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Case Report

Self-Extrusion of Giant Sialolith in an Adolescent: Case Report

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Abstract

Introduction: Sialolithiasis can cause obstructive symptoms and sialadenitis. Rarely, large sialoliths self-extrude from ductal openings instead of requiring surgical removal.

Case Report: A 17-year-old boy presented with a painful 3-day history of right submandibular swelling, fever, and malaise, with prior similar symptoms 5 years ago. Examination showed a tender, erythematous 2x2 cm swelling and purulent discharge from the right Wharton's duct. CT revealed an edematous right submandibular gland, an 18*8 mm sialolith proximally, and a 6*6 mm stone distally in the duct. He was managed conservatively with intravenous antibiotics, fluids, and analgesics. On day 3, the smaller than the larger stone self-extruded through the duct over 8 hours, followed by purulent discharge. Over 2 days, the swelling and erythema subsided. At 2-week follow-up, he was symptom-free.

Conclusion: Even giant sialoliths can self-extrude with conservative treatment, averting surgical removal in acute sialadenitis.

Keywords: Sialolith; Giant Sialolith; Self Extrusion; Sialadenitis

Introduction

Sialolithiasis is the most common disease of the salivary gland [2,5]. It affects about 1.2% population [2,5-7]. Around 10% of all cases of sialadenitis are localized to the submandibular gland [8,9]. Larger sialoliths causing obstruction and stasis of saliva can cause sialadenitis and may require surgical procedures for stone removal after initial conservative management. But rarely, sialoliths can extrude themselves from the ductal opening or form a sialo-oral fistula after ulceration in the duct.

Case Report

An adolescent boy came to our secondary health care center with painful swelling in the right submandibular region for three days that aggravated when taking food. He had high-grade fever and malaise for the past two days. He had a history of similar swelling in the right submandibular region five years back, for which he was treated with oral antibiotics on an outpatient basis.

A smooth, ovoid swelling of 2x2 cm in size was found in the right submandibular area during a local examination. The skin in the area was tense and showed signs of erythema, local temperature rise, and tenderness over the swelling. Upon bimanual palpation, it was suggested that the right submandibular gland was swollen, with stony concretions present along the gland's duct on the oral cavity floor. No lymph nodes were felt during the examination. The mucosa of the oral cavity over the floor of the mouth was edematous, and pus discharge from the opening of the right Wharton's duct was seen when pressure was applied to the swelling.

The patient underwent a complete hemogram, and the results were within normal limits. Additionally, a non-contrast computed tomography of the neck was performed, which revealed edematous glands and soft tissue in the right submandibular region. Two homogenous calcified masses were seen in the right Wharton's duct near its opening on the floor of the mouth (Figure 1; Figure 2).

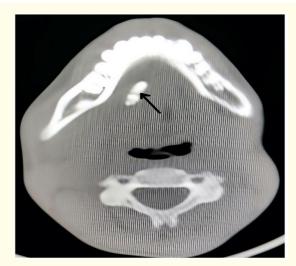


Figure 1: CT image showing proximally situated, larger sialolith in right Wharton's duct.

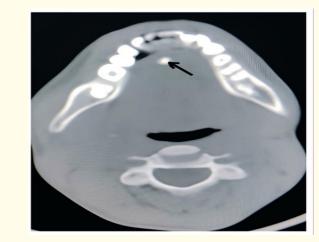


Figure 2: CT image showing distally situated, smaller sialolith in right Wharton's duct.

The patient was started on conservative management with intravenous cefotaxime at 1g thrice daily, intravenous fluids, analgesics, and antipyretics. The patient was advised to massage the swelling gently. Continuous monitoring of vitals was done.

On day 3 of the admission, the patient reported the self-extrusion of one of the sialoliths that measured 6*6 mm (Figure 3), followed 8 hours later by the second, larger sialolith. Larger sialolith measured around 18*8 mm (Figure 3). Purulent discharge was observed from the opening of the right Wharton's duct in the floor of the mouth, surrounded by edematous mucosa (Figure 4). The size of the swelling, along with erythema over the swelling, subsided over the next two days. The wound on the floor of the mouth was allowed to heal by secondary intention. The patient was discharged on oral antibiotics and analgesics and reviewed on an outpatient basis.



Figure 3: Two self-extruded sialoliths, one measuring 18*8 mm in size (**) and the other measuring 6*6 mm in size (*).

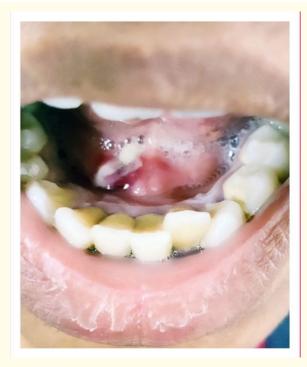


Figure 4: Floor of the mouth after self-extrusion of two sialoliths from right Wharton's duct.

The patient was reviewed in the outpatient department after two weeks. The patient did not report any complaints. The wound over the duct had healed well, and there was no tenderness or pus discharge on palpation.

No complications were observed, and the patient returned to his daily activities. The patient was asked to report to the department in case of any recurrence.

Discussion

Patients with sialolithiasis may present with chronic fluctuating pain and swelling in the submandibular region. Here, the patient gave an account of two such incidents five years apart.

Not all cases of sialolithiasis cause obstructive symptoms. Even large sialoliths can be asymptomatic as saliva seeps around the irregularly shaped sialolith and exits through the ductal opening [1]. In this case, it is possible that saliva surpassed the obstruction caused by sialolithiasis, and the patient managed to remain symptom-free for a long time between the two episodes. Gradually, complete obstruction of the Wharton's duct must have led to the stasis of saliva. When salivary flow normally occurs, gland tissue and duct

are cleared of pathogens. Failure of normal production and flow of saliva causes commensal oral microbes to migrate back into the duct and parenchyma, where they proliferate using stagnant saliva as a medium [4,10]. Due to suppuration and abscess formation, the duct must have undergone ulceration, leading to self-extrusion of the giant sialolith, which was already present near the distal end of the duct. Causative organisms for acute suppurative sialadenitis include Staphylococcus aureus, Streptococci, E. coli, Pseudomonas aeruginosa, and Moraxella catarrhalis [9].

Sialoliths usually measure from 1mm to less than 10 mm in size [11,12]. Sialoliths with a size larger than that are referred to as sialoliths of unusual size and those more than 15 mm are called giant sialolith [6,7,11,13]. Wharton's duct is 5-6 cm long and about 1-3 mm in diameter. In this report, the size of the larger sialolith was found to be 18 mm*8 mm. It was located in the duct of the right submandibular gland.

There are rare incidences of self-perforation of the duct by sialoliths. 8 cases of self-extrusion of sialolith from the submandibular duct occurred in the last decade were collected from literature by Alhamdani [13]. The largest of those stones measured 40 mm in size and was reported by Hegde., et al. Alhamdani also writes about a case of an 18 mm sialolith expulsion in a 35-year-old male in his report [13]. Cases of self-extrusion of giant submandibular sialolith were reported in a 36-year-old male and a 65-year-old male, respectively, by Suzzane., et al. and Saluja., et al. [6,11]. More recently, in 2021, Dam., et al. reported self-extrusion of a giant sialolith in a 17-year-old male [7]. In our case report, the patient was relatively younger, 17 years of age.

Occlusal radiograms were a standard approach in prior times, but they are insensitive for radiolucent stones and detecting abscess formation. Non-contrast computed tomography and magnetic resonance imaging are reliable modalities for detecting stones and abscess formation and deciding on management options. Hence, we subjected our patient to CT scan imaging. Sialography was contraindicated as the patient was in the acute stage of infection.

The use of antibiotics was warranted in this case as there was an active infection, and the patient was maintained on intravenous fluids in view of dehydration and decreased oral intake. The self-expulsion of the stones was aided by gentle massage over the swelling.

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Conclusion

Every case of submandibular sialadenitis caused by sialolithiasis need not undergo surgical procedures and can be managed conservatively. Symptomatic management and antibiotics help resolve and improve clinical conditions in patients.

There is a chance of self-extrusion of the sialolith, especially if the stone is present towards the distal end of Wharton's duct.

Ethics Approval and Consent to Participate

Descriptive case report and consent was waived.

Consent for Publication

Written informed consent for publication of their clinical details and/or clinical images was obtained from the patient.

Availability of Data and Material

The datasets during and/or analyzed during the current study are available from the corresponding author upon reasonable request.

Author's Contribution

All the authors have equally contributed to the case report. KR and VK were the major contributor in writing the manuscript. VMS, NK and PR participated in writing, editing and data interpretation along with KR and VK. All authors have approved the manuscript.

Competing Interests

The authors declare that they have no competing interests

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