Giant Submandibular Sialolith presenting with Sialocutaneous and Sialo-Oral Fistula: A Case Report and Review of Literature

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ABSTRACT
Sialolithiasis is a common disease of the salivary glands and a major cause of salivary gland dysfunction. It commonly affects middle-aged and has male predominance. Submandibular gland or its duct is most commonly affected. The size of salivary calculi may vary from less than 1 mm to a few cm in the largest diameter. Salivary stones that exceed 15 mm in any dimension are classified as giant. Association of sialocutaneous or sialo-oral fistula with salivary stones is considered rare. Long-standing stones with chronic inflammatory process may lead to fistula formation.

Keywords: Giant sialolith, Sialo-oral, Sialocutaneous fistula, Oxalate.

INTRODUCTION
Sialolithiasis is the most common disease of salivary glands which accounts for more than 50% of the salivary gland diseases. Its estimated frequency is 1.2% in the adult population with male patients affected as twice as much as female patients. Most salivary calculi (80-95%) occur in the submandibular gland, whereas 5 to 20% are found in the parotid gland. The sublingual gland and minor salivary glands are rarely affected. Salivary stones are mainly composed of calcium and phosphate with small amounts of carbonates in the form of hydroxyapatite as well as small amounts of magnesium, potassium and ammonia. Submandibular stones are composed of 82% inorganic and 18% organic material, whereas parotid stones are composed of 49% inorganic and 51% organic. Presence of oxalate is uncommon.

We report a case of giant sialolith measuring 26 × 21 × 20 mm in dimension and weighing 9.5039 gm containing oxalate presenting with sialocutaneous and sialo-oral fistula.

CASE REPORT
A 52-year-old man reported complains of pain and recurrent swelling and pus discharge from the right submandibular region for duration of 2 years. The swelling was there for the past 25 years. There were episodes of pain in the same region for the past 3 years. The pain become aggravated during eating and was relieved by rest. Swelling was gradual in onset, progressing to the present size.

On extraoral examination, the patient showed diffused swelling with scars over the right submandibular region measuring 8 × 7 cm (Fig. 1). There was a fistula opening in the inferior aspect of the swelling from which pus was discharged on manual pressure. The swelling was warm, fluctuant and tender. Intraoral examination reveals another fistula opening in the right posterior floor of the mouth (Fig. 2). The surrounding mucosa appears normal in color and texture. On palpation, a firm mass was felt at a deeper location.

Bacterial culture and antibiotic sensitivity test were done. Culture revealed growth of mixed organisms including Staphylococcus, Escherichia coli after 48 hours of incubation. Short course antibiotics were given and the patient was symptomatically relieved.

Radiographic examination with a lateral oblique of mandible revealed a well-defined radiopacity in right submandibular region measuring 8 × 7 cm (Fig. 1). There was a fistula opening in the inferior aspect of the swelling from which pus was discharged on manual pressure. The swelling was warm, fluctuant and tender. Intraoral examination reveals another fistula opening in the right posterior floor of the mouth (Fig. 2). The surrounding mucosa appears normal in color and texture. On palpation, a firm mass was felt at a deeper location.

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Radiographic examination with a lateral oblique of mandible revealed a well-defined radiopacity in right submandibular region (Fig. 3). Computed tomography revealed a homogenous hyperdense lesion measuring 1.8 × 2.4 cm at the hilum of right submandibular gland. Three-dimensional computed tomography (3D-CT) reveals the 3D-picture of the lesion in relation to the craniofacial structures (Fig. 4). Magnetic resonance imaging revealed a well-defined single void lesion measuring 22 × 22 mm in the right submandibular space. It also revealed pressure effect...
over adjacent right posterior tongue and tonsillar fossa. The carotid and jugular vessels were displaced posteriorly (Fig. 5).
The sialolith was removed extraorally under general anesthesia along with the associated salivary gland. No smaller calculi were associated with the sialolith. The fistulas were surgically closed. The sialolith recovered measured $26 \times 21 \times 20$ mm and weight 9.5039 gm (Fig. 6). It is yellowish brown in color and the surface is having a corrugated texture. In cross section, concentric lamellations were seen. Biochemical analysis reveals it is mainly composed of calcium, phosphate and oxalate. After 7 months follow-up, the patient was asymptomatic and salivation was satisfactory. Consent was taken from the patient for publication of this case.

DISCUSSION
Sialolithiasis is a common disease of the salivary glands and a major cause of salivary gland dysfunction. There are three requirements for the formation of a sialolith: Salivary stagnation, a nidus and precipitation of salivary salts. Infection, inflammation, salivary stagnation, physical trauma, introduction of foreign bodies and the presence of desquamated epithelial cells seem to be the initial events for the formation of a nidus that later will be the site for the precipitation of mineral salts contained in the salivary secretion. Several factors seem to be involved in the development and growth of salivary calculi in the submandibular salivary gland tissue, such as wider diameter of duct, salivary flow against gravity, alkaline secretion and higher calcium and phosphate content.

Salivary stones that exceed 15 mm in any dimension are classified as giant. Commonly, sialoliths measure from 1 mm to less than 1 cm. They rarely measure more than 1.5 cm. Giant sialoliths are rare. Bodner found only 14 well-documented cases in his review published in 2002 and to the best of our knowledge, this case is the only case of giant sialolith presenting with both sialocutaneous and sialo-oral fistula after Paul D, Chauhan MS.

The ability of a calculus to grow and become a giant sialolith depends on the reaction of the affected duct. If the duct adjacent to the sialolith is able to dilate, allowing nearly normal secretion of saliva around the stone, it might be asymptomatic for a long period and eventually a giant calculus will be created.

Fistula formation either oral or cutaneous from a salivary calculus is rare. A fistula develops most likely when bacteria...
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JOURNAL OF INDIAN ACADEMY OF ORAL MEDICINE AND RADIOLOGY

Fig. 5: MRI showing the sialolith (cal), its effect on surrounding structures, swelling with pus collection and lymph node

Fig. 6: Gross specimen of the sialolith

set up an acute exacerbation in the stagnating and retained saliva located behind the stone. The inflammatory debris obstructs the residual narrowed duct lumen, further exacerbating the inflammation. The inflammatory process around a large stone may lead to tissue breakdown leading to intraoral\(^1\) or extraoral fistula formation.

Oxalate in salivary stones is seldom found. Consumption of food with high oxalate content, high ambient temperature and dehydration with poor oral hygiene may precipitate oxalate in salivary stones. In the body, oxalic acid can combine with metallic ions, such as Ca\(^{2+}\) and Mg\(^{2+}\) to form crystals of the corresponding oxalates. Mg oxalate is 567 times more soluble than Ca oxalate, so that Ca oxalate crystals are more likely to precipitate out when Mg levels are low and Ca and oxalate levels are high.

The standard occlusal radiograph is the most commonly used to view submandibular sialolith. But, it is unsuitable for giant sialoliths which occur frequently in the posterior portion of Wharton’s duct and the glandular parenchyma. In contrast to the small-sized calculi, 20 to 30% of which are radiolucent, giant sialoliths are mostly radiopaque and are easily depicted on standard radiographs, probably because their lithogenesis is long enough for calcification to be completed.\(^4\) Calcifications, however, can be visualized very early by use of a computed tomography scan, which is sensitive even to stones that are radiolucent on standard radiographs.\(^1\) Sialography is particularly useful in obstructions of duct by radiolucent stones and detection of strictures in the course of duct. Magnetic resonance imaging is useful in visualizing the effects of the stone on surrounding tissue but cost limits its use.

The management of giant sialolith has always been a therapeutic challenge. The giant sialolith should be removed in a minimally invasive manner via a transoral sialolithotomy to avoid the morbidity associated with sialadenectomy.\(^4\) Submandibular gland removal is indicated only when there is a stone of substantial mass within the gland itself that is not surgically accessible intraorally and when there are small stones present in the vertical portion of Wharton’s duct from the comma area of the hilum.\(^1\) Recent advances in optical technology have led to the development of sialoendoscopy, a new diagnostic means of directly visualizing intraductal stones which allows complete exploration of duct including shockwave lithotripsy, sialoendoscopy, interventional radiology, endoscopically video-assisted transoral and cervical surgical retrieval of stones and botulinum toxin therapy.\(^1\) Recent progress in endoscopic surgery has facilitated the nonsurgical removal of sialoliths from the duct of the submandibular gland. However, the limitations of endoscopic techniques are adhesion of stones to the ductal walls, ducts that are too narrow, strictures, ducts with sharp angles, giant sialoliths and intraparenchymal stones.\(^5\)\(^6\) Newer treatments modalities, such as extracorporeal shock wave lithotripsy (ESWL), and more recently, the use of endoscopic intracorporeal shock wave lithotripsy (EISWL),\(^7\) endoscopy using wire baskets and balloons\(^8\) are effective alternatives to conventional surgical excision for smaller sialoliths. However, for giant sialoliths, transoral sialolithotomy with sialodochoplasty or sialadenectomy remains the mainstay of management.\(^1\)

REFERENCES
