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Case report

Megalith of the Wharton's duct: Review of literature and a case report

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Abstract

Sialolithiasis, the most common disorder affecting salivary glands, occurs when calcified formations obstruct the ducts or glands. Typically, these formations, known as sialoliths, measure between 5 and 10 mm; however, those exceeding 15 mm are classified as megaliths. While sialoliths commonly manifest in the submandibular glands, they can develop in any salivary gland duct, with the Wharton's duct of the submandibular gland being a frequent site. These stones, formed from calcified organic material within the secretory system, can lead to pain, swelling, and disruptions in saliva flow. Chronic sialolithiasis, the primary contributor to acute and chronic infections in salivary glands, is closely associated with stone formation. The precise cause remains unclear, yet it is linked to chronic sialadenitis and partial obstruction. On the contrary, megaliths are exceptionally rare and tend to occur more frequently in male patients. The present article presents a case involving a 19 mm long megalith detected at the duct mouth of the left submandibular gland, which was treated through surgical intervention. Additionally, a comprehensive literature review on this specific topic was conducted.

Keywords: **Salivary gland calculi; sialolithiasis; Wharton's duct**

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Introduction

Sialolithiasis, the primary concern affecting salivary glands, involves the development of salivary stones within ducts or glands and is estimated to occur in approximately 1.2% of adults.[1,2] This condition, also called salivary calculi, predominantly manifests in the submandibular gland and its duct, constituting more than 80% of documented cases.[3,4]

The submandibular gland is particularly susceptible to sialolithiasis due to the distinctive characteristics of Wharton's duct, which include its lengthy and convoluted path and the specific properties of saliva.[2,3] Notably, there is a significant gender difference, with males being twice as likely as females to experience this condition. It tends to affect individuals in their fourth to sixth decades of life.[1,4]

While most cases exhibit intermittent pain and swelling, the severity depends on the degree of obstruction and the resulting pressure in the gland, particularly noticeable during meals.[3,4] Radiographically, salivary stones usually appear as visible masses, but not all are discernible through standard X-rays. The diagnostic arsenal incorporates cone-beam computed tomography (CBCT) for assessing the submandibular duct.[3,5]

Giant sialoliths, commonly known as megaliths, represent a rare form of sialolithiasis characterized by salivary stones exceeding 15 mm in any dimension.[6,7] This distinct subset of sialoliths is primarily found in the submandibular gland and its duct, presenting diagnostic challenges due to their infrequency and potential impact on salivary gland function. A comprehensive literature review reveals only 29 reported cases of megaliths in Wharton's duct in the past 22 years, underscoring their rarity and emphasizing the importance of heightened clinical awareness.[6]

This article presents a case involving a megalith, a sizable salivary stone measuring 19 mm in length, detected at the duct mouth of the left submandibular gland. The case was successfully treated through surgical intervention. Additionally, a comprehensive literature review was conducted to provide a detailed exploration of relevant information and insights from existing studies and reports.

Case Report

A 54-year-old man sought treatment at Istanbul Aydın University, Faculty of Dentistry, Department of Oral and Maxillofacial Surgery, due to persistent and painful purulent discharge from the unilateral submandibular ducts. The patient had no significant medical history, and no underlying conditions contributing to sialolith formation were identified.

Upon thorough examination, it was revealed that the issue originated as a minor swelling, evolving over nine months and eventually rupturing, leaving a yellowish-white mass on the right floor of the mouth. The patient underwent discomfort and the release of pus from the affected area. The enlargement exhibited a slow initiation, progressively growing in magnitude. The patient also reported an altered taste sensation for one year, accompanied by occasional pus discharge.

No symptoms of dysphagia or tongue elevation were reported. The patient was conscious, cooperative, and within normal vital sign limits. Pain, described as dull and intermittent, was associated with meals, with the swelling enlarging during and after meals.

Extraoral examination showed a localized swelling in the left submandibular region, exhibiting tenderness and softness upon palpation, with normal overlying skin. Intraoral examination revealed edematous flooring on the left side of the mouth. Bimanual palpation detected a hard mass measuring approximately 2 × 1 cm in the left canine region. The overlying mucosa displayed standard color, except where a break exposed the underlying yellowish mass. Milking of the left submandibular gland resulted in minimal saliva but pus discharge. Notably, the entity has not adhered to underlying structures (Figure 1).

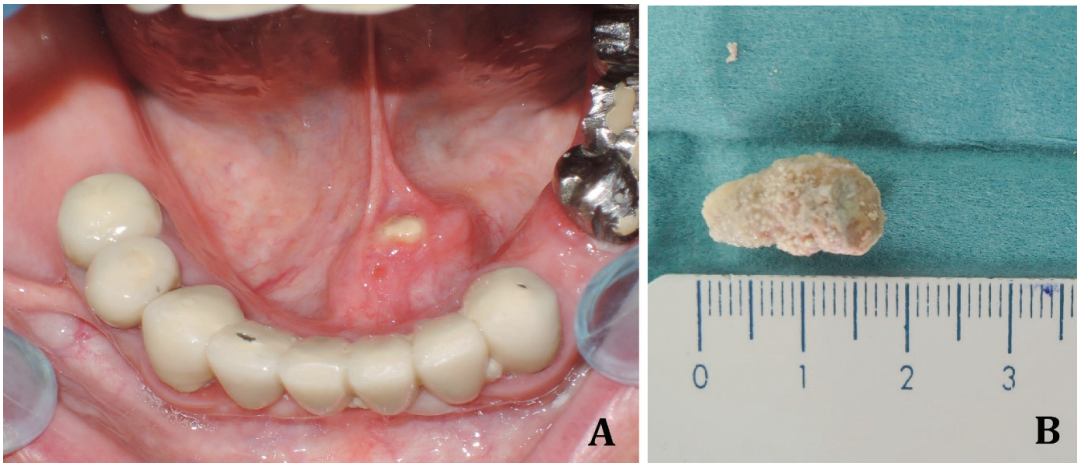

Figure 1. *(A) Preoperative intraoral image, (B) Appearance after the sialolith removal, measuring 19 mm in length and 12 mm in diameter.*

The mandible's orthopantomogram (OPG) showed a calcified mass resembling an impacted canine tooth in the left canine region. The OPG displayed a radioopaque mass in the left body of the mandible, measuring approximately 2 × 1 cm, extending 1 cm above the apices of teeth 33 and 34 and 4 cm below the lower border of the mandible. Subsequent computerized tomographic (CT) scans confirmed the presence of a cylindrical and elongated sialolith within the left Wharton's duct, leading to the diagnosis of the left submandibular duct sialolith (Figure 2).

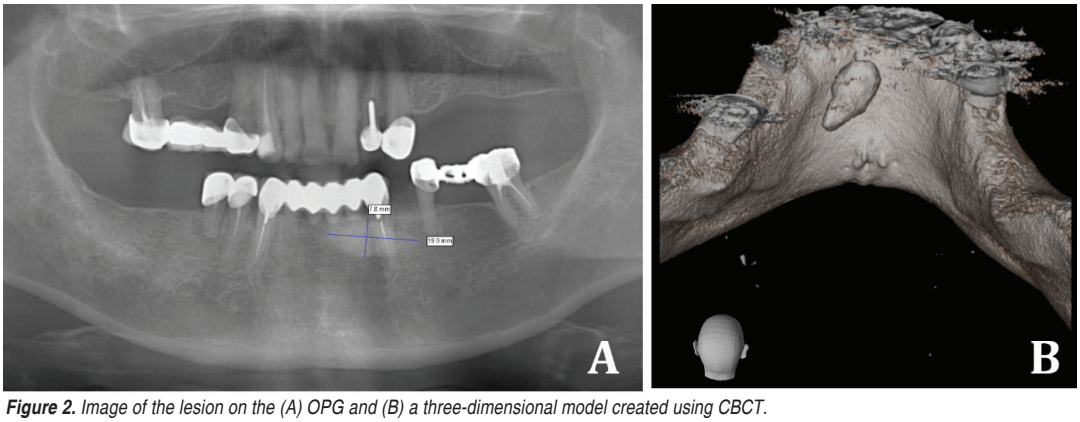

Figure 2. *Image of the lesion on the (A) OPG and (B) a three-dimensional model created using CBCT.*

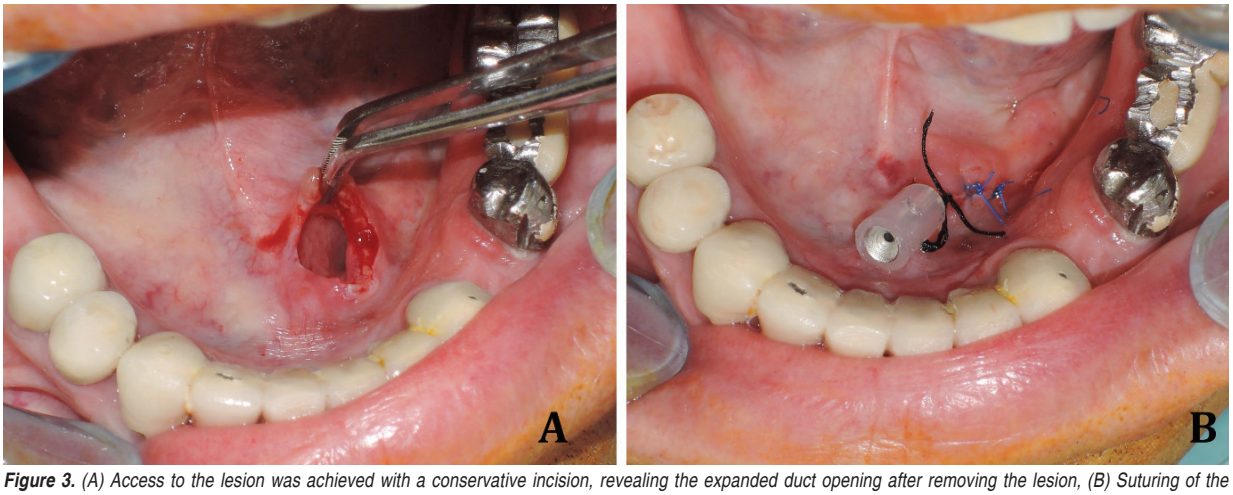

Figure 3. *(A) Access to the lesion was achieved with a conservative incision, revealing the expanded duct opening after removing the lesion, (B) Suturing of the incision line with polypropylene sutures and fixation of the catheter placed to preserve canal integrity with silk stitches.*

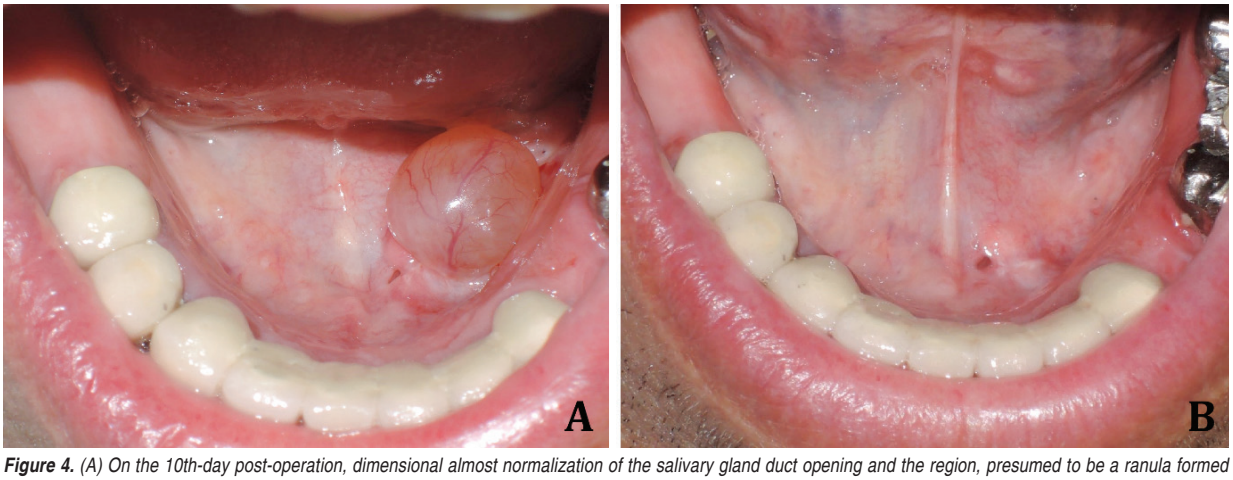

Figure 4. *(A) On the 10th-day post-operation, dimensional almost normalization of the salivary gland duct opening and the region, presumed to be a ranula formed after surgical trauma, (B) Intraoral view in the first month after the operation.*

With informed consent, the giant sialolith was surgically removed via an intraoral approach under local anesthesia, using transoral sialolithotomy and sialodochoplasty for minimal invasiveness. Upward and medial pressure was applied to stabilize the floor of the mouth and accurately locate the sialolith. A longitudinal incision was made over the stone, followed by meticulous mucosal dissection to expose and mobilize the sialolith. The wound was closed only at the mucosal layer. Examination post-removal revealed a hard, yellow, elongated stone measuring 19 mm in length and 12 mm in diameter. A drainage catheter was inserted postoperatively to manage purulent discharge. On the tenth day post-surgery, a ranula formation was noticed in the area, believed to be a result of surgical trauma. The ranula was addressed through a straightforward surgical intervention (Figure 3).

Postoperatively, the patient received antibiotics (amoxicillin + clavulanate, 2000 mg/day) and an analgesic (Dexketoprofen 25 mg/day) for five days. The drainage catheter was removed after 48 hours. Reassessment on the seventh postoperative day indicated normalcy in the left submandibular gland, with clear salivary flow from the healed duct (Figure 4). After a three-year follow-up, it was observed that the patient showed no symptoms, displaying adequate glandular function and normal, uninterrupted salivary flow.

Discussion

In the literature review using the PUBMED database, a total of 59 case reports were found in 51 articles as a result of the search with the terms "Giant Submandibular Sialolith," "Large Submandibular Sialolith" and "Submandibular Megalith" among the articles published between 2000 and 2023 (Table 1).

When the dimensions of the sialoliths reported in the case reports are listed according to the largest value in the dimensions, the largest sialolith was reported as 72 mm by Rai and Burman[8] in 2009. The average size of 59 cases was found to be 32.5 mm. When the area between 15-35 mm was divided into megaliths, and those over 35 mm were divided into giant sialoliths, the average size of 35 megalith cases was found to be 25.2 mm, and the average size of 24 giant sialoliths was found to be 43.1 mm. While the general average age of the cases is 47.5, it is 46.5 for giant sialoliths and 48.1 for megaliths. Fifty-one of the cases are male, and only 8 of them are female. This age range and male dominant gender status appear to be compatible with the data in the literature.[1,9]

Sialoliths, common in sialadenitis, are calcified formations in salivary ducts or glands. They consist of glycoproteins, mucopolysaccharides, and inorganic substances. These stones form around a central core, appearing as radiopaque masses mostly in submandibular glands.[9,10]

Giant sialoliths or megaliths are a rare subset of salivary gland disorders. In the literature, stones exceeding 15 mm in any dimensional value are referred to as megaliths, while those surpassing 35 mm are classified as giants.[11,12]

They mainly affect males, with the submandibular gland being the primary location in 94% of cases.[1,13] The etiology and pathogenesis align with conventional sialolithiasis theories, involving the deposition of calcium salts around an initial organic nidus.[2,13] Diagnosis involves various radiographic studies, and treatment plans vary based on the stone type and location.[7,14] Options include simple approaches, surgery, or advanced methods like shock wave treatment and sialoendoscopy.[3,15]

Sialolithiasis, a condition characterized by the formation of salivary gland stones, is prevalent among individuals between the ages of 30 and 50, and it tends to affect males more frequently.[1,9] It is notably rare in children. The submandibular duct or gland (80%) is the most commonly affected, followed by the parotid gland (18%), and rarely the sublingual gland (2%).[2,16,17]

The etiology and pathogenesis of sialoliths are intricate and not completely comprehended, reflecting the complex and multifaceted nature of their origin and development. Sialoliths develop when calcium salts accumulate around an initial organic core. This core consists of altered salivary mucins, bacteria, and shed epithelial cells.[11,15,18] Traditional theories propose a

Table 1. *Evaluation of 59 cases included in 51 articles obtained as a result of scanning between 2000 and 2023 using the PUBMED database*

REFERENCE	SIZE (mm)	AGE	GENDER
Rai and Burman [8]	72	60	M
Chaidas *et al.*[6]	58	72	M
Rauso *et al.*[41]	56	56	M
Shahoon *et al.*[42]	55	25	M
Lim *et al.*[43]	50	59	M
Sakthivel *et al.*[44]	50	42	M
Bodner[45]	45	50	M
Rodrigues *et al.*[21]	45	48	F
Akinyamoju and Adisa[32]	44	54	M
Singh *et al.*[15]	42	40	M
Fowell and MacBean[46]	41	58	M
Iwai *et al.*[47]	41	53	M
Abraham *et al.*[12]	40	45	M
Pandarakalam *et al.*[48]	40	68	M
Mathew *et al.*[49]	39	50	M
Omezli *et al.*[2]	37	35	M
Ledesma-Montes *et al.*[50]	36	34	M
Arslan *et al.*[24]	35	42	M
Chan and Patel[51]	35	27	M
El Gehani *et al.*[52]	35	41	M
Emir *et al.*[53]	35	57	M
Goh *et al.*[54]	35	34	M
Iqbal *et al.*[16]	35	55	M
Raveenthiran *et al.*[55]	35	10	F
Krishnan *et al.*[56]	34	41	M
Alkurt and Peker[57]	31	65	M
Bhullar *et al.*[58]	31	45	M
Bodner[45]	30	45	M
Bodner[45]	30	25	M
Cottrell *et al.*[59]	30	75	M
Demircan and Isler[60]	30	62	M
Oliveira *et al.*[22]	30	42	M
Yıldırım[61]	30	56	M
Alkurt and Peker[57]	28	45	M
Al Hussona[62]	28	21	M
Gupta *et al.*[4]	28	48	M
Kumar *et al.*[63]	28	29	M
Mohsin *et al.*[14]	28	47	M
Shetty and Sharma[64]	27	50	M
Biddle and Arora[65]	26	48	M
Bodner[45]	26	46	M
El Gehani *et al.*[52]	25	32	F
Gadve *et al.*[66]	25	45	M
Krishnan *et al.*[56]	25	32	F
Mao *et al.*[11]	25	75	F
Soares *et al.*[67]	25	54	F
Thong *et al.*[20]	25	71	M
Rivera-Serrano and Schaitkin[68]	23	69	M
Ungari *et al.*[69]	23	70	M
Boffano and Gallesio[36]	22	48	M
Graziani *et al.*[70]	22	61	M
Arunkumar *et al.*[71]	20	10	M
Nilesh *et al.*[72]	20	37	M
Oteri *et al.*[13]	20	40	F
Bodner[45]	19	61	M
Brooks *et al.*[7]	19	63	M
Bodner[45]	18	61	M
Ben-Shoshan and Lacroix[73]	17	16	M
Oteri *et al*[13]	15	51	F
MEAN	**32.5**	**47.5**	**51M, 8 F**

mm: Milimeter, M: Male, F: Female

two-phase process: forming a central core through salt precipitation and layered deposition. Metabolic factors and variations in the sphincter-like mechanism of the duct may contribute. Reduced salivary flow, stagnation, dehydration, changes in salivary pH, foreign bodies, and physical trauma further contribute.[9,14] Sialoliths are not exclusive to systemic issues and can also be linked to conditions like gout, Sjögren's syndrome, the use of anticholinergics and antisialogogues medications, local trauma, head and neck radiotherapy, advanced age, and renal impairment, all of which can contribute to the formation of sialoliths.[9,19]

Clinical features of sialolithiasis encompass symptoms such as pain, swelling, and tenderness, particularly heightened during meals when there is an increase in salivary flow. Secondary infections may lead to fever, redness, and pus discharge. Common indicators include tenderness, trismus, decreased salivary flow, and purulent discharge at the duct's opening. Palpation along the duct may reveal a sialolith. Some cases may be asymptomatic.[19-22]

The Mealtime Syndrome presents as moderate to severe pain and swelling in major salivary glands, particularly the submandibular gland, triggered by meal-related salivary flow stimulation. Obstruction by sialoliths leads to saliva accumulation, causing pain and swelling.[15,23] Patients may experience difficulty swallowing, tenderness, trismus, decreased salivary flow, and occasionally, purulent discharge. Careful clinical examination and radiographic imaging are essential for diagnosis and management.[23]

Radiographic features play a crucial role in the diagnosis of sialolithiasis, offering essential details about the size, location, and mineral composition of the calculi. Submandibular gland sialoliths are usually visible on conventional radiographs as they are radiopaque in 80–94% of cases. The elongated or smooth cylindrical appearance of ductal sialoliths and the round or oval configuration within the gland help characterize them.[24,25] However, sialoliths might be too small or insufficiently mineralized in the early stages to show up on X-rays. Radiolucent submandibular sialoliths, which do not appear clearly on X-rays, pose a diagnostic challenge and are documented in 20% to 43% of cases.[12,23] Various imaging methods, including sialography, xeroradiography, ultrasonography, scintigraphy, and advanced techniques like computerized tomography and magnetic resonance imaging, complement conventional radiography, providing a comprehensive diagnostic approach.[3,5,14,25]

The diagnosis of sialoliths entails a comprehensive evaluation, including an analysis of patient history, clinical examination, and the utilization of imaging methods. Symptoms include pain, swelling, and issues during or after meals. Clinical examination helps identify potential problems. Radiographic methods like panoramic and occlusal radiographs are crucial for visualizing calcified sialoliths. Advanced techniques such as sialography, ultrasound, computed tomography, and magnetic resonance imaging provide additional details.[14,25,26] Sialography is useful for radiolucent stones, while ultrasound helps locate stones within the gland. These tools together provide a precise understanding of the sialolith's characteristics, guiding personalized management plans.[8,27]

Histopathologic features of sialoliths reveal gross characteristics such as rigid calcified masses exhibiting concentric layers and a central core. The stones, mainly made up of glycoproteins, mucopolysaccharides, and cellular debris, may exhibit concentric layers surrounding a central core of amorphous material.[28,29] Microscopically, the calcified mass reveals a layered structure, with microcrystalline apatite or whitlockite as the primary minerals. If the associated duct is removed, there may be evidence of squamous, oncocytic, or mucous cell metaplasia. Periductal inflammation is a common finding, indicating the interaction between stone formation and the glandular environment.[30,31]

The prognosis for individuals with sialoliths is generally favorable, especially when prompt and appropriate management is implemented. Small stones treated conservatively often lead to complete resolution.[22,32,33] Larger stones may require surgical removal or advanced techniques but are also associated with favorable outcomes. Surgical removal of the gland may be necessary in cases of chronic inflammation or recurrent infections, potentially affecting salivary function. Early diagnosis and intervention are crucial to prevent complications.[3,9,14,32-34]

The management and treatment of sialoliths are contingent upon factors such as size, location, and the presence of multiple stones. For small, single stones, conservative measures such as applying local heat, massaging the gland, and using sialogogues may be employed. Mechanical removal or sialolithectomy may be attempted for stones located at the periphery. Sialolithectomy, involving intraoral ductotomy, is an option for stones in specific ductal regions. Surgical removal, either intraorally or extraorally under general anesthesia, becomes necessary for multiple stones, those near the gland hilum, or when recurrent infections compromise the gland.[18,33-37]

Advanced, non-invasive techniques like Extracorporeal Shortwave (ESW) and Intracorporeal Endoscopic lithotripsy (sialoendoscopy) provide alternatives for fragmenting stones without major surgery. ESW lithotripsy uses shock waves externally, while sialoendoscopy delivers shock waves directly to the stone surface, allowing for precise intervention.[18,33,38-40] Tailoring the approach based on individual sialolith characteristics helps clinicians select the most appropriate management strategy, emphasizing a personalized approach for optimal outcomes.

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Wharton kanalı megaliti: Literatür derlemesi ve bir olgu sunumu

Özet

Tükürük bezlerini etkileyen en yaygın hastalık olan siyalolitiyazis, kalsifiye oluşumların kanalları veya bezleri tıkamasıyla ortaya çıkar. Tipik olarak, sialolit olarak bilinen bu oluşumlar 5 ila 10 mm arasındadır; ancak 15 mm'yi aşanlar megalit olarak sınıflandırılır. Sialolitler herhangi bir tükürük bezi kanalında gelişebilirken, en sık olarak submandibular bezlerde, özellikle de submandibular bezin Wharton kanal ağzında ortaya çıkar. Salgı sistemi içindeki kalsifiye organik materyalden oluşan bu taşlar ağrıya, şişmeye ve tükürük akışında bozulmalara yol açabilir. Tükürük bezlerindeki akut ve kronik enfeksiyonlara birincil katkıda bulunan kronik siyalolitiyazis, taş oluşumuyla yakından ilişkilidir. Kesin nedeni tam olarak bilinmemekle birlikte, kronik siyaladenit ve kısmi tıkanıklıkla bağlantılıdır. Aksine, megalitler son derece nadirdir ve erkek hastalarda daha sık görülme eğilimindedir. Bu makalede, sol submandibular bezin kanal ağzında tespit edilen ve cerrahi müdahale ile tedavi edilen 19 mm uzunluğunda megalit içeren bir olgu sunulmuştur. Ayrıca, konuyla ilgili kapsamlı bir literatür taraması yapılmıştır.

Anahtar Kelimeler: Siyalolitiyazis; tükürük bezi taşları; Wharton kanalı

