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"A GIANT SIALOLITH WITH PERFORATION OF THE FLOOR OF MOUTH."

*Sanjana V. Nemade *Vidya V. Rokade. *Dr. Netra A. Pathak

*Smt. Kashibai Navale Medical college and General Hospital, Pune.

ABSTRACT

Salivary gland calculus (Sialolith) is the most common disease of the salivary glands. The majority of sialoliths occur in the submandibular gland or its duct and are a common cause of acute and chronic infections. We report a case of a male patient who had intraductal giant sialolith in the proximal aspect of the submandibular duct impacted at the posterior curve of the duct with perforation of the floor of mouth. The patient was treated successfully with excision of submandibular salivary gland and intraoral removal of the calculus.

KEY WORDS

Giant sialolith; Submandibular duct; Submandibular salivary gland; Perforation of the floor of mouth

INTRODUCTION

Sialolithiasis is the most common disease of the submandibular glands in middle aged patients. It is estimated that it affects 12 in 1000 of the adult population. Males are more frequently affected than females and children are rarely involved . Sialolith can form in any of the salivary glands of the head and neck, but the submandibular gland is the commonest site (80.0% - 90.0%). Lower incidence of calculi is found in the parotid duct and the sublingual duct (6.0% to 20.0%), (1.0 to 2.0%) respectively [1,2].Submandibular calculus close to the hilum of the gland or in the gland itself, tend to become large before they become symptomatic . Commonly, sialoliths measure from 1mm to less than 10mm. Giant salivary gland calculi are those stones measuring over 10 mm and have been rarely reported in the medical literature [3,4].

Very few cases of submandibular calculus with perforation of the floor of mouth have been reported. Zakaria [5] presented a case of a 70 years old male patient with a 20 years history of calculus manifested at the floor of the mouth as fibrosis and erosion. Management was done via transoral surgical sialolithectomy. The case reported by Akin and Esmer [6] showed a giant sialolith located completely inside the submandibular gland that finally led to erosion of floor of the mouth. On the other hand, Asfar et al [7] reported three cases of giant submandibular intraglandular stones. One of them had eroded through the floor of the buccal cavity forming an orocervical fistula and pus discharge. It was preceded by swelling at the submandibular area and side of the neck. The other two cases led to fibrosis and erosion of the mouth floor. Management

of all the previous intraglandular calculi was via extraoral complete surgical excision by gentle extirpation of gland and stone.

Most of the reported giant sialoliths are intraglandular. Intraductal giant sialoliths are rarely reported in the medical literature. We report this case for its exclusive presentation of giant sialolith in the proximal aspect of the submandibular duct impacted at the posterior curve of the duct with long standing perforation of the floor of mouth and chronic submandibular sialoadenitis. The patient was treated successfully with submandibular gland excision and intraoral removal of the calculus through the perforation.

CASE REPORT

A 46-year-old male patient came for consultation in ENT outpatient department of our Institute. He complained of a, firm mass in the left submandibular area. He had a history of having episodes of left submandibular swelling occurring with meals since last 5 years. 15 years back, he had history of abscess formation in the floor of mouth that burst open in the oral cavity with a huge calculus within. This episode was followed uneventful for 10 years. After this he started getting the left submandibular region swelling and pain on and off especially during meals. The past medical history was unremarkable.

On otolaryngologic examination, palpation revealed a firm and non tender swelling in the left submandibular region. No dental caries was detected clinically. We found perforation of the left floor of mouth adjacent to third molar.(Fig.1) It was like a well-established channel with smooth lining. The part of calculus could be visualised deep inside the channel. Ultrasonography

revealed 16 * 10 mm calculus in the left submandibular duct in its proximal aspect with changes of chornic sialoadenitis. Fine needle aspiration cytology showed changes of chronic sialoadenitis. Computerised tomography showed a calculus in the submandibular region medial to the angle of mandible (Fig.2). Findings on blood and serum biochemistry were within normal limits.

The treatment plan was complete surgical excision of submandibular salivary gland and the calculus under general anesthesia The incision was taken 2 finger breadth below and parallel to the margin of the left mandible. Dissection was performed to expose the gland. It was fibrosed and firmly adherent to the mylohyoid muscle. The submandibular duct was unusually dilated (Fig.3) We tried to remove the calculus through the duct, but was unsuccessful as the calculus was impacted at the mylohyoid curve of the duct. We excised the submandibular gland and ligated the duct proximal to the calculus after identifying the lingual nerve where it curves around the duct. The wound was closed in layers. We planned for second surgery by intraoral approach for the removal of calculus after external wound healing. On third post operative day, patient complained of purulent discharge through the intraoral perforation along with swelling in the submandibular area. On aspiration, 5cc pus was aspirated through the wound. We posted the patient for intraoral removal of the calculus. The calculus was found impacted deep in the floor of mouth at the posterior curve of the submandibular duct (Fig.4). We widened the perforation by elliptical incision around the perforation. We removed the calculus and drained the pus intraorally. Post operative period was uneventful .

Histhpathology of the salivary gland revealed features of chronic sialoadenitis. There was no malignancy in the sections studied. The sialolith measured 16 * 10 mm. The yellowish sialolith was oval and had irregular surface. (Fig.5)

DISCUSSION

Sialolithiasis is the main cause of obstructive salivary diseases, being involved in 66% of cases and accounting for about 50% of major salivary gland diseases [8].Several factors contribute to the development of salivary stones in the submandibular gland. The saliva from the submandibular has a high content of mucin and flows uphill in a wider and longer duct as compared to the parotid gland. Stenson's duct (parotid gland) is narrower and the serous saliva from the parotid gland flows down hill. In addition, the saliva secreted by the submandibular gland is more alkaline and has a higher content of calcium and phosphate which promotes stone formation. [8,9]

The submandibular duct (also known as Wharton's duct) allows the passage of saliva from the submandibular gland to the sublingual papilla located anteriorly. The duct extends anteriorly from the submandibular gland curving over the posterior edge of the mylohyoid muscle (comma area) into the sublingual space. It is approximately 5 - 6cm in length and has a diameter of approximately 1 - 3mm on conventional sialographic images.[10] 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.[11] Giant sialoliths are a rare finding; their sizes vary from approximately 10 mm to 7 cm. Our sialolith was 16 mm* 10 mm in size.

It was reported that salivary gland calculi may reside intraductally or within the gland substance itself (intraglandular).Intraductal calculi are divided into proximal and distal ones depending on their relative position in Wharton's duct (Above or below the mylohyoid muscle as the duct curves around the muscle to come in the anterior 2/3 of the floor of mouth.) About 40% of calculi are located distally in Wharton's duct (In anterior 2/3 of the floor of mouth). The rest are either intraglandular or near the hilum of the gland and prone to increase in size by time. This enlargement occurs due to an extra deposition of minerals, duct lining cells, debris and bacteria. In this case the giant calculus (16mm*10mm) was impacted at the posterior aspect of the submandibular duct where it curves around the mylohyoid muscle with perforation of the floor of mouth.

The symptoms are consequence to obstruction of salivary flow. They may be presented mainly at meal time as pain and swelling distal to stone obstruction area. This usually makes the diagnosis clearer and straight forward. Our patient had past history of submandibular calculus with abscess in the floor of mouth 15 years before and abscess had drained with expulsion of the calculus through the mouth. The perforation had healed forming a well established channel and now the patient had with recurrence of calculus in the duct. The degree of inflammation, atrophy and fibrosis of salivary glands are related to the duration of symptoms and the stone in-place period. The more the stone reside in-place, the more chronic manifestations may proceed .[12]

The conventional diagnostic methods for detecting obstructions in the salivary ductal system are mostly been based on clinical examination in combination with plain radiography (occlusal and panoramic radiographs) or sialography. Submandibular gland calculi have been reported to be radiopaque in 80% to 94.7% of cases.[5,9] However, diagnostic ultrasound and

Computerised Tomography can furnish the surgeon with much more detailed information on the exact stone localization, the number of stones, and the functional status of the glandular parenchyma.[13,14,15] As many as 90% of all stones larger than 2 mm can be detected as echodense spots on Ultrasonography. A Ultrasound and the Computerized Tomography helped us to come to a final diagnosis and to plan the treatment in our patient

Submandibular calculus is typically removed surgically via either an intraoral or an external approach.[16,17] The most appropriate mode of treatment depends primarily on the location of the calculus. An intraoral approach is often utilized when the calculus is located anterior to the lingual nerve and artery i.e in the distal part of the Wharton's duct (The part of the duct above the mylohyoid muscle and in the anterior 2/3 rd of floor of mouth). However, proximal and intraglandular calculus is difficult to remove transorally because of its position deep in the floor of mouth. Calculus impacted within the duct posterior to the first molar or even more proximally in the so-called comma area (where the duct turns inferiorly at the posterior border of the mylohyoid muscle) is difficult and may be hazardous to the lingual nerve.[18]
Furthermore, it is widely believed that proximal calculus causes permanent structural damage to the gland, thereby predisposing the gland to recurrent infection. Thus, proximal calculus is generally removed from the submandibular gland using a transcervical approach.[16]

In the treatment plan of our patient, in view of long standing disease, chronic sialoadenitis and the calculus in the proximal aspect of the Wharton's duct; we planned for submandibular gland excision along with removal of calculus by extraoral approach. As the calculus was impacted at the comma area (posterior curve of the Wharton's duct), it was difficult to remove the calculus and we had to remove it through the perforation in the floor of mouth. Thus we had

to go for a dual approach to manage this patient. The patient is still asymptamatic for 1 year after surgery.

CONCLUSION

Although giant sialoliths have been reported to be intraglandular or near the hilum of the gland, they have rarely been reported inside the duct. We report a giant sialolith impacted at the posterior curve (comma area) of the Wharton's duct with perforation of the floor of mouth. Submandibular stones are typically removed surgically via either an intraoral or an external approach. In our case, due to chronic sialoadenitis, we excised the gland by external approach and removed the calculus intraorally through the perforation. Thus a dual approach was needed in our case.

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Fig. 1: Perforation of the floor of mouth with calculus within.

Fig.2: Computerized Tomography showing the calculus .

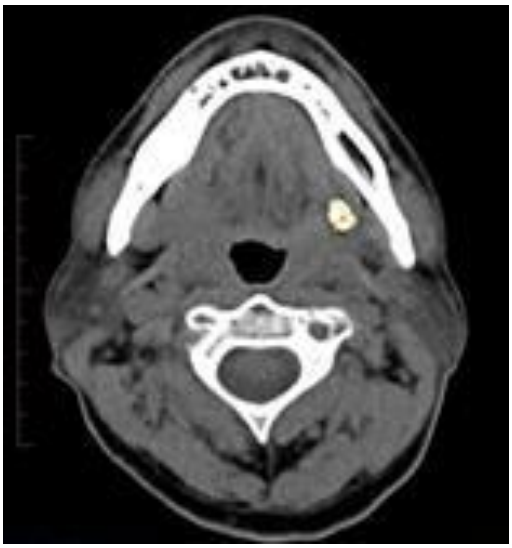

Fig. 3: Dilated Submandibular duct

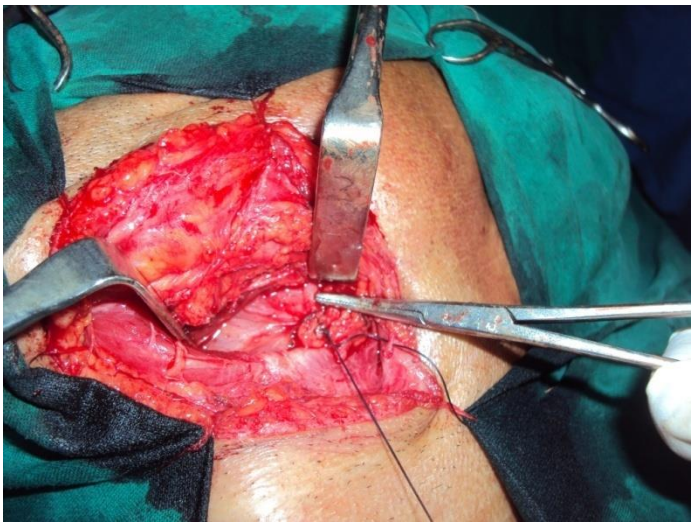

Fig. 4: Endoscopic picture of the calculus within the perforation

Fig. 5: Sialolith.