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RARE GIANT SUBMANDIBULAR GLAND CALCULUS: A CASE REPORT

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ABSTRACT: Salivary gland calculus / Sialolithiasis is the most common disease of the salivary glands. Sialolithiasis is most commonly found in the submandibular gland and its duct (Wharton's duct). Giant salivary gland stones measuring over 3cm are extremely rare with only scanty reported cases¹. This report describes the case of a patient who had a painless very rare giant Submandibular sialolith visible per orally on the floor of mouth. A 5× 2 cm sized stone was excised.

INTRODUCTION: Sialolithiasis (also termed salivary calculi, or salivary stones), is a condition where a calcified mass forms within a salivary gland. The term is derived from the Greek words sialon (saliva) and lithos (stone), and the Latin -iasis meaning "process" or "morbid condition".

Sialolithiasis is the most common disease of salivary glands ¹. Stones or calculi can be found in all major and minor salivary glands and their ducts. More than 80 % of the calculi are found in Submandibular gland and its duct (more common)². Parotid gland accounts for 10% of cases. Remaining cases involve Sublingual and minor salivary glands².

Submandibular calculi are most commonly seen in young and middle aged adults with equal affection of both sexes³. Longer and upward course of the submandibular duct, non- dependent drainage, high calcium and mucin content, more viscid secretion are the factors which favour the formation of submandibular duct stones ³. Bilateral or multiple gland sialolithiasis occurs in less than 3% of cases⁴.

Commonly sialoliths measure from 1mm to less than 1cm. Giant salivary gland stones (GSGS) are those stones measuring over 1.5cm and have been rarely reported. GSGS measuring over 3cm are extremely rare with only scanty reported cases ⁵. The aim of this case report is to highlight a case of a very rare giant sialolith.

CASE REPORT: A 40 yr old male, farmer by occupation presented to the General Surgery out patient department for a firm, painless, yellowish brown mass in the right side of floor of mouth for 2 yrs. He was a chronic smoker and tobacco chewer with a poor oral hygiene. Medical history of the patient was unremarkable. Patient complained of occasional episodes of mild pain over right jaw during meals.

No palpable swelling or mass was revealed on extra oral examination. In intraoral examination a hard, mobile, non tender, yellowish brown mass was palpable along the anatomical location of the Wharton's duct. All the teeth were stained. OPG revealed a large calcified mass at that area. With informed consent of the patient, under local anaesthesia, the mass was excised per orally after breaking adhesions using an artery forceps. Patient was discharged on outpatient basis with prescription of oral antibiotics and povidone iodine gargles. Recovery of the patient was complete with no fresh complaints on follow up.

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DISCUSSION: There are thought to be a series of stages that lead to the formation of a calculi (lithogenesis). Initially, factors such as abnormalities in calcium metabolism,⁷ dehydration,⁶ reduced salivary flow rate,⁶ altered acidity (pH) of saliva caused by oropharyngeal infections,⁶ and altered solubility of crystalloids,⁶ leading to precipitation of mineral salts, are involved.

The next stage involves the formation of a nidus which is successively layered with organic and inorganic material, eventually forming a calcified mass^{6,7}. In about 15-20% of cases the sialolith will not be sufficiently calcified to appear radiopaque on a radiograph,⁷ and therefore be difficult to detect.

Other sources suggest a retrograde theory of lithogenesis, where food debris, bacteria or foreign bodies from the mouth enter the ducts of a salivary gland and are trapped by abnormalities in the sphincter mechanism of the duct opening (the papilla), which are reported in 90% of cases. Fragments of bacteria from salivary calculi were reported to be Streptococci species which are part of the normal oral microbiota and are present in dental plaque.⁶

Sialolithiasis may also develop because of the presence of existing chronic infection of the glands, dehydration (e.g. use of phenothiazines), sjogren's syndrome and/or increased local levels of calcium, but in many instances the cause is idiopathic (unknown).

Salivary calculi sometimes are associated with other salivary diseases, e.g. sialoliths occur in two thirds of cases of chronic sialadenitis⁸, although obstructive sialadenitis is often a consequence of sialolithiasis. Gout may also cause salivary stones⁸, although in this case they are composed of uric acid crystals rather than the normal composition of salivary stones.

Giant sialoliths are classified as those exceeding 1.5 cm in any one dimension. Giant salivary calculi are thought to form in salivary ducts, which allow expansion and permit salivary flow around the stone. Stones may slowly increase in size, remaining asymptomatic for a more substantial period of time⁴. Subsequently, most giant salivary calculi adopt an oval or elongated shape. Giant calculi are described as being hard in texture, yellow in colour and with a porous aspect⁹.

The predisposition to calculi, and ability to tolerate expansion, lead to a higher incidence of giant calculi associated with submandibular gland.

Although large sialoliths have been reported in both salivary glands and their ducts, stones larger than 3cm are rare⁵. Mean size is reported as 6 to 9mm¹¹. A review of literature in 2007 found only 16 reported cases of sialoliths having a size up to 3.5cm¹⁰.

Patient's generally present with pain, discomfort and swelling before or during meals. 80-90% of Submandibular stones are radio-opaque due to their high content of calcium and magnesium carbonates & phosphates³ leading to their easy detectability in standard x-ray films. Other diagnostic methods include Sialography, USG, CT and Scintigraphy. Submandibular stones can be removed surgically through either intraoral or an external approach¹¹. For stones located entirely in the duct and close to the papillae, intraoral approach is sufficient. Extra oral approach is indicated for intra glandular stones and stones embedded into the hilum of gland.

Newer treatment methods are External lithotripsy and Interventional sial-endoscopy (including wire basket extraction and fibre optic laser lithotripsy)¹¹. Sial-endoscopy uses minimal invasive surgical techniques which allows for optical exploration of the salivary ductal system and extraction of the stones by a basket under endoscopic view. This technique can be performed in most cases as an ambulatory, outpatient procedure under local anaesthesia. They are two types of sial-endoscopy, diagnostic and interventional. Interventional sial-endoscopy is used to treat

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disorders discovered during diagnostic sial-endoscopy. The attitude is the same for the submandibular and parotid glands. For small stones less than 4 mm in diameter in submandibular cases and less than 3 mm for parotid cases, extraction is performed with custom designed wire baskets of various sizes. In cases of bigger stones, a priori fragmentation is necessary, using a laser system, or possibly an extracorporeal lithotripter.

Development & recurrence of salivary gland stones can be prevented by following simple habits like drinking plenty of water every day, massaging the salivary gland after meals to break up thickened saliva, cessation of smoking & tobacco chewing, practising good oral hygiene, with regular tooth brushing and flossing. Sucking on sour drops will help maintain a good flow of saliva. Salivary stimulants, such as Salagen, can further facilitate saliva flow. Avoiding older-style over-the-counter antihistamines in favour of newer, non-sedating antihistamines may be useful. Choosing antidepressants that have less drying effects may also be preventive.

CONCLUSION: This case highlights a case of very rare, giant Submandibular duct stone in a patient with poor oral hygiene and neglect for his own health. Patients should be educated regarding the underlying pathology and emphasizing the value of adequate hydration and oral hygiene. Once the diagnosis is established attempts at removal by minimally invasive techniques should be considered. However, excision of a salivary stone of such a large size is a rare medical entity.

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Fig. 1: Per oral extraction of submandibular calculus

Fig. 2: Cavity on the floor of mouth after calculus extraction

Fig. 3: Extracted calculus

Fig. 4: 5x 2 cm sized calculus

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