ABSTRACT

Context: Sialolith is a common salivary gland disorder occurring commonly in the duct submandibular salivary glands. Sialolith size < 10 mm is considered to be common and > 10 mm is considered to be unusual.

Aims: This case report highlights the occurrences of a large unusual sialolith in the parenchyma of the submandibular salivary gland.

Settings and designs: The representative sample was 10% formalin fixed hard tissue measuring 45 × 35 mm and 10 gm in weight; and soft tissue measuring 40 × 30 × 20 mm in size.

Materials and methods: The soft tissue received was sent for routine processing, sectioned, and stained with hematoxylin and eosin and was observed under optical compound microscope.

Conclusion: The clinician should carefully evaluate the painful and nonpainful lesions of salivary glands and consider the possibility of a sialolith even in the parenchyma apart from the duct. Timely intervention will eliminate or minimize the chances of complications to the patients.

Keywords: Sialodenuctomy, Sialolithiasis, Submandibular gland.

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INTRODUCTION

Sialoliths are calcareous concentrations and most salivary calculi occur in major and minor salivary glands. However, sialolith most commonly affects the submandibular glands or ducts (80–95%), whereas 5 to 20% occur in parotid glands with sublingual glands being rarely (1–7%) affected.1–4 Sialoliths occur commonly in the ducts and less commonly in the parenchyma of the glands as well. They are commonly seen as unilateral presentations and appear clinically as round or ovoid, rough or smooth, and yellow in color masses. Males are twice more affected than females.1,2 Etiological factor and pathogenesis of salivary calculi are unknown, but inflammation is considered to be an important contributing factor.5 Mostly, salivary calculi are clinically characterized by local pain, swelling, less saliva production, and dry mouth. Sometimes, when the salivary calculi become large enough, dysphagia, dysphonia, and lymphadenopathy of involved area are presented. The basic imaging methods are orthopantomogram and mandibular occlusal view; ultrasonography followed by computed tomography (CT) may assist the diagnostic protocol and thereby assist in favorable treatment. Differential diagnosis of other diseases, such as epidemic parotitis-mumps, obstructive sialadenitis, and benign or malignant salivary gland tumors might be verified to establish and eliminate them. The literature presents different sizes and shapes of sialoliths, but giant sialoliths of submandibular gland and duct are rarely reported (> 40 mm). The location and dimensions of sialolith will determine the management modalities, conservative or surgical procedure.

CASE REPORT

A 67-year-old male patient presented with complaint of pain, swelling in the posterior part on the left side of the floor of mouth with associated symptoms like dry mouth, bad breath, and inability to eat and drink during the last 3 to 4 months, and with no history of previous investigations and treatment. During the anamnesis, the patient gave us a past medical history of arrhythmia, high blood pressure, and bronchitis; the patient was under medication Aspirin (salicylate drug), Diazepam (benzodiazepine), Losartan (angiotensin 2 receptor blockers), and Verapamil (calcium channel blockers). The patient was a chronic smoker. A clinical examination with bimanual palpation revealed tenderness and palpable hard mass on the left submandibular region followed by regional lymphadenopathy. An occlusal radiograph was advised, which showed that the lesion was placed posteriorly, interpreted as half-oval...
sialolith in the lower left premolars and molars region. The orthopantomogram radiographic examination view revealed big triangle radiopaque mass, confirming the diagnosis of major submandibular sialolith.

Computerized tomography was performed in two projections: Posterior–anterior and right–left, to confirm the diagnosis of submandibular sialolith (Fig. 1). The patient underwent sialoadenectomy (surgical removal of the gland) via extraoral approach under general anesthesia with preservation of nearby structures. During surgical extrication, a small part of sialolith was broken; hence it was removed in two parts. The sialolith was present in the parenchyma of the submandibular gland, measuring $45 \times 35$ mm and 10 gm in weight (Fig. 2). Histopathological examination of the submandibular gland shows chronic submandibular sialadenitis. All symptoms of the patient disappeared after surgical procedure. He was discharged from the hospital on the 3rd postoperative day.

**DISCUSSION**

Salivary calculi are formation of calcific concretions either in the salivary duct or glands. They are usually small and measure 1 mm to less than 1 cm, and they rarely measure more than 1.5 cm. Sialoliths more than 3.5 cm are rare and known as giant sialolith, commonly found in the body of submandibular gland. According to Iqbal et al, the submandibular gland is most involved because of its specific anatomical location, viscous secretion, and long tortuous duct. Sialoliths are usually manifesting between the 3rd and 6th decade of life. It is believed that sialoliths annually increase by 1 to 1.5 mm per year. There are cases of large salivary calculi presented in the literature, with the largest one measuring around 70 mm and resembling an hen egg size. Iqbal et al reported a sialolith from the left submandibular duct of 55-year-old man measuring 35 mm in length and 30 mm circumference. Fowell and MacBean removed irregular elongated submandibular sialolith, weighing 3.0 g and measuring 41 mm in a 58-year-old male patient, with history of pain within weeks. Krishnappa removed multiple submandibular duct calculi of unusual size and shape in a 42-year-old male patient. Alkurt and Peker reported two unusual cases of large submandibular sialoliths in 45- and 65-year-old men, with lesion size in the 1st case $2.8 \times 0.8 \times 0.4$ and $3.1 \times 1.0 \times 0.7$ cm in the

**Fig. 1:** Preoperative CT scan shows large radiopaque mass localized within the left submandibular gland

**Fig. 2:** Enucleated salivary sialolith having unusual size and shape
2nd case. Singhal et al reported that patients who had multiple calculi in major salivary gland can be located in different anatomical location as well as salivary duct or body of salivary gland. The etiology and pathogenesis of major salivary calculi are still uncertain. These include infective, increased alkalinity pH, and calcium content of the saliva, inflammatory, infective, mucus, or desquamates cells, using certain medication that affects production of saliva. The most common symptoms noted with sialolith are recurrent pain, swelling of the involved salivary gland, and pus secretion. There are various clinical and X-ray imaging methods available like occlusal views, panoramic views, computerized tomography, and ultrasonography to detect and visualize the sialolith.

Parkar et al showed that sialoliths appear as radiopaque masses in X-ray imaging as reported in 80 to 94.7% of cases. Standard mandibular occlusal radiograph view is considered to be the best for visualizing radiopaque stones. Ultrasonography is the preferred investigation of choice in cases of clinical suspicion of sialolithiasis as both the sialoliths and the gland affected are visualized. Occult sialoliths on radiographs can show up on ultrasonography. Sialoliths appear as strongly hyperechoic lines or points with distal acoustic shadowing. However, sialoliths < 2 mm may not manifest on the ultrasound. In acute obstructive cases, the gland appears to be enlarged, and excretory ducts proximal to the sialolith may be visibly dilated. Examination is best performed with small high-frequency intraoral probes as against CT which exposes to radiation. The treatment depends on size and location of sialolith, and treatment options for salivary calculus are generally dictated by the size and position of the stone. Sialoliths which are smaller in size can be milked out through the ductal orifice using bimanual palpation. For larger sialoliths or sialoliths situated in the proximal duct, piezoelectric extracorporal shock wave lithotripsy or surgical removal of the stone or gland may be required. Sialoendoscopy is a new way and minimally invasive technique for treating obstructions of the ductal system and can be used with operation in large salivary stones. Intraglandular sialoliths located in the body of major salivary glands require sialodenedectomy or partial sialodenedectomy. In our case, the giant submandibular sialolith was removed with open extraoral surgery under general anesthesia.

CONCLUSION

Clinical investigation followed by diagnostic methods is especially important to determine the existence of salivary calculi. Giant salivary gland calculi might be asymptomatic or followed by local pain and swelling in the affected area. The location and dimensions of sialolith determine the management modalities, but surgical treatment of giant salivary gland calculi is necessary.

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CONSENT

Written informed consent was obtained for publication of this case report and the accompanying images. A copy of the written consent is available for review by the editor of this journal.

ETHICAL APPROVAL

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. Informed consent was obtained from all individual participants included in the study.

REFERENCES