



The Largest Asymptomatic Sialolith: Report of a Rare Case

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Abstract

Sialolithiasis is the most common pathology affecting the salivary glands. The submandibular gland is most commonly affected. Sialoliths typically causes pain and swelling in the affected gland. They usually range in size from 1 to 10 mm, while those larger than 15 mm are considered rare. The exact etiology of sialolith formation remains unknown. We report the case of a 62-year-old patient with submandibular gland sialolith, that was incidentally discovered during imaging performed for dental implant planning. Remarkably, the patient was completely asymptomatic, despite the stone being giant in all three dimensions. Cone-Beam Computed Tomography (CBCT) of the mandible revealed a radiopaque lesion posterior to the mandibular Body, measuring approximately 2.8×1.8×2 cm. The sialolith was successfully removed using a minimally invasive transoral sialolithotomy.

Keywords: Dental implants, Mandible, Pain, Salivary gland calculi, Spiral cone-beam computed tomography, Submandibular gland

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Introduction

Sialolithiasis is the most common pathology of salivary glands (1). It is characterized by obstruction of salivary flow due to a calculus, which is accompanied by pain, swelling, and inflammation of the affected gland (2).

Sialolithiasis affects approximately 1% of the general population (3), most commonly between the third and sixth decades of life (2,4). Men are affected twice as often as women, and the condition rarely occurs in children (5,6). More than 80% of the sialoliths occur in the submandibular gland (6-8), whereas 5-20% are found in the parotid gland (8,9), and only 1-2% in sublingual and minor salivary glands (6,9). Sialolith formation is less common in intraglandular locations compared to intraductal locations (5). Submandibular calculi are mainly formed in Wharton's duct, whereas parotid stones are usually located in the gland parenchyma (9). Sialolithiasis is more likely to occur in the submandibular gland due to its anatomical position requiring drainage against gravity, alkaline pH and high viscosity of its saliva, higher mineral content, and the long, tortuous, and narrow orifice of its ductal system (5). Although the size of sialoliths is variable, most (88%) are less than 10 mm (6).

The diagnosis can easily be made through a detailed history and clinical examination, especially in the presence of a large sialolith. However, imaging can be helpful, particularly for small stones that are difficult to detect by palpation (10). Conventional radiography, digital subtraction sialography, ultrasonography,

computed tomography, and Magnetic Resonance (MR) sialography are imaging modalities that can be utilized to investigate salivary calculi (3). Sialoliths of the submandibular gland are typically recognized as radiopaque formations on plain radiographs and appear as radiolucent filling defects on sialography (5). In the literature, sialoliths measuring more than 15 mm in their maximum dimension are referred to as "giant". Giant sialoliths, also called megaliths have been rarely reported (5,10). This report presents a rare case of an asymptomatic giant sialolith of the submandibular gland, measuring 2.90 cm in its maximum dimension.

Case presentation

A 62-year-old male patient with a medical history of hypertension and diabetes mellitus was referred to our clinic for evaluation of an oral lesion, that was incidentally detected during imaging conducted for dental implant planning. Remarkably, the patient was completely asymptomatic, despite the considerable size of the lesion. He presented to us solely upon the recommendation of his dentist.

Intraoral clinical examination through bimanual palpation revealed a stony hard, non-tender mass located in the floor of the mouth, measuring approximately 3.5×3.5 cm in size. No swelling or bulging was observed on extraoral examination (Figure 1). However, a mild elevation was noted in the left floor of the mouth (Figure 2), which was not perceived by the patient. A saliva milking test

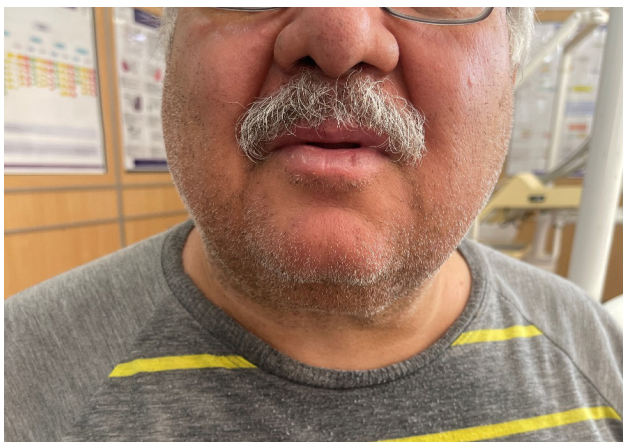


Figure 1. Extraoral clinical photograph showing no evident swelling or asymmetry in the left submandibular region.



Figure 2. Mild elevation in the floor of the mouth on the left side.

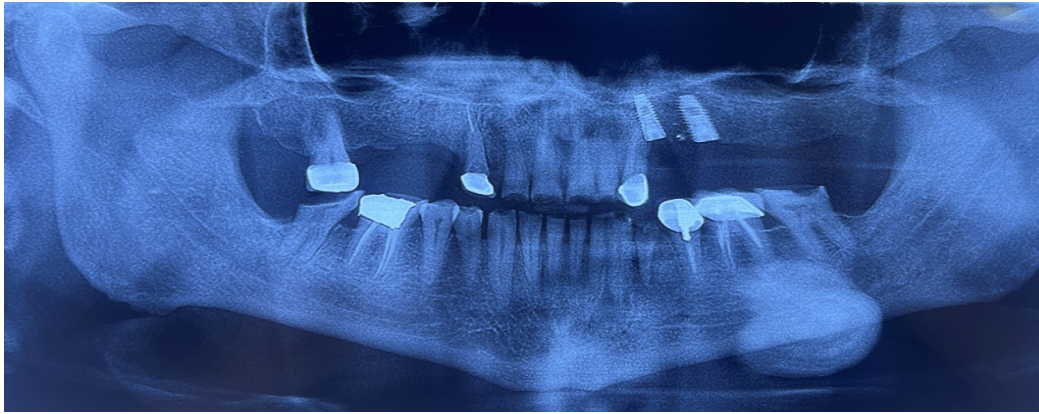


Figure 3. Panoramic radiograph revealing a well-defined radiopacity with a lamellar structure located in the posterior region of the left mandible.

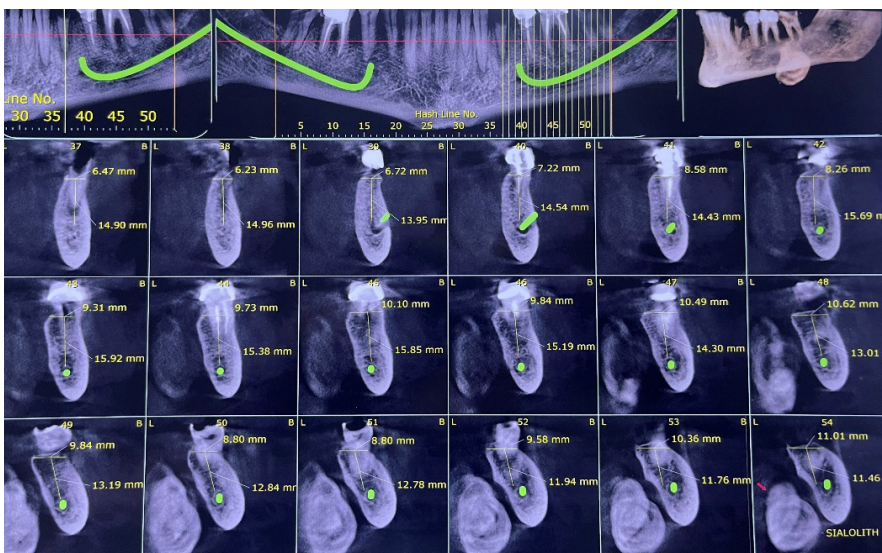


Figure 4. Sagittal sections of Cone-Beam Computed Tomography (CBCT) demonstrating a well-defined radiopacity with an onion-like appearance adjacent to the lingual cortex of the left mandibular molar region.



Figure 5. The excised sialolith following sialoadenectomy, placed beside a graduated ruler for size reference.

demonstrated a slightly reduced volume of saliva with normal coloration. The test did not provoke any discomfort or pain. Panoramic radiography revealed a well-defined radiopaque mass superimposed over the left posterior body of the mandible, in proximity to the first and second mandibular molars, measuring approximately 3.5×3.2 cm (Figure 3).

A provisional diagnosis of sialolithiasis was considered. Cone-Beam Computed Tomography (CBCT) of the mandible revealed a radiopaque lesion posterior to and near the mandibular body, measuring approximately $2.8 \times 1.8 \times 2$ cm (Figure 4). Ultrasonography of the left submandibular gland, with and without Doppler

study, was performed. It showed a normal-sized left submandibular gland with mild heterogeneity and a calcified sialolith (anteroposterior size: 2.07 cm). No abnormal vascularization was observed on color Doppler ultrasonography. Based on clinical and radiographic findings, a differential diagnosis of left submandibular duct sialolithiasis was proposed. Laboratory investigations revealed an HbA1c level of 6.7% and blood pressure was measured at $135/85$ mmHg. Under local anesthesia, the sialolith was removed *via* a minimally invasive transoral sialolithotomy. The excised stone measured $2.90 \times 1.90 \times 2.18$ cm (Figure 5).

Table 1. Comparative table of various cases of giant submandibular sialolithiasis with different characteristics and treatments

Author	Year of publication	Age/gender	Medical history	Location	Size	The basis of measurement	Symptom	Treatment approach
Soopanit Thanion (19)	2020	54/M	-	Right submandibular area	60×40×33 mm	Stone after surgery	Swelling	Surgery
Thong H <i>et al</i> (18)	2021	71/M	Hypertension, Diabetes mellitus	Right submandibular area	25 mm	Stone after surgery	Swelling	Surgery (Excision)
Mathew <i>et al</i> (20)	2022	50/M	Diabetes mellitus	Right submandibular area	39×5 mm	Stone after surgery	Swelling	Sialolithotomy using diode 810 μm laser
Aoun G Maksoud C (21)	2022	34/M	-	Right side of the floor of the mouth	17 mm	Stone after surgery	Swelling	Surgery (Excision)
Ungari C (9)	2022	70/M	Repeated sialolithiasis in the left Wharton's duct	Left side of the mouth floor,	18×10 mm	Stone after surgery	painful swelling	Surgery (Excision)
Konstantinos Chaidas <i>et al</i> (10)	2023	72/M	-	Right submandibular area	58×17 mm	Stone after surgery	Pain, Swelling Tenderness, and mild odynophagia	Surgery (Excision)
Chia-Wei Liu <i>et al</i> (6)	2023	48/F	Iron deficiency, Anemia, Biopsy of a benign right tongue tumor	Left side mouth floor	35×14 mm	Stone after surgery	Lumping sensation	Surgery (Excision)
Jit-Swen Mao (22)	2023	72/F	-	Right submandibular area	35×20 mm	Computed tomography	Swelling	Excision of the right submandibular gland and sialoliths was performed
Ashindoitiang John Adi (4)	2023	50/F	-	Left submandibular region	27×15 mm	Stone after surgery	Painless swelling	Surgery
Butt (23)	2023	38/M	-	Left submandibular area	45*×40×25 mm	Stone after surgery	Swelling	Surgery
Present case	2024	62/M	Hypertension, Diabetes mellitus	Left submandibular area	2.90×1.90×2.18 mm	Stone after surgery	-	Surgery

Abbreviations: F, female; M, male.

Discussion

Sialolithiasis is the most common cause of major salivary gland swelling (11). It refers to the formation of calculi within the salivary glands or their ducts (10). The composition of sialoliths can vary depending on

the relative concentrations of organic and inorganic components (12). Although the exact pathogenesis remains unclear, it is widely accepted that sialoliths form *via* the deposition of calcium-rich salts around a central nidus. This nidus may consist of desquamated

epithelial cells, foreign bodies, bacteria or their degradation products (13).

It has also been suggested that food debris or bacteria from the oral cavity can migrate into the salivary ducts and initiate stone formation (14). Salivary stones are composed of both organic and inorganic materials, including calcium carbonates and phosphates, cellular debris, glycoproteins, and mucopolysaccharides. Typically, the core of the stone is composed predominantly of organic matter, whereas the peripheral layers are mostly inorganic (15). Sialolith usually causes pain and swelling in the affected salivary gland with symptoms ranging from very mild to very severe (6). In the present case, despite the presence of a giant sialolith, the patient was completely asymptomatic and unaware of its existence (Table 1).

Prolonged obstruction in the absence of infection can eventually lead to glandular atrophy, loss of secretory function and subsequent fibrosis (16). Therefore, surgical treatment is recommended even in asymptomatic patients to prevent potential future complications. Sialolithiasis is most commonly observed between the third and sixth decades of life (2,4). Consistent with the typical age range, the present case involved a 62-year-old man. Some studies suggested that systemic diseases, medications, smoking, and alcohol consumption play no or only

a limited role in the development of salivary stones (17). Sialoliths typically range in size from 1 to 10 mm with an average of 6 to 9 mm. Stones larger than 15 mm are considered rare (18). In present case, the sialolith measured 29 mm, placing it within the rare “giant sialolith” category.

The primary goal in treating giant sialoliths is to restore normal salivary flow. Treatment options for submandibular sialoliths include interventional sialendoscopy, intraoral stone removal, and gland resection. The appropriate method depends on the stone’s location, size, and number (5). Stones located within 2 cm of the duct orifice may be removed intraorally, as indicated in the present case; whereas those located posteriorly in the duct or within the gland may necessitate submandibular sialoadenectomy (4).

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Conflict of Interest

There was no conflict of interest in this manuscript.

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