

Giant sialoliths of submandibular gland duct: Report of two cases with unusual shape

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Abstract

Giant sialoliths are classified as those exceeding 15 mm in any one dimension. Although, large sialoliths have been described in the body of salivary glands, they are rarely found in the salivary ducts, particularly when the patients have no painful symptoms. Sialolithiasis is one of the most common diseases of the salivary glands in middle-aged patients and approximately 80% of all reported cases of sialoliths occur in the submandibular gland. Here, we report two cases of giant sialolith of submandibular gland duct. Interestingly, sialolith found in one of our cases had an unusual similarity with the canine tooth and mimicking it in both size and shape. In both cases the calculus was removed surgically via intraoral approach. No recurrence was seen in any of the cases on follow-up.

Keywords: Giant, sialolith, submandibular duct, unusual shape

Introduction

Sialolithiasis is one of the most common diseases of the salivary glands in middle-aged patients, and it is estimated that 12 of every 1000 adult patients are affected with sialolithiasis.^[1] Approximately 80% of all reported cases of sialoliths occur in the submandibular gland.^[2] In literature, giant sialoliths are classified as those exceeding 15 mm in any one dimension or 1 g in weight.^[3,4] Giant sialolith of submandibular duct are rarely reported. Here, we report two cases of giant sialolith of submandibular gland duct. Interestingly, sialolith found in one of our cases had an unusual similarity with the canine tooth.

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Case Reports

Case 1

A 48-year-old male patient reported with the complaint of intermittent, dull aching pain, and swelling in left submandibular area for the last 3 months. These symptoms occurred 6-7 times per week, during meals. The patient noted that sour foods were more likely to produce symptoms than were other types of food.

On examination, he was found to have a firm mass of approximately 3 cm × 1 cm on the floor of his mouth, along the course of the left submandibular duct. Salivary flow was negligible from left submandibular duct orifice. The left submandibular gland was tender and enlarged measuring approximately 3 cm × 2 cm. An occlusal radiograph showed a large radiopaque calculus in the floor of mouth. Left submandibular sialogram showed obstruction of the left submandibular duct by a radiopaque calculus approximately 3 cm proximal to the orifice of the duct [Figure 1a]. The calculus was removed via intraoral approach under local anesthesia [Figure 1b] and transposition of ductal opening was done. The shape of calculus was unusually similar to the canine tooth [Figure 1c]. The thicker and thinner portions of calculus looks like the crown and root of canine tooth. The length and width was measured to be 28 mm and 11 mm, respectively, which are close to the dimensions of canine tooth. In subsequent clinical follow-up of 2 years, the complete remission of the symptoms, the effectiveness of the salivary drainage, and the normal functioning of the submandibular gland were observed.

Case 2

A 45-year-old female reported with complaints of swelling in mouth associated with pain over left side of face during intake of food. On intraoral examination, hard mass of

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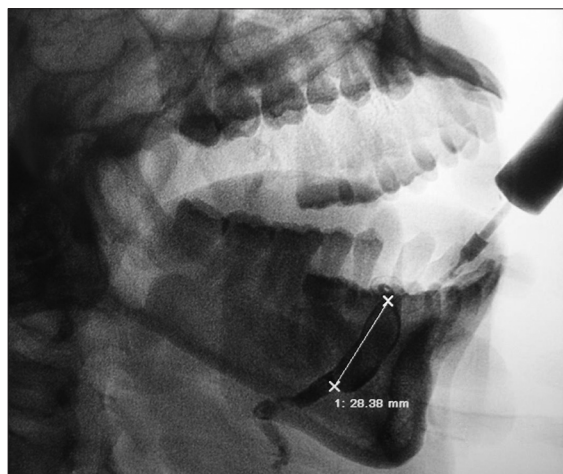


Figure 1a: Sialogram showing calculus with length of 28 mm in submandibular duct of case 1

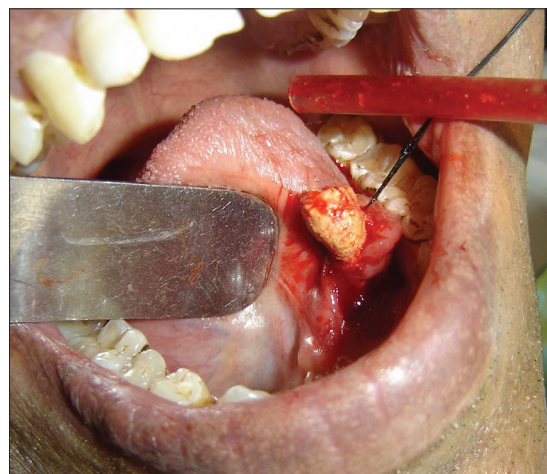


Figure 1b: Intraoral photograph of case 1 showing surgical removal of calculus from the submandibular duct



Figure 1c: Photograph showing specimen of the calculus removed from case 1 having unusual similarity with the canine tooth in size and shape

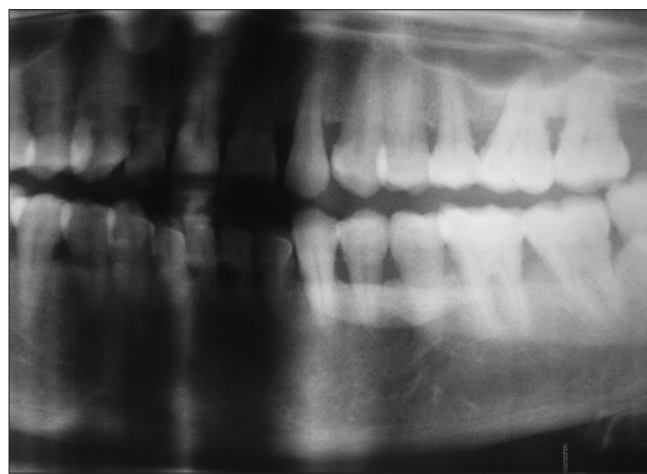


Figure 2a: Orthopantomogram of case 2 showing radiopaque calculus on left side

approximately 2 cm × 1 cm is palpated in the floor mouth with mild tenderness over left submandibular region. On orthopantomogram and occlusal radiograph large radiopaque mass is detected of size approximately 20 mm × 6 mm located within the right Wharton's duct [Figures 2a and b]. The calculus was removed intraorally and duct opening was transposed into the floor of mouth under local anesthesia. The unusually large size of calculus was measured to be 19 mm × 5 mm [Figure 2c]. Patient was followed-up for 1 year and is asymptomatic without any recurrence. The submandibular salivary gland regained its normal function and effective salivary drainage in the follow-ups.

Discussion

Sialolithiasis is often accompanied by recurrent bouts of pain and swelling in the involved salivary gland. The pain and swelling are usually associated with eating. Because salivary duct calculi are usually symptomatic, patients often seek

medical attention long before their calculi become large. Judging from the size of the unusual calculus described in this report, we suspect that it must have been present for years. Although, case 2 patient reported pain over left side of face during intake of food; adding to our interest, case 1 patient reported that he had only intermittent, dull, aching pain, and swelling in the left submandibular area for the last 3 months. His symptoms occurred 6-7 times per week, during meals. In our opinion, the nature of symptoms could be explained by the fact that the obstruction to the flow of saliva caused by the large calculus was never complete and that some saliva managed to seep through the cracks or crevices in the wall of calculus. Few cases of giant sialolith with unusual shape have been reported in the English literature but with such an unusual shape (case 1) is probably the first case in the literature.^[5] Interestingly, this unusual similarity with the canine tooth is found to be incidental and supposed to have no significance with the management. However, this unusual shape requires reporting to aware with the possibility of such variant shapes of sialoliths in submandibular duct.

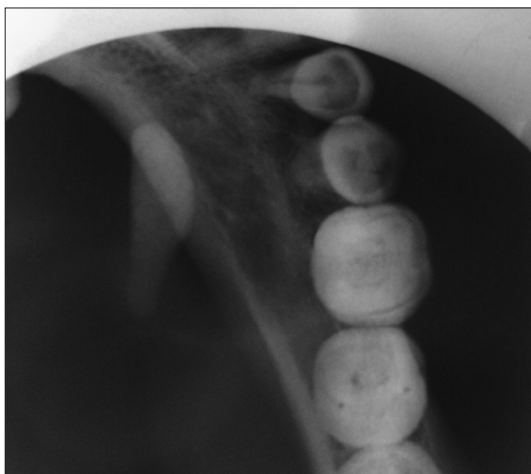


Figure 2b: Occlusal radiograph of case 2 showing radiopaque calculus on left side of floor of mouth

It is believed that salivary calculi develop as a result of deposition of mineral salts around a nidus of bacteria, mucus, or desquamated cells.^[6] Salivary stagnation, increased alkalinity of the saliva, increased calcium content of the saliva, infection or inflammation of the salivary duct or gland, and physical trauma to the salivary duct or gland may predispose to calculus formation. The submandibular gland is susceptible because its saliva is far more alkaline, and has a greater concentration of calcium and phosphate. It is known that the submandibular gland has a higher mucus content than the saliva of the parotid and sublingual glands.^[7] Although, large sialoliths have occasionally been reported in the salivary gland, they have rarely been reported in the salivary ducts. Rust and Messerly removed a sialolith 51 mm long that occupied the entire length of Stensen's duct in a 66-year-old man.^[7,8]

Imaging studies are very useful for diagnosing sialolith. To visualize the radiopaque stones in the submandibular duct, the best view is a standard mandibular occlusal radiograph. Other traditional diagnostic methods include sialography, ultrasound, computed tomography, and scintigraphy for sialoliths.^[9]

The primary objective of the treatment in a case of standard sized stones in the case of giant sialolith is the restoration of normal salivary secretions. Different treatment options may be selected according to the size and location of the sialolith. Small stones often may be "milked out" through the ductal orifice using bimanual palpation. If the stone is too large



Figure 2c: Photograph showing specimen of calculus removed from case 2 with ruler

or located in the proximal duct, piezoelectric extracorporeal shock wave lithotripsy or surgical removal of the stone or gland may be required.^[10]

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