Case Report

https://doi.org/10.36377/ET-0135

Sublingual sialolithiasis in a patient with myofascial pain and pulp stones: A case report

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Abstract

Multidisciplinary evaluation and management are essential for achieving optimal outcomes in complex cases involving recurrent calcifications and chronic pain. This case highlights the rare coexistence of sublingual sialolithiasis, myofascial pain dysfunction syndrome (MPDS), and pulp stones or pulp canal obliteration. A 54-year-old male patient was diagnosed with sublingual sialolithiasis (2 × 1.6 cm), MPDS, and multiple pulp stones. Radiographic investigations, including bitewings and CBCT, revealed and confirmed the size of calculi and showed a significant pulp stone in posterior teeth. The patient underwent transoral sialolith removal and was treated with muscle relaxants and oral physiotherapy for MPDS. This case highlights the complex interplay among sialolithiasis, MPDS, and pulp stones. A multidisciplinary approach is essential for comprehensive management, especially in cases involving recurrent calcifications and chronic pain.

Keywords: sublingual sialolithiasis, myofascial pain dysfunction syndrome, pulp stones

Article info: received – 11.09.2025; revised – 14.10.2025; accepted – 20.10.2025

Conflict of interest: The authors report no conflict of interest.

Acknowledgements: There are no funding and individual acknowledgments to declare.

For citation: Alelyani A.A. Sublingual sialolithiasis in a patient with myofascial pain and pulp stones: A case report. *Endodontics Today.* 2025;23(4):597–602. https://doi.org/10.36377/ET-0135

Подъязычный сиалолитиаз у пациента с миофасциальной болью и пульпарными камнями: клинический случай

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Резюме

Междисциплинарная оценка и комплексное ведение пациента имеют ключевое значение для достижения оптимальных результатов при сложных клинических случаях, сопровождающихся рецидивирующими кальцификациями и хронической болью. В представленном клиническом наблюдении описано редкое сочетание подъязычного сиалолитиаза, миофасциального болевого дисфункционального синдрома (МБДС) и пульпарных камней либо облитерации корневых каналов. Пациент – мужчина 54 лет, у которого диагностированы подъязычный сиалолитиаз (2 × 1,6 см), МБДС и множественные пульпарные камни. Радиографические исследования, включая прицельные снимки и КЛКТ, позволили визуализировать и подтвердить размеры конкрементов, а также выявили значительный пульпарный камень в области жевательных зубов. Пациенту была проведена трансоральная операция по удалению сиалолита, после чего назначены миорелаксанты и курс оральной физиотерапии для коррекции МБДС. Данный клинический случай подчеркивает сложное взаимодействие между сиалолитиазом, МБДС и пульпарными кальцификациями. Комплексный междисциплинарный подход является обязательным условием эффективного ведения таких пациентов, особенно при сочетании рецидивирующих кальцификаций и хронической боли.

Ключевые слова: подъязычный сиалолитиаз, миофасциальный болевой дисфункциональный синдром, пульпарные камни

Информация о статье: поступила – 11.09.2025; исправлена – 14.10.2025; принята – 20.10.2025

Конфликт интересов: Авторы сообщают об отсутствии конфликта интересов.

Благодарности: Финансирование и индивидуальные благодарности для декларирования отсутствуют.

Для цитирования: Алилйани А.А. Подъязычный сиалолитиаз у пациента с миофасциальной болью и пульпарными камнями: клинический случай. *Эндодонтия Today.* 2025;23(4):597–602. https://doi.org/10.36377/ET-0135

INTRODUCTION

Sialoliths are calcified masses, known as salivary calculi, which occur within the ducts of salivary glands [1; 2]. The submandibular gland (Wharton's duct) exhibits the highest propensity for calculi development (80%), followed by the parotid gland (19%), with the lowest incidence occurring in the sublingual gland (1%). This condition is typically observed in male patients between 30 and 60 years old [3]. The clinical symptoms were a history of pain, swelling, and discomfort during mastication, which typically indicate the diagnosis [4]. Risk factors include bacterial infections, inflammation, systemic diseases such as diabetes mellitus, and abscess formation accompanied with decreased salivary flow [5].

Advanced diagnostic techniques, such as CBCT, and conventional radiographic techniques, such as occlusal radiography, ultrasonography, MRI, CT, and sialendoscopy, can be used in the assessment of sialoliths [6; 7].

Among the symptoms of the sublingual gland is mucus viscosity that is six times more than normal, with a low and antigravity flow pattern [3; 8]. Sialoliths located in the floor of the mouth near the lingual frenum cause discomfort in the patient, followed by purulent discharge in the later stages [3]. The presence of large calculi is associated with MPDS, characterized by pain in the jaws and muscles extending to the temporomandibular joint [9].

The treatment modalities for sialoliths encompass palliative care and duct milking for small calculi, whereas surgical management is necessary for large stones, including procedures such as sialography and sialendoscopy [3; 10].

Urolithiasis has a recurrent nature and is associated with pulp stones in posterior teeth, as evidenced by the patient's medical and dental history; dental radiographs clearly showed pulp stones in many teeth [11]. The association between renal stone and pulp stones is unconclusive. Almadhoon et al. demonstrated a correlation between pulp stones and cardiovascular diseases, but no association was found between pulp stones and renal stones [12]. Conversely, Gabardo et al. reported that patients with pulp stones were nearly twice as likely to present with kidney stones [13], as documented in a recent clinical study [14].

The present case reports a patient with a large calculus accompanied with multiple small, salivary calculi associated with a sialolith in the floor of mouth originating from the sublingual gland duct. The patient also complained of myofascial pain dysfunction syndrome with several painless pulp stones in the posterior teeth.

CASE PRESENTATION

A 54-year-old Egyptian patient presented to the emergency clinic with a chief complaint of discomfort behind the tongue for the past 5 days and an inability to open his mouth for the last 3 months. The medical history revealed a prior occurrence of urolithiasis, with kidney stones surgically removed 18 years ago, followed by a recurrence necessitating a second surgery after 4 years. The patient also underwent sialoadenectomy for the removal of a sialolith in the submandibular gland 24 years ago.

No gross facial asymmetry was detected during extraoral examination. Well-defined solitary nodular swelling was observed intraorally on the floor of the mouth, lingual to the mandibular incisors, measuring 1.5–2 cm in length and width, with a history of increasing size over the last month. The swelling had a yellowish hue, was firm and non-tender upon probing, and had a smooth surface (Fig. 1). The patient had no pus discharge or bleeding, yet he experienced restricted tongue mobility and perceived an abnormality beneath his tongue. The patient's history indicated bilateral tenderness in the masseter, identified as trigger points. During physical examination, gentle pressure was applied to assess the tightness of muscle bands, and postural abnormalities were visually inspected. The patient occasionally experienced clenching and grinding. He was given analgesics such as ibuprofen and applied ice packs for pain relief.

Standard bitewing radiographs were obtained, which revealed generalized pulpal stones or pulpal canal obliteration in the posterior teeth (Fig. 2, *A*). The coronal CBCT image revealed a well-defined radiopaque structure measuring 15 mm x 12 mm in the mediolateral region of teeth # 44 and 45 (Fig. 2, *B*). The sagittal section exhibited a well-defined radiopacity measuring 16 x14 mm in the lingual region of tooth # 42 which was in close proximity to the alveolar bone of teeth # 41 and 42 (Fig. 2, *C*). Similarly, the axial section revealed a well-defined radiopaque structure measuring 14 mm x 12 mm in the lingual bone of tooth #42, which might impede the salivary gland (Fig. 2, *D*).

After data interpretation, the patient was diagnosed with MPDS, and the radiopaque mass was diagnosed as a 2 cm sublingual sialolithiasis accompanied with several pulp stones in molar teeth. The treatment plan was discussed with the patient, who agreed to the surgical removal of the stone, management of MPDS symptoms, and periodic assessment of pulpal stones via periapical X-rays. During this visit, the patient signed a consent form.

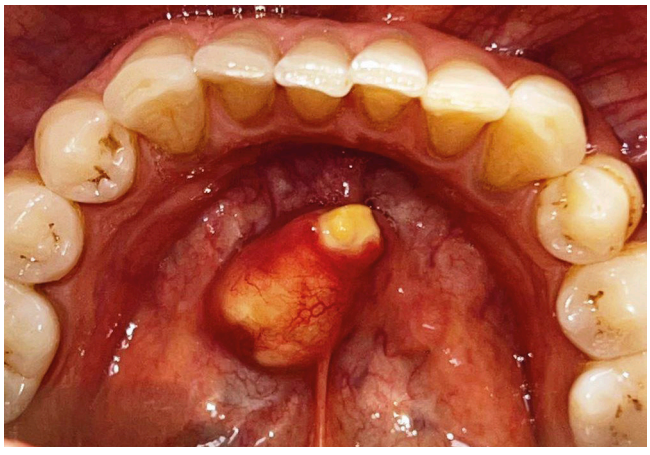

Fig. 1. Intraoral view with appearance of hard swelling lingually to incisors

Рис. 1. Внутриротовой вид с наличием плотного отека (уплотнения) на язычной стороне резцов

At the second appointment, the treatment was conducted via a transoral incision. Local infiltration of anesthesia was initiated, followed by a mucosal incision across the sialolith to expose the area. A single large calculus (Fig. 3, *A*) and many small salivary calculi (Fig. 3, *B*) were easily extracted. The duct was squeezed to ensure the removal of any residual calculi, and salivary flow was carefully assessed. The mucosa was sutured (Fig. 4). The excised portion was sent for histopathological analysis, which confirmed the diagnosis of sialolith.

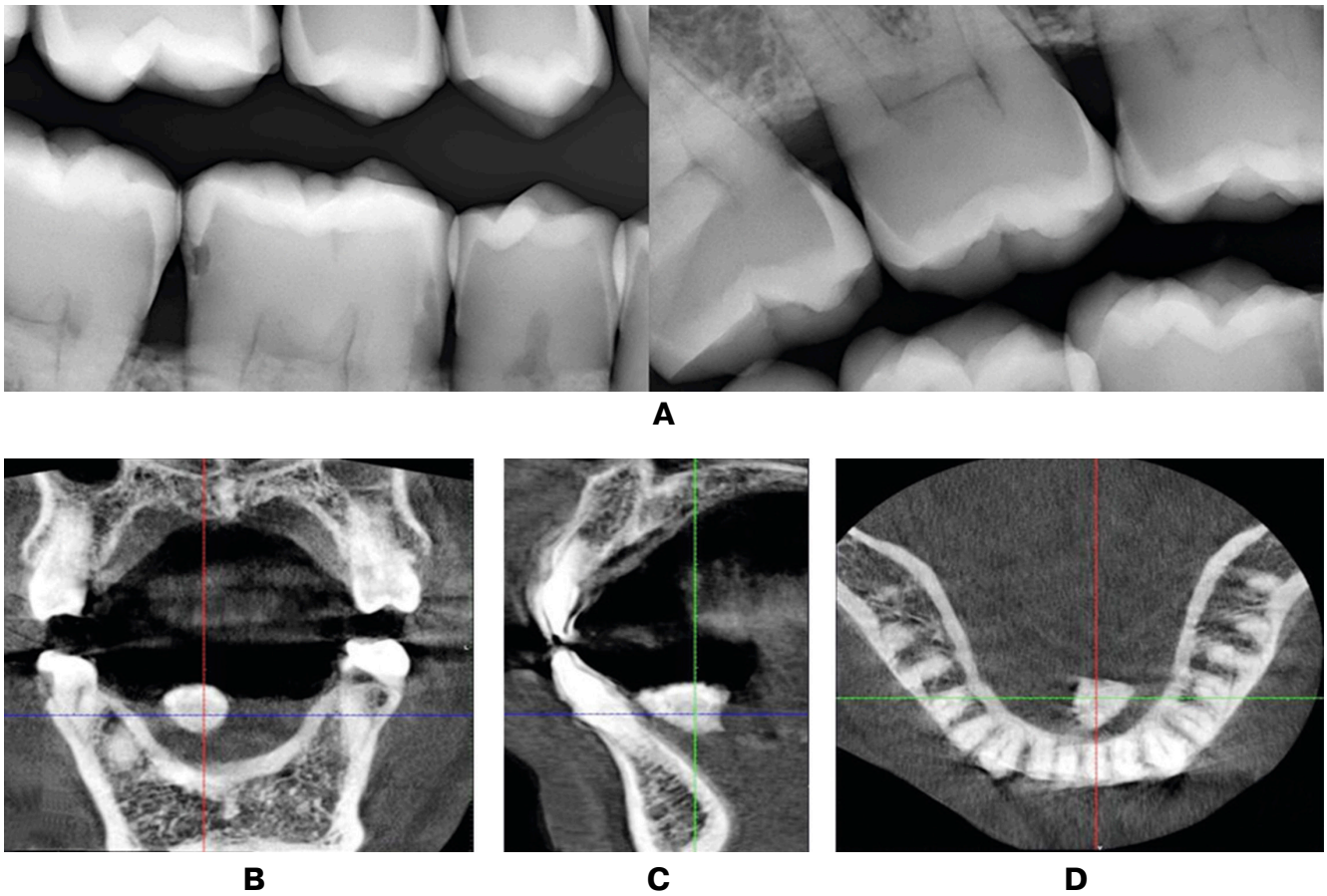

Fig. 2. Bitewing radiographic views, OPG (*A*), coronal (*B*), sagittal (*C*), axial (*D*) CBCT views

Рис. 2. Рентгенограммы прицельные (bitewing), ортопантомограмма (*A*), корональный (*B*), сагиттальный (*C*) и аксиальный (*D*) срезы КЛКТ

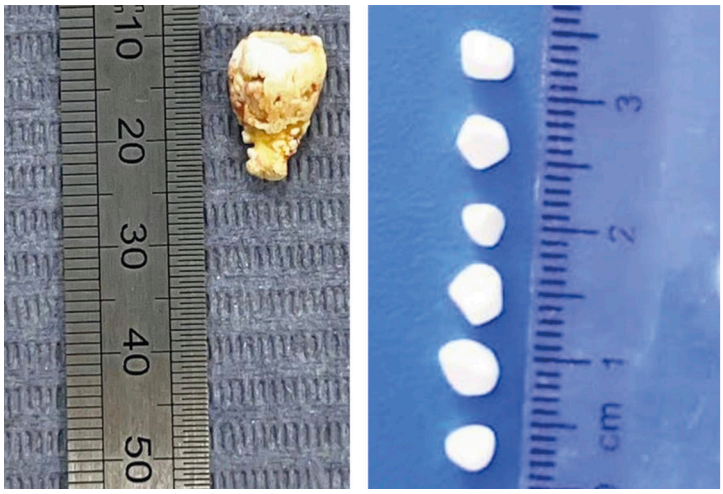

Fig. 3. The big calculus (*A*) and small salivary calculi (*B*)

Рис. 3. Крупный зубной камень (*A*) и мелкие слюнные конкременты (*B*)

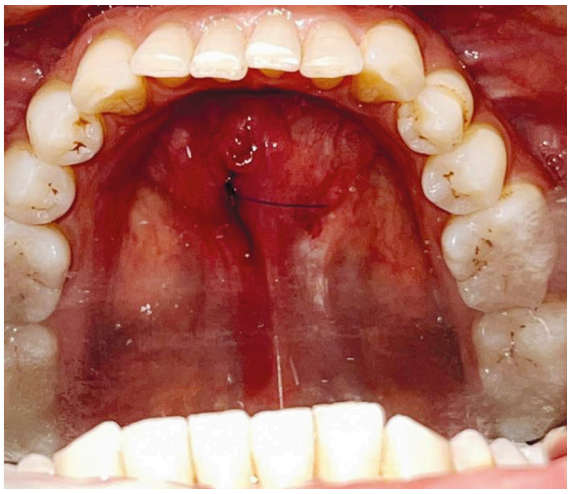

Fig. 4. Postoperative intraoral view after calculus removal

Рис. 4. Послеоперационный внутриротовой вид после удаления конкремента

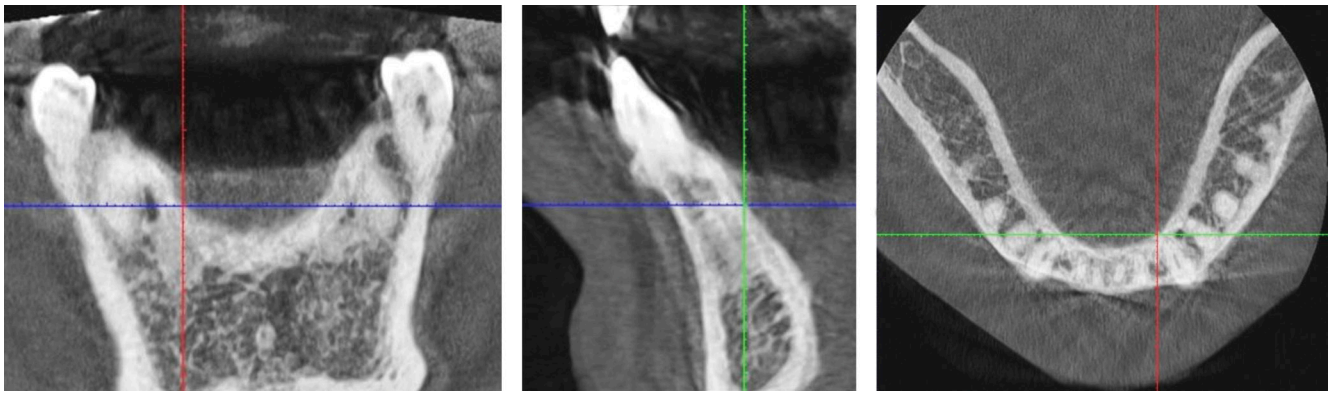

Fig. 5. Postoperative CBCT views, coronal (*A*), sagittal (*B*), axial (*C*)

Рис. 5. Послеоперационные снимки КЛКТ: корональный (*A*), сагиттальный (*B*) и аксиальный (*C*) срезы

MPDS was treated with the administration of an effective muscle relaxant tinzanidine hydrochloride (2 mg; Tilax Tinzanidine, RIADH PHARMA, SA) tablet three times daily. The patient was instructed not to exceed 36 mg throughout a 24-hour period for 30 days. The patient was advised about the potential side effects of the medicine, including nervousness, pain or burning during urination, and unusual fatigue.

Supplementary physical therapy, including stretching to alleviate muscle tightness, was advised as part of oral physiotherapy, and a numbing spray was administered for any pain experienced during stretching. The patient was instructed to avoid consuming hard food and chewing gum. The therapist used elongated strokes along the muscle or applied pressure to specific areas of the patient's muscle to relieve tension. Finally, a 2 mm-thick hard acrylic mouth guard was constructed and delivered to the patient, who was advised to wear it in the maxillary arch for 2–4 h during sleep to treat bruxism due to clenching and grinding habits.

After 15 days, the suture was removed, and the patient reported no post-surgical complaints or complications. Healing was satisfactory, and salivary flow was normal. A postoperative CBCT was conducted, revealing normal proper healing, intact structures, and absence of calculus or salivary calculi (Fig. 5). The patient was booked for a 3-month follow up to manage MPDS and observe any potential recurrence of calculus or salivary calculi.

DISCUSSION

Sialoliths lack a definite etiology, though they are known to form from a nidus of salivary mucin, desquamated epithelial cells, and mineralization of accumulated debris in the salivary duct lumen [3]. Sialoliths are formed by the aggregation of calcium salts, predominantly hydroxyapatite, which consists of calcium phosphate and traces of ammonium and magnesium carbonate [3]. This case presents a male patient with MPDS with a large calculus and multiple small salivary calculi, accompanied with a pulpal canal obliteration in most of the posterior teeth.

Rai and Burman reported that 16% of sialoliths occur in the sublingual gland, with sizes ranging from 5 mm to a maximum of 72 mm [15]. Males are commonly affected, as reported by Pachisia et al. [3; 16]. Similar cases were reported in surgical treatments for large, non-accessible cases of conservative treatment involving sublingual sialolithiasis, with satisfactory outcomes [3; 17–19]. This coincided with the findings of the current case.

Factors such as dehydration, changes in salivary pH and flow, increased calcium content, and physical trauma to the salivary gland duct may predispose individuals to calculus development [3; 20]. The formation of stones is due to a combination of gastric and environmental factors, including risk factors such as high urine calcium levels, medication, obesity, and systemic diseases (like diabetes, hypertension, and gout), as well as insufficient fluid intake and familial predispositions [3; 21]. In the present case, a large sublingual sialolith was detected; however, inspection, palpation, and ductal milking confirm the diagnosis.

The clinical differential diagnosis includes salivary gland tumors, arterial and venous calcifications, phleboliths, calcified lymph nodes, mandibular tori, and osteomas [22]. A sublingual sialolith was detected in this case, which can be considered a rarity. The supported reasons may be the anatomical point, as the duct of the sublingual gland is shorter and possesses multiple openings, and the patient only experienced discomfort with no other complaints. However, in our case, the calculi were around 1.6–2 cm, and the chief complaint was resolved accordingly [23].

The current case involved the surgical treatment of a sublingual sialolith through a transoral approach, which included the resection of the sublingual gland. In the treatment of small salivary calculi, milking or stimulation with sialagogues or lemon juice may be employed for their expulsion. Nevertheless, in our case, surgical excision was needed due to the large size [24]. Sialoliths often form in the central and peripheral stages, where mineral salts and organic substances accumulate through the deposition of organic and inorganic mate-

rials around the central layer. The other theory posits an unknown metabolic phenomenon that results in the precipitation of salivary calcium, increasing salivary bicarbonate levels and altering the solubility of calcium phosphate [25].

The patient in this case had a history of urolithiasis and a previous history of sialolithiasis, with no familial history given. Hemminki et al. evaluated the familial and spousal risks associated with sialo, urolithiasis, and cholelithiasis, and they concluded that familial clustering is unique for each individual [26]. Additionally, the patient previously reported submandibular calculi, indicating a history of recurrent salivary calculi. This coincided with a previously stated observation that 8.9–10% of patients reported recurrent sialoliths with stones such as urolithiasis or nephrolithiasis [15; 27]. Similarly, sublingual sialolithiasis is asymptomatic in terms of pain and swelling unless there is edema, infection, and blockage of salivary flow, as evidenced in this patient.

Pulp stones, calcifications in the dental pulp chamber, are commonly observed in radiographs and associated with systemic disorders such as cardiovascular diseases, diabetes, and nephrolithiasis [28]. Gabardo et al. reported a significant association between pulp stones and kidney stones, suggesting a possible shared systemic predisposition to calcification [8]. However, conflicting evidence exists, with some studies finding no conclusive link between pulp stones and systemic calcifications [13].

CONCLOUSION

This case highlights the rare coexistence of sublingual sialolithiasis, MPDS, and pulp stones. A multidisciplinary evaluation, incorporating surgical removal of calculi, muscle relaxants, and oral physiotherapy, is essential for achieving optimal results in complex cases involving recurrent calcifications and chronic pain. Further research is warranted to elucidate the common mechanisms underlying these conditions.

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AUTHOR'S CONTRIBUTION

The author was fully responsible for the conception and design of the study, data collection and analysis, manuscript preparation, critical revision for important intellectual content, and final approval of the version to be published.

ВКЛАД АВТОРОВ

Автор полностью несет ответственность за замысел и дизайн исследования, сбор и анализ данных, подготовку рукописи, критический пересмотр в части значимого интеллектуального содержания, а также окончательное одобрение версии статьи для опубликования.