Case Report

Mealtime syndrome: An Enigma to Resolve

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Abstract
Mealtime syndrome also known as sialolithiasis is the most common disease affecting the submandibular glands. Symptoms of intermittent swelling in the salivary glandular region and pain during mealtime due to increased intraglandular pressure help in the diagnosis. Radiographically calculi may occur as radiopaque structure, and sometimes may not be visible due to poor calcification. Salivary calculi, most commonly occur in the submandibular glands (up to 90% of cases) and parotid glands (5 to 20%). The sublingual gland and minor salivary glands are rarely affected. The right and left side glands are equally affected, and bilaterally arising stones being rare occur in less than 3% of cases. This article reports a case of submandibular salivary calculi in 22-year-old male patient with recurrent symptoms of pain and swelling during mealtime. The salivary calculi were removed surgically by intraoral approach under general anesthesia.
Keywords: Sialography, Sialolith, Submandibular salivary gland.

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Introduction

In humans, three pairs of major salivary glands and numerous minor salivary glands secrete saliva. Variety of pathological processes like inflammatory, infectious, obstructive, autoimmune, systemic and neoplastic disorders affects the salivary glands. Sialolithiasis is the second most common disease of the salivary glands after mumps. It is also the most common disease of submandibular glands in middle-aged adults. Sialolithiasis is estimated to affect 12 in 1000 of the adult population with males being affected almost twice as much as females,[1] and Children are very rarely affected.[2] Calculi size may vary from less than 10 mm to abnormally large up to 15 mm in size.[3]

Case Report

A 22-year-old male patient reported with a complaint of retained deciduous tooth in the upper left front region of jaw. He gave a history of recurrent intermittent swelling on his left lower third of face while having

food, which relieves by itself since eight years. There was no facial asymmetry or deformity observed on extra oral examination. Intraoral examination showed retained upper left deciduous canine and clinically missing maxillary permanent left canine. A hard mass was palpated in the floor of the mouth during bimanual palpation of left submandibular gland (Figure 1).

Figure 1: Clinical photograph of left submandibular region where calculi was palpated by bimanual palpation.

Based on history and clinical examination, a provisional diagnosis of impacted upper permanent left canine and left submandibular sialolith were considered. Radiological examination of periapical radiograph showed retained maxillary left deciduous canine and impacted maxillary left permanent canine. Mandibular cross sectional occlusal radiograph showed, a well-defined radiopacity measuring about 1× 0.5cm in size at the left side floor of the mouth (Figure 2). Panoramic view showed retained maxillary left deciduous canine with impacted maxillary left permanent canine, bilaterally missing mandibular first and second molars.

A well-defined radiopacity measuring about 1.5×1cm superimposed over the roots of mandibular left premolars was observed (Figure 3). Radiographic diagnosis of left submandibular calculi with a differential diagnosis of enostosis, sclerosing ostetitis and calcified lymph node were considered.

Figure 2: Mandibular cross sectional occlusal radiograph showing calculi in left submandibular duct.

Figure 3: Panoramic radiograph showing impacted maxillary left permanent canine with calculi superimposed on left mandibular premolar region.

Sialography was carried out to know the amount of destruction to the gland caused by calculi, using urograffin (1 ml of aqueous iodine based contrast media) by simple injection technique. Preoperative, filling phase and emptying phase panoramic radiographs were captured. The panoramic radiograph captured during filling phase showed non-uniform and incomplete filling of the radiopaque contrast media within the submandibular duct and the gland. There was severe dilatation of duct posteriorly with numerous loculations. The peripheral ducts of gland were dilated and incompletely filled, giving a typical appearance of bush in winter appearance (Figure 4).

Figure 4: Filling phase panoramic radiograph showing incomplete filling of gland and duct with dilatation of duct posterior to calculi.

Panoramic radiograph captured at emptying phase after five minutes showed retention of contrast media in the duct and gland suggestive diminished glandular function.

Figure 5: Photograph showing surgical removal of calculi by intraoral approach.

Figure 6: Extracted upper deciduous and permanent left canines with calculi.

Sialolith was removed surgically by intraoral approach, and impacted maxillary left permanent and deciduous canines were extracted under general anesthesia (Figures 5 and 6).

Discussion

Salivary calculi may occur within the intraglandular ductal tributaries or within the main ducts and are usually unilateral, and do not cause dry mouth. Multiple calculi in the submandibular glands are rare,[4] as is simultaneous lithiasis in more than one salivary gland.[5]

The exact aetiology and pathogenesis of salivary calculi is unknown. For calculi formation, it is likely that intermittent stasis produces a change in the mucoid element of saliva, which forms a gel. This gel produces the framework for deposition of salts and organic substances creating a stone.[6] Traditional theories suggest that the formation occurs in two phases: a central core and a layered periphery.[7] The central core is formed by the precipitation of salts, which are bound by certain organic substances. The second phase consists of the layered deposition of organic and non-organic material. Submandibular stones are thought to form around a nidus of mucous,[8] whereas parotid stones are thought to form most often around a nidus of inflammatory cells or a foreign body.[8-10] Submandibular sialolithiasis is more common as its saliva is (i) more alkaline, (ii) has an increased concentration of calcium and phosphate, and (iii) has a higher mucous content. According to Gorlin & Goldman, 92% of salivary calculi occur in the submandibular gland, 6% in the parotid gland, and 2% in both the sublingual and minor salivary glands. Rauch determined the following locations of submandibular stones, near the ostium (30%), middle third of the duct (20%), right angle bend of Wharton duct (35%), and proximal to right angle bend and hilum of gland (15%).

Sialolith may cause pain and swelling during salivary stimulation and is intensified at mealtimes, so called “mealtime syndrome”. Calculi may cause stasis of saliva, leading to bacterial ascent into the parenchyma of the

gland,[8] and therefore infection, pain and swelling of the gland. When calculi are present in the gland itself, the symptoms may be relatively minor, whereas ductal sialoliths usually have a more precipitous presentation. In our case the calculi was present in the duct. Some calculi may be asymptomatic until the stone passes forward and can be palpated in the duct or seen at the duct orifice. Long-term obstruction in the absence of infection can lead to atrophy of the gland with resultant lack of secretory function and ultimately fibrosis. Diagnosis of submandibular stone is often straightforward from a through history and examination. Plain radiographs such as occlusal view, orthopantogram , lateral oblique view of mandible can detect opaque stones (80-95% of sialoliths). Ultrasonography, Computed tomography, MRI and sialography are the other tools of detection of calculi. There are various methods available for the management of salivary stones, depending on the gland affected and location of the calculi. Surgical removal by intraoral or extraoral approach, sialoendoscopy and extra and intracorporeal lithotripsy are few of them. In our case, surgical removal of the calculi was carried out through intraoral approach under general anaesthesia.

Conclusion

Intermittent swelling and pain are often associated with sialolith. They occur in mealtime due to elevated intraglandular pressure resulting from an increased salivary secretion in the obstructed gland or duct. Careful history and thorough clinical and radiographic examination is important in the diagnosis of sialolithiasis. Early diagnosis and prompt treatment helps reducing patient discomfort and complications.

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