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Multiple Giant Sialolith in an Unusual Location: Report of a Case

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ABSTRACT

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INTRODUCTION

Sialoliths are calcium-rich crystallized minerals that can occur in any of the salivary glands. It occurs in 1.2% in the adult population and has a strong male predilection, with a peak incidence between ages 30 and 50[1]. They may be single or multiple but a majority of cases occur as a single entity with no preference towards the left or right side[2]. Multiple stone formation occurs in approximately 25% of cases[3]. More than 80% of sialoliths are found within the submandibular gland (80%-90%), to a lesser extent within the parotid gland (5%-20%), and rarely in the sublingual and minor salivary glands[2]. This higher incidence in the submandibular gland is due to the anatomy and course of Wharton's duct as well as the salivary composition. Anatomically, the duct is narrower in diameter and follows a more tortuous course. The duct makes a sharp turn in 2 areas; the first is around the posterior edge of the mylohyoid muscle and the second occurs before the duct empties into the oral cavity. These particular areas are therefore more prone to salivary stone formation. Other contributing factor is the salivary composition. The saliva from the submandibular gland is twice as viscous as saliva from the parotid gland and also has a higher pH and calcium content[4]. Systemic factors which causes salivary calculi formation includes dehydration of the patient and the antisialogogues effects of certain medications (antihistamines, antipsychotic medications)[5]. Classical manifestations of sialolithiasis include pain and swelling, especially whenever the salivary flow is stimulated, as during the pre-meal period. The sialolith obstructs the salivary ducts hindering the normal flow of salivary secretions, thus aggravating the symptoms.

Here we present a case with multiple giant sialoliths in the left submandibular duct in an unusual location and briefs on the clinical examination, radiological investigation and the surgical excision.

CASE REPORT

A 64-year-old male patient reported to Department of Oral and Maxillofacial Surgery with chief complains of pain and swelling in the floor of the mouth for the past 9 years. The swelling was smaller in size at the beginning which increased over the period and attained the present size. Pain and swelling was noted to be increased at meal time. On clinical examination, the swelling was hard in consistency and tenderness present on the left submandibular gland on palpation. Decreased salivation were noted. The swelling was extending from the lingual aspect of 36, posteriorly up to the 38 region. The patient was afebrile and his vital signs were normal. On the basis of the history and clinical findings, a clinical diagnosis of left submandibular sialolithiasis was made.

INVESTIGATIONS

All the routine blood examinations were performed. Occlusal radiograph was taken and no obvious sialolith was noted on the anterior aspect of the floor of the mouth. OPG was carried out and it revealed a well-demarcated radiopacity in relation to 37 and 38 region and continuous downwards towards the lower border of mandible suggesting of salivary calculi in the posterior aspect of the left submandibular duct.

TREATMENT DONE

Excision of sialolith was planned under local anesthesia. 2% lignocaine hydrochloride with 1:2,00,000 adrenaline was infiltrated lingually in relation to 37 and 38 region. A 1 cm incision was placed on the lingual aspect of 37 and 38 region and blunt dissection was carried out further to explore the left submandibular duct and to access the calculi. The submandibular salivary calculi were identified, which was 2 in number. Using digital pressure, the calculi was pushed upwards and retrieved (figure 2). The two calculi measured approximately 1.5 x 0.5 cm and 1 x 0.8 cm respectively (figure 3). Milking of the gland was done to stimulate saliva secretions. Hemostasis was achieved. Closure done with 3-0 vicryl. Post-operative antibiotics and analgesics were given for 3 days. The surgical site was healed without any complications.

DISCUSSION

Several theories have been proposed for sialolith formation which includes inflammatory, infective, mechanical, neurogenic, and chemical origin[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[7,8]. Calculi formation occurs when organic material such as inspissated mucous, ductal epithelial cells, salivary proteins, and foreign bodies that serve as a scaffold for the deposition of inorganic material (calcium salts)[4]. Crystallization takes place right after and give rise to the initial hydroxyapatite focus. This focus acts as a scaffold onto which further deposition is added, expanding the size of the sialolith. The submandibular gland is most susceptible because its saliva is more alkaline, has a greater concentration of calcium and phosphate and has a higher mucus content than the saliva of the parotid and sublingual glands. In addition, the submandibular duct is longer and has a tortuous course. The submandibular gland also has an antigravity flow.

Large salivary calculi are either oval or elongated in shape. They are hard, yellow in color with a porous aspect[9]. The calculi in our case followed the same typical appearance. In our present case report, unusual size and location of sialoliths was noted. The two sialoliths measured 1.5 x 0.5 cm and 1 x 0.8 cm respectively and present on the posterior aspect of the left submandibular duct, near the hilum. Although large sialoliths have occasionally been reported in the salivary gland, they have rarely been reported in the salivary ducts[10-13]. The detection of giant sialolith is often based on clinical history, a thorough clinical examination, and radiographic evidence. Investigations such as plain radiography shows the radio opaque stones in most cases (80–95% of sialoliths). Intraoral occlusal radiographs have also proven to be useful. Due to the unusual location as in this case, sialolith was not detected with intra oral occlusal radiograph. OPG was taken and it revealed a well-demarcated radiopacity in relation to 37 and 38 region and continuous towards the lower border of mandible. Computerized tomography scan is the most accurate non-invasive diagnostic tool for establishing the position of calculi[9,14]. Other investigations includes sialography and ultrasound imaging. Ultrasound is an another non-invasive method of identifying sialoliths. For an accuracy of 99% in ultrasound imaging, the size of the sialolith should be bigger than 1.5 mm, and its mineral content should be high[15].

The treatment is determined by the history of the patient's symptoms (duration of symptoms and number of repeated episodes), size and location of the sialoliths. Conservative management should be considered in cases of small stones. This includes application of warm compresses, massaging the gland, using sialagogues to increase salivary flow, ensuring that the patient is well hydrated, and antibiotics in case of infections[16]. Extracorporeal shock wave lithotripsy (ECSWL) has been successful in treating small salivary gland stones[3]. Large sialoliths can be removed surgically either through a transoral approach or through a transfacial approach. If the stone is located within the horizontal portion of the duct, the stone can be retrieved through a small mucosal incision and the ductal lining is sutured to the mucosa of the floor of the mouth. This procedure

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is known as sialodochoplasty. Sialendoscopy is an another intervention, for the elimination of giant salivary sialolith[17]. Submandibular gland excision is advocated in substantial intra-glandular sialolith, which cannot be approached through a transoral approach. Transfacial sialadenectomy is the preferred treatment when stones are located close to the hilum or in patients with persistent sialadenitis that is refractory to other methods of treatment. The risk of mandibular nerve palsy after excision of the gland is around 8%.

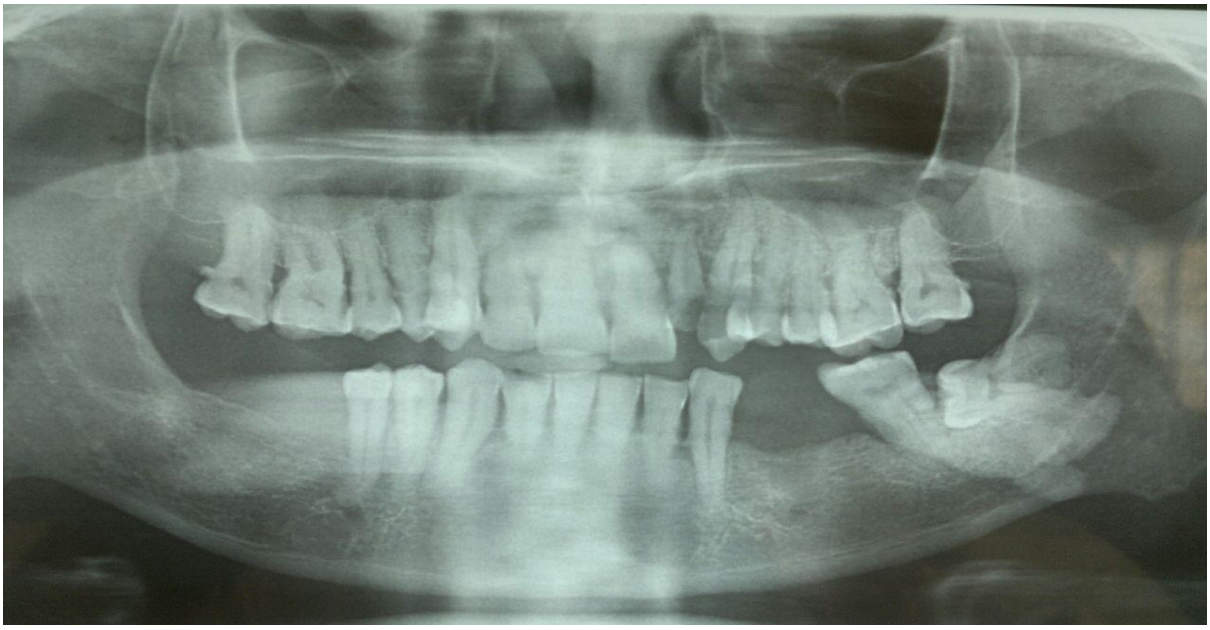

Figure 1: OPG showed radiopaque mass noted on the 37, 38 region suggestive of salivary calculus on the posterior aspect of the submandibular duct

Figure 2: Surgical excision of the salivary calculi

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Figure 3: Excised salivary calculi; 2 in number

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