection of the gland. The appropriate treatment method is chosen according to the size, location and number of sialoliths. Small sialoliths can be removed through the duct orifice using bimanual palpation. Larger or proximally located sialoliths in the duct require surgical removal through oral cavity (Steiner, 1997). Submandibular gland resection is indicated when a sialolith with a substantial mass (12 mm or more) is located within the gland and intraoral surgical access is not possible.

Newer treatment modalities such as endoscopic intracorporeal shockwave lithotripsy and extracorporeal shockwave lithotripsy have been introduced as alternatives to conventional submandibular gland surgery (Walvekar, 2009). Other techniques for sialolith fragmentation such as electrohydraulic and pneumoblastic devices have been described. Sialendoscopy has been used as both a diagnostic and an interventional modality in salivary sialolithiasis. Small salivary intraductal calculi may be removed and biopsies may be taken using sialendoscopy systems. Submandibular sialoliths of large sizes are extremely rare and should be treated after considering their size (McGurk, 2005 and Abdullah, 2016). However, for giant sialoliths with chronic sialadenitis causing atrophy to the gland as presented in our case, submandibular gland resection remains the main stay of treatment.

Conclusion

Giant sialolith of remarkable size may pose diagnostic and therapeutic challenge for the clinician. They are usually seen in 4^{th} & 5^{th} decade of life in males predominantly in submandibular region. Transoral sialolithotomy or sialadenectomy remains the main stay of management. Newer treatment modalities are effective alternatives to conventional

surgical treatment for smaller but not probably for giant sialoliths.

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