

Chronic Sialolithiasis Of The Submandibular Gland With A Gigantic Collection Of Stones: A Case Report

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Abstract

Objective: Sialolithiasis, characterized by the accumulation of calcium salt deposits, often affects the submandibular gland nestled within the submandibular triangle. This disorder primarily impacts the submandibular gland due to the lower positioning of Wharton's duct, facilitating retrograde infection by oral flora. The alkaline saliva produced by the gland actively promotes the precipitation of calcium salts, consequently fostering the formation of sialoliths.

Methods: This case study showcases a 35-year-old male patient who developed a huge sialolith and persistent sialadenitis after recurring submandibular gland swelling and pain. The patient revealed a ten-year history of recurrent painful, persistent swelling and a gradual increase of dysphagia during his last seven-month illness. Chronic sialolithiasis was diagnosed when imaging revealed a large sialolith collection. A contrast-enhanced neck CT confirmed a massive intra-parenchymal stone aggregate and an enlarged gland. Excision of the submandibular gland was decided as the definitive treatment. Following submandibular sialoadenectomy, unusually shaped stones weighing 189 grams were discovered. A histopathological study confirmed the presence of squamous and mucinous metaplasia together with persistent sialadenitis, ruling out cancer. At the three-month follow-up, the patient showed favourable healing and improved speech.

Results: The objective is to highlight a gigantic collection of stones with unusual presentation in submandibular sialolithiasis, clinical features, diagnostic techniques, and treatment options.

Conclusions: Highlighting the complexities of submandibular sialolithiasis, this case report emphasizes the need for early and easy access to timely healthcare provision in getting precise diagnoses and tailored treatment plans and avoiding complications related to delayed and unreachable healthcare. Successfully performing a submandibular sialoadenectomy led to significant relief of symptoms, demonstrating the effectiveness of surgical approaches in managing chronic sialadenitis with large sialoliths.

Keywords: Sialolithiasis, Submandibular Gland, Chronic Sialolithiasis and Sialadenitis, Sialoadenectomy, Gigantic Stones

Introduction

The submandibular gland resides in the submandibular triangle, a bony depression formed by the base of the jaw and the anterior and posterior digastric muscles. From the gland's medial surface, Wharton's duct protrudes.¹ Aggregations of calcium salts, known as sialoliths, are the most common cause of disorders in major and minor salivary glands. Due to the lower level of the oral cavity and the location of Wharton's duct, oral bacteria can induce a retrograde infection in it. This anatomical arrangement makes it easier for oral germs to enter the submandibular gland. Moreover, the saliva of the submandibular gland has an alkaline pH, which encourages the synthesis of calcium salts.² Because of its alkalinity, calcium salts like hydroxyapatite precipitate more easily and can build up to create sialoliths inside the gland or duct, possibly resulting in sialolithiasis.

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Sialolithiasis is the most common condition in this category, accounting for 1.2% of unilateral large salivary gland swellings. In 80% of instances, the submandibular gland is the most susceptible, followed by the parotid (19%) and sublingual (1%) glands.³ Typically observed in people in the thirty to sixty age range, sialolithiasis is rare in children, accounting for only 3% of pediatric cases that are documented. Males are disproportionately affected by the illness; their incidence is double that of females.⁴

Although experts don't know the exact cause of this ailment, they attribute it to variables such as decreased salivary flow and duct inflammation, which can cause an injury characterized by mucus obstructions and eventual calcification. The bigger size and downward angle of Wharton's duct are the reasons it tends to happen in the submandibular gland. Ductal alterations and foreign objects are additional important causes.⁵

Clinical symptoms include discomfort and swelling in the affected gland; the signs tend to aggravate if the duct is obstructed.⁶ Recurrent symptoms are typical, and they are usually more noticeable during meals. Sialoliths larger than 15 mm in any dimension are referred to as enormous, while larger than 35 mm are referred to as gigantic.⁷ Stones can be clinically diagnosed, asymptomatic, found by imaging (such as X-rays or ultrasonography), or present with problems (such as infection or blockage). The position and size of the stone determine the different treatment approaches.⁸

In this case study, a large, painful submandibular sialolith in a male patient aged 35 is presented in this report. There is a thorough analysis of the literature that highlights aetiology ideas, typical and atypical presentations, clinical characteristics, diagnostic techniques, and treatment modalities.

Case Presentation

A 35-year-old male who had been experiencing pain and swelling in his right submandibular region for seven months, along with mild dysphagia, came to the outpatient department (OPD) of King Abdul Aziz Specialist Hospital, Taif, to seek treatment. Surprisingly, the patient revealed a ten-year history of comparable complaints with a progressive deterioration of symptoms. Notably, the patient's medical history was free of any known allergies or chronic conditions. Examining the patient in detail revealed stable vital signs, a typical physical appearance, and a noticeable enlargement in the right submandibular region. There was crepitus and tenderness in the right submandibular gland. Even though there was modest dysarthria, there was no paresthesia and normal tongue movements, which added clinical context.



Figure 1(a)

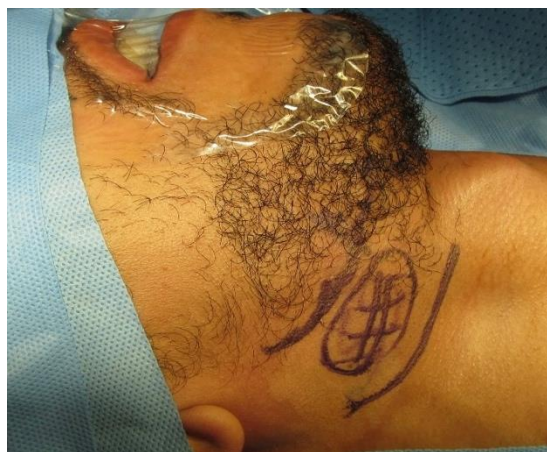


Figure 1(b)

Figure 1. (a), (b) Pre-operation photograph depicting the right submandibular gland swelling in a 35-year-old male diagnosed with submandibular sialolithiasis.

A significant accumulation of stones was discovered during a routine orthopantomogram (OPG) X-ray and neck ultrasonography, which is diagnostic imaging for chronic sialadenitis with sialolithiasis affecting the right submandibular gland. An enlarged right submandibular gland with a wide lobulated outline, laminated texture, and an intra-parenchymal stone mass measuring

33x37x45mm was seen upon further investigation using a contrast-enhanced neck CT scan. Notably, there was no discernible mass effect, and a few air locules were seen surrounding the stone bulk.

The patient was then admitted for a submandibular sialoadenectomy, a surgical procedure performed under general anaesthesia. There were no indications of mass impacts throughout the procedure. Examination of the removed gland showed the presence of oddly shaped stones weighing 189 grams in total.

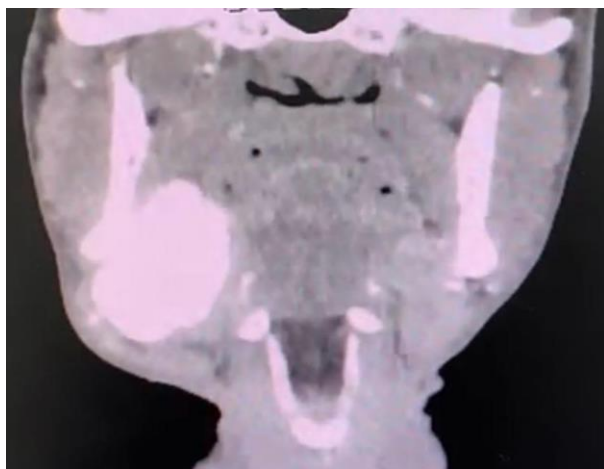


Figure 2(a)



Figure 2(b)

Figure 2. (a), (b) CT scan of the neck revealing the presence of a sialolith. (b) Radiographic presentation confirming the presence of a sialolith.

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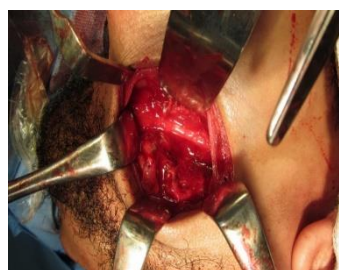


Figure 3(a)



Figure 3(b)

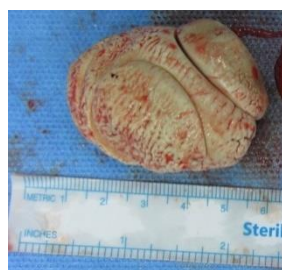


Figure 3(c)



Figure 3(d)

Figure 3. (a) Intraoperative image capturing the submandibular gland in its original position in a 35-year-old male patient. (b), (c), (d).

Histopathological investigation of the removed specimen validated the diagnosis of chronic sialadenitis, revealing squamous and mucinous metaplasia in the ducts. Crucially, cancer was ruled out. The patient demonstrated better speech and favourable surgical site healing during the three-month follow-up, highlighting the effectiveness of the surgical intervention and all-encompassing care for chronic sialadenitis with sialolithiasis.

Discussion

An estimated 12 out of 1000 persons are believed to be affected with sialolithiasis, a frequent disorder that affects the salivary glands. Adults are more likely than children to suffer from this condition.⁹ Although it can happen to anyone at any age, it usually affects people in their third to sixth decades of life; the patient, in this case, is in their third decade. With a 2:1 male-to-female ratio, this disease is more common in men and can present unilaterally or bilaterally without showing a clear preference for the right or left side.^{8,10} In contrast to the 1:1 male-to-female ratio reported by Seldin et al.¹¹ and Lustmann et al.¹² Our findings align with the general tendency of greater male sensitivity to sialolith development.

More than 80% of sialoliths are located in the submandibular gland or its ducts; the remaining 5–20% are located in the parotid gland, and the remaining 1–7% in the sublingual gland.^{5,9} Major salivary glands are not the only salivary glands affected by sialoliths; minor salivary glands account for about 6% of instances. The duct contains a large portion of the stones in the submandibular gland; 34% of the stones are in the distal duct, and 57% are in the hilum. 10% of sialoliths are located in various small salivary gland locations, 35% are located in the buccal mucosa, and only 10% of them are found in the lower lip.^{9,11} Interestingly, prior research has shown that sialoliths seem to grow more commonly in the left submandibular gland than in the right. This, however, is the same as our scenario. These stones can be intraglandular or intraductal, and the current case showed squamous and mucinous metaplasia in the ducts and significant ductal epithelium squamous metaplasia.¹³

Sialoliths typically have a maximum diameter of 5 mm; those that are more than 10 mm are regarded as unusually large. Monster sialoliths are stones greater than 15 mm in any dimension. Rai and Burman described one of the biggest sialoliths, measuring 72 mm. Salivary stones can have radiolucent cores and outside radiopaque portions.^{5,9,10} However, Rust and Messerly found a 51 mm sialolith in a 66-year-old male that extended the full length of Stensen's duct.¹⁴ The diameter of the stone found in our case was 33x37x45mm, which was also considered a giant sialolith.

The genesis of sialolith production needs to be properly understood. Reports, however, indicate that the creation of intracellular microliths serves as a foundation for the production of stones. Secretory problems result from a rise in microorganisms brought on by dysfunction and rising microlith formation, which in turn causes localized blockage and acinar cell atrophy. According to a different opinion, the development is caused by an increase in calcium and phosphate salts, which deposit bacteria, salivary mucus, and desquamated cells together.¹⁵ Infections, abnormalities of the salivary glands, foreign objects, ductal malformations, and metaplasia of the ductal epithelium are additional possible causes of sialolithiasis. The key factor influencing the calculi's capacity to enlarge beyond reasonable limits is the afflicted duct's response.

Sialoliths might appear as "mealtime syndrome," a recurring uncomfortable swelling that occurs during meals, or they can be asymptomatic.⁴ Acute suppurative sialadenitis and abscess formation are examples of complications that could also happen. Along with symptoms like trismus, pus discharge, lymphadenopathy, and, in rare instances, systemic infections, sialolithiasis pain can also present itself. Phleboliths, cervical lymphadenopathy, abscessed teeth, calcified hemangiomas, tonsillitis, and benign salivary gland tumours are among the differential diagnoses.¹⁶ In this instance, the patient's duct dilation next to the sialolith caused painful swelling, which allowed normal salivation around the stone and contributed to its significant growth.

It is believed that mineral salts gather around a core of bacteria, mucus, or lost cells to produce salivary calculi.¹⁷ Several factors, including decreased salivary flow, high saliva alkalinity, and excessive calcium levels, both physical harm to the salivary duct or gland and inflammation of the duct or gland, can facilitate calculus formation. Compared to the parotid and sublingual glands, the submandibular gland produces more mucus and more alkaline saliva with higher amounts of calcium and phosphate. This makes it more vulnerable.¹⁸

A complete approach combining a detailed history, clinical and physical tests, and appropriate imaging modalities is necessary for accurate diagnosis and therapy planning. One can use a variety of radiographic examinations, including scintigraphy, magnetic resonance imaging (MRI), CT scans, sialography, ultrasonography, standard radiography, and sial endoscopy.¹⁹ It is noteworthy to avoid performing sialography when the calculus is in the distal portion of the duct because the contrast agent may impede clearance by moving the calculus in the direction of the gland. Scintigraphy is advised in certain situations.

Due to their radiopacity, X-rays may not be able to detect every case of submandibular lithiasis; nonetheless, CT scans are the best noninvasive method for identifying sialoliths. But ultrasonography has its limitations, especially when it comes to identifying calculi that are less than 2 mm. When CT scans fail to identify the calculus, sial endoscopy—a minimally invasive treatment that provides excellent visualization of the ductal system is recommended.¹⁶

The position and size of the stone are among the variables that affect sialolithiasis treatment methods. For tiny stones, conservative treatment is appropriate and includes sialagogues to encourage saliva flow, warm, moist heat, gland massage, and refreshment. Infection requires the prescription of antibiotics. Depending on the position of the stone, sialolithotomy or sialodochoplasty may be necessary. For big proximal stones, extracorporeal shock wave lithotripsy (ESWL) is an option; however, sial endoscopy and endoscopic intracorporeal shock wave lithotripsy (EISWL) are becoming more and more popular as less intrusive options.²⁰

The therapeutic approach used is based on the particulars of the patient. Stones in the gland or the posterior portion of the duct may require submandibular sialoadenectomy, but stones close to the duct orifice may be removed intraorally.⁵ Our patient also went through submandibular sialoadenectomy and completely recovered after three months of follow-up.

Conclusions

To summarize, the case study clarifies the intricate characteristics of submandibular sialolithiasis, highlighting its incidence, subtle diagnostic challenges, and effective treatment approaches and the need for early and easy access to the timely provision of healthcare in getting precise diagnoses and tailored treatment plans and avoiding complications related to delayed and unreachable healthcare. This 35-year-old male patient's unusual presentation of repeated symptoms culminating in a large stone collection is consistent with unusual clinical features. CT scans and other diagnostic instruments were very helpful in directing the submandibular sialoadenectomy that was performed.

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S.A.K, A.H.E - Experimentation/Study Conduction

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