

Giant submandibular gland duct sialolith mimicking an impacted canine tooth

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ABSTRACT

Sialolithiasis is the most common disease affecting the salivary glands and accounts for 80% of salivary gland disorders. Chronic sialolithiasis promotes stone formation. Size of the salivary stones may range from 0.1 mm to 30 mm or be even bigger. Those salivary stones, the size of which exceeds 15 mm in any one dimension or 1 g in weight are classified as giant sialoliths. Giant sialoliths of the submandibular gland duct are rarely reported. Here, we report a case of a giant sialolith of the submandibular gland duct mimicking an impacted mandibular canine tooth on routine radiographic examination and its surgical management through an intraoral approach.

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INTRODUCTION

Sialolithiasis is the most common disease of the salivary glands and is a major cause of salivary gland dysfunction.^[1] Its estimated frequency is 1.2% in the adult population with a slight male predominance. More than 80% of the salivary gland calculi appear in the submandibular gland, more frequently in the excretory duct as compared to the gland parenchyma.^[2] According to one of the etiopathogenetic theories, the formation of salivary gland calculi results from a deposition of calcium salts around a core made up of desquamated epithelial cells, foreign bodies, or mucus. Their main inorganic components are phosphates and calcium carbonates.^[3] Usually sialoliths measure from 1 mm to less than 1 cm. They rarely measure more than 1.5 cm. In the literature, giant sialoliths are classified as those exceeding 15 mm in any one dimension or more than 1 gm in weight.^[4] We report a case of a giant sialolith of the submandibular gland duct bearing an unusual similarity to

the canine tooth and mimicking an impacted canine tooth on routine radiographic examination.

CASE REPORT

A 45-year-old male walked into our center with a history of pain in the left submandibular region for the past 6 months. The pain was dull, intermittent, and associated with meals, and the swelling enlarged in that area during and after meals.

On extraoral examination, there was a localized swelling in the left submandibular region measuring approximately 4 cm × 3 cm. The swelling was soft and tender on palpation with normal overlying skin. Intraoral examination revealed an edematous floor of the mouth on the left side. Intraorally, on bimanual palpation on the floor of mouth, a hard mass measuring approximately 3 cm × 2 cm was felt in the premolar-molar region. Milking of the left submandibular gland yielded very less saliva but pus discharge.

Orthopantomogram (OPG) [Figure 1] and lateral oblique view of the mandible [Figure 2] revealed a calcified mass, exactly mimicking an impacted canine tooth in the premolar-molar region. In the occlusal view x-ray of the mandible [Figure 3], this calcified structure was

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seen bearing a close resemblance to a canine tooth but the radiopacity was homogenous. On the basis of clinical and radiological findings, a diagnosis of the sialolith of the left submandibular salivary gland duct was done.

Considering the large size of the sialolith (giant), transoral sialolithotomy was done under local anesthesia after giving a proper antibiotic cover. Upward and medial pressure was applied at the submandibular area for stabilizing the floor of the mouth and for proper localization of the sialolith. Intraoral mucosal incision was placed directly over the sialolith. After careful mucosal dissection, the submandibular duct was identified and incised over the sialolith. The giant sialolith was mobilized and gently picked with tissue forceps and the wound was closed only at the mucosal layer. In the subsequent follow-up at 6 months, there was complete remission of the symptoms with effective salivary drainage and normal function of the submandibular gland. The sialolith removed [Figure 4] measured 31 mm in length and 16 mm in circumference, weighing 1.250 g.

On the basis of clinical, radiological and histopathological findings, the diagnosis of a giant sialolith of the

submandibular salivary gland duct was confirmed. Stone analysis report revealed 100% carbonate apatite, as done by the fourier transform infrared spectroscopy (FTIR) technique.

DISCUSSION

Salivary calculi are usually small and measure from 1 mm to 1 cm. They rarely measure more than 1.5 cm and the mean size reported is between 6 mm and 9 mm². Giant sialoliths are classified as those exceeding 15 mm in any one dimension or 1 g in weight.^[4] In this report, clinical and radiological features of a giant sialolith, with dimensions of 31 mm × 16 mm and a weight of 1.250 g were present. It was located in the Wharton duct opposing the mandibular left premolar and the first molar region. The location and shape of the calcified mass exactly mimicked an impacted canine tooth. Large sialoliths may perforate the floor of the mouth by ulcerating the duct or may result in orocutaneous fistula by causing a suppurative infection.^[5] The largest sialolith reported in the literature was 70 mm in the length of the Wharton duct and was described as having a “hen’s egg” size.^[2]

Traditional and recent etiopathogenic factors include a reduced salivary flow rate, a change in pH, dehydration,



Figure 1: OPG showing sialolith mimicking an impacted canine tooth



Figure 2: Lateral oblique view of mandible showing giant sialolith



Figure 3: Occlusal view of mandible showing giant sialolith



Figure 4: Sialolith removed

duct anomalies, and retrograde migration of food, bacteria, or foreign bodies from the oral cavity favoring stone formation.^[6] The exact mechanism of lithogenesis is unclear; however, the presence of microcalculi in 80% of the normal submandibular gland and in only 10% of the normal parotid gland may correspond well to the overt incidence of calculi in these two glands.^[4] The ability of a calculus to grow and become a giant sialolith depends mainly on the reaction of the affected duct. The growing stone causes an increasing obstruction of salivary secretion, which leads to swelling, pain, and infection of the gland and eventually medical intervention. However, if the duct adjacent to the sialolith is able to dilate, allowing nearly normal secretion of the saliva around the stone, it might be asymptomatic for a long period and eventually a giant calculus will be created. As the stone increases in size beyond the ability of the duct to dilate, a sialo-oral fistula will develop and the sialolith will partially protrude into the oral cavity.^[4] It is believed that a calculus may enlarge at the rate of approximately 1-1.5 mm/year.^[5]

Sialolithiasis typically causes pain and swelling of the involved salivary gland by obstructing the food related surge of salivary secretions.^[7] Calculi may cause stasis of saliva, leading to bacterial ascent into the parenchyma of the gland, therefore, causing infection, pain, and swelling of the gland.^[4] In this case report, the patient gave a history of preprandial and postprandial submandibular swelling since the last 6 months explaining the infection and partial obstruction of the duct. Long-term obstruction in the absence of infection can lead to atrophy of the gland with resultant lack of secretory function and ultimately fibrosis.^[5]

Ultrasonography (USG) is a method of choice in salivary stone diagnostics. Its sensitivity in calculi detection amounts to 94%, specificity 100%, and accuracy 96%. USG allows detection of nonopaque calculi with sensitivity of 80-96%.^[8] But small stones <2 mm may not produce any acoustic shadow leading to diagnostic mistakes, especially small stones in the intraparenchymal ducts with no duct distension. Sialendoscopy allows direct visualization of the salivary duct lumen, i.e., visualization of calculi, mucosal plugs, foreign body, and polyps. It is mainly used in the treatment of inflammatory condition of the salivary glands and obstruction of the salivary ducts.^[9] Sialendoscopy enables the surgeon to examine the salivary duct from inside as well as combine the therapeutic procedures for better results.

Restoration of the normal salivary secretion is the treatment objective of giant sialolith removal as the standard sized stones. The treatment options available for stone removal are transoral sialolithotomy, interventional sialoendoscopy, extracorporeal shock

wave lithotripsy (ESWL), and resection of the gland. ESWL is an effective, noninvasive alternative approach in the surgical removal of salivary stones, reserving surgery for recurrent or complicating salivary lithiasis. The principle of ESWL is to reduce the size of calculi to small fragments, the diameter of which does not block the flow of saliva and can be washed away by natural flow. Anderetta *et al.*^[10] discussed the limitations of ESWL in the removal of stone with a diameter of >10 mm, as it is difficult to reduce the fragments and also in cases of acute inflammation. Minimal invasive approach should be the treatment of choice so as to avoid morbidity associated with the radical ones.^[4] If the stone can be palpated, it is best to remove it transorally. In this case report, the giant sialolith was easily palpated on the floor of the oral cavity. The cardinal rule while performing the sialolithotomy is to first isolate the duct, stabilize it, and then provide a longitudinal incision onto the duct over the stone to retrieve it. Direct cutdown technique, where the incision is taken directly to the surface of the stone without primary isolation of the duct, may lead to stenosis of the duct.^[5]

After elimination of the obstruction, the apparent resiliency of the submandibular gland results in no adverse symptoms. Submandibular gland removal is only indicated when there is substantial mass within the gland itself that is not surgically accessible intraorally and when there are small stones present in the vertical portion of the Wharton duct from the comma area to the hilum. Surgical removal of the gland is also indicated in a situation where opening of the duct surgically created causes recurrent infection of the gland due to ingress of oral fluids.^[5]

CONCLUSION

The treatment objective of a giant sialolith, as for its standard sized stones, is restoration of normal salivary secretion.^[7] The sialolith should be removed with the minimal invasive method via a transoral sialolithotomy to avoid the morbidity associated with sialoadenectomy. Intraglandular sialoliths necessitate sialoadenectomy.^[4]

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