Submandibular giant sialoliths—2 case reports and review of the literature

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Abstract Sialolithiasis is one of the most common diseases of the salivary gland. The submandibular gland and its duct appear to be the most susceptible. However, giant sialoliths have rarely been reported in the literature. Two case reports of giant sialoliths in the submandibular duct alone are presented. In both patients, a transoral removal of the stone was done under local anesthesia. The stones were 35 mm and 25 mm long, respectively. Follow-up showed asymptomatic and normal functioning glands. The etiology, pathogenesis and management of such giant sialoliths are discussed.

It is interesting to note that both patients remained relatively asymptomatic in spite of such long-standing sialoliths. The glands also recovered normal function quickly following removal of stones, which is interesting as such long-standing stones usually produce irreversible functional damage to the gland. A conservative approach to the gland will spare the patient the morbidity associated with gland removal.

Keywords Giant salivary sialoliths · Submandibular duct

Introduction

Sialolithiasis is one of the most common diseases of the salivary glands in middle aged individuals [1, 2]. The majority of sialoliths (80%) occur in the submandibular gland or its duct and are a common cause of acute and chronic infections of the gland [3].

The size of salivary calculi may vary from less than 1 mm to a few cm in size, with most calculi being less than 10 mm in size. However, giant sialoliths (>15 mm) in the submandibular duct have rarely been reported [4, 5].

The purpose of this paper is to report two cases of giant submandibular duct calculi and to review the literature on giant sialoliths.

Case reports

Case 1

A 41-year-old Sudanese man reported to the Department of Oral and Maxillofacial Surgery, Al Arab Medical University, following referral by a the general dental practitioner, for management of a firm mass in the anterior part of the left side of the floor of the mouth. History revealed that this patient had complaints of recurrent pain and swelling for the past several years (8 years), which were managed symptomatically with analgesics. The pain characteristically increased during meal times only. However, he also reported a total cessation of symptoms during the past 2 years.

Extraoral examination revealed a palpable, non-tender, left submandibular salivary gland, while intraoral examination showed a large mass, protruding and rupturing through the mucosa of the left floor of the mouth (Fig. 1).

On the basis of history and clinical examination, a diagnosis of left submandibular duct calculi was made. Lower occlusal and panoramic radiographs showed a large radio opaque mass located in the left floor of the mouth,

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extending beyond the distal surface of the lower left second molar (Fig. 2). Blood and serum biochemistry findings were within normal limits and his health history was unremarkable.

At a subsequent appointment, the stone was removed by a transoral approach, with sharp dissection under local anesthesia (2% lignocaine with 1:80,000 adrenaline). The wound was left to heal by secondary intention. The stone was measured to be 34 mm along its greatest length (Fig. 3). The postoperative period was uneventful. The patient was advised to perform warm saline mouth rinses and prescribed analgesics and sialogogues.

Due to the presence of a palpable left submandibular gland and the large size of the stone, the patient was reviewed after a period of 1 month postoperatively. On review, the gland, although still palpable, continued to be asymptomatic and clear saliva could be expressed from the submandibular duct on massage of the gland. The gland

was no longer palpable after 3 months following the stone removal.

Case 2

A 32-year-old Libyan woman reported to the Department of Oral and Maxillofacial Surgery, with a mass in the left side of the floor of the mouth. She gave a history of multiple episodes of pain and swelling in the left lower part of the mandible, during the past 4–5 years, especially at meal times. The patient was examined by her dentist and diagnosed with a carious lower molar as the causative factor, which was subsequently extracted. However, the persisting submandibular swelling resulted in the patient being referred to us. On enquiry, she gave an additional history of a significant reduction of symptoms in the last 1 year.

On extraoral examination, the left submandibular gland was palpable and non-tender. Intraorally, a giant sialolith was seen piercing through the mucosa of the left floor of the mouth in the region of the first and second lower molars. Radiographs confirmed the provisional diagnosis of a left submandibular duct sialolith. Ultrasonography did not reveal any calcifications in the glandular structure. Laboratory investigations were within normal limits.

The stone was removed through a transoral approach, with sharp dissection under local anesthesia (2% lignocaine with 1:80,000 adrenaline). Postoperative analgesics and sialogogues were prescribed. The stone was measured to be 25 mm along its greatest length (Fig. 4). On review after 6 months, the patient was totally asymptomatic. The submandibular gland was no longer palpable and clear saliva could be expressed from the duct on massage of the gland.

Fig. 1 Sialolith perforating through the oral mucosa

Fig. 2 Lower occlusal radiograph showing the presence of the stone extending beyond the second molar region

Discussion

Salivary stagnation, increased alkalinity of saliva, increased calcium content of the saliva, infection or inflammation of the salivary duct or gland and physical trauma to the sali-

Fig. 3 Giant sialolith measuring 35 mm along its greatest length

Fig. 4 Giant sialolith extending 25 mm along its greatest length

vary duct and gland may predispose to calculus formation [6]. There are currently two theories about the pathogenesis of salivary calculi. The first is that intracellular microcalculi develop within autophagosomes in normal salivary tissue and are naturally voided through the duct system. However, if these became impacted during their discharge, they will act as a nidus for stone formation [7]. An alternative explanation is that a mucous plug forms, which calcifies after being exposed to a supersaturated solution [8].

The submandibular gland is most susceptible because its saliva is more alkaline, has a greater concentration of calcium and phosphate, and has a higher mucous content than other major salivary glands. Also, the submandibular duct is longer with a kink over the posterior border of the mylohyoid muscle and has an antigravity flow. However, the hypothesis of a correlation with the consumption of hard water has been disproved [9].

Although large sialoliths have occasionally been reported in the salivary gland [10, 11] they have been rarely reported in the salivary ducts [12–14]. The largest reported stone in the submandibular duct was about 55 mm in length [15]. The salivary sialoliths, in both our cases, are comparable to the other giant sialoliths reported in the literature.

The ability of a calculus to grow and become a giant sialolith depends mainly upon the reaction of the affected duct. The growing stone causes increasing obstruction of salivary secretion, which leads to swelling, pain and infection of the gland and finally to surgical intervention [16]. However, if the duct adjacent to the sialolith is able to dilate, allowing normal secretion of saliva around the stone, it might be asymptomatic and allowing the creation of a giant calculus eventually. As the stone increases in size beyond the ability of the duct to dilate, a sialooral fistula may develop and the sialolith may partially protrude into the oral cavity. The affected gland may take advantage of the fistula to improve salivary secretion, allowing further enlargement of the stone. Satisfactory salivary flow can result in an asymptomatic period [17]. The creation of a similar fistula in

both our patients may account for the relatively symptom-free period at the time of presentation.

The treatment objective of giant sialoliths is restoration of normal salivary secretion. The sialolith should be removed with a minimally invasive procedure, usually through a transoral sialolithectomy, to avoid morbidity associated with sialadenectomy. Conflicting views exist with regard to the surgical treatment of a salivary stone in the knee of the submandibular duct, the options being excision of the submandibular gland or transoral removal of the stone alone. Nearly all submandibular stones, including those at the knee of the duct, can successfully be removed transorally [18]. Palpability of the calculi, regardless of its location or size, is considered to be the most important factor in the successful transoral removal of the stone [19]. The key question is whether or not a gland with such a stone has suffered irreparable functional damage. While experimental studies show extensive regeneration of the glandular parenchyma after prolonged ligation of the excretory duct of the submandibular gland [20], clinical studies have shown satisfactory return to normal functioning of the salivary glands in a vast majority of patients with transoral removal of the stone alone [21–24]. However, factors such as glandular infection, calculus diameter and patient's age can influence the recovery of function [25].

Both our patients showed a total relief of symptoms following transoral removal of the stone. A clear flow of saliva could be expressed from the ducts during the massage of the gland. A sialolith logically must result in an irreparable functional damage to the gland. However, the fact that, in both cases, there was a rupture of the overlying mucosa, creating a fistula, could have contributed to the absence of significant backpressure to damage the gland. The good recovery of the gland in both cases leads us to conclude that a conservative approach to the gland may be considered even in the presence of a large, long-standing sialolith in the submandibular duct. This not only leaves the submandibular gland intact as a functioning organ but also obviates the risk of surgical complications associated with its removal.

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