

# Sublingual Bezde Dev Tükürük Bezi Taşı

## *Giant Sialolith of Sublingual Gland*

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### Özet

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Tükürük bezi taşı majör tükürük bezlerinin yaygın bir hastalığıdır ve sıklıkla submandibular bezde fakat nadiren sublingual bezde görülür. 2/1 erkek/kadın oranıyla birlikte erişkin popülasyonun 1000’de 12’sini etkiler. Tükürük bezi taşı, özellikle yemek sırasında tekrarlayan ağrılı şişkinliklere sebep olarak tükürük bezi tıkanıklığının yaygın bir nedenidir ve disfaji geniş lezyonlarda görülebilir. Literatürde büyük sublingual tükürük bezi taşı nadiren rapor edilmiştir. Amacımız, mevcut tanı usulleri ve tedavi seçeneklerini gözden geçirerek, 23cm uzunluğunda ölçülen sublingual tükürük bezi taşı için sunulan 41 yaşında bayan hasta olgusunu rapor etmektir.

**Anahtar Kelimeler:** Büyük, Tükürük Bezi Taşı, Sublingual, Tedavi, Tükürük Bezi.

### Abstract

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*Sialolithiasis is a common disease of the major salivary glands and frequently seen in the submandibular gland but rarely seen in the sublingual gland. It affects 12 out of 1000 in the adult population with a male/female ratio of 2/1. Sialolithiasis is a common etiology of salivary gland obstruction, causing recurrent painful swelling (especially while eating) and dysphagia can be present in extended lesions. With regard to the literature, large sublingual sialoliths have rarely been reported. Our aim is to review the literature and report a case of a 41-year-old woman, who presented due to a sialolith in the sublingual gland measuring 23 mm in length, with review of the various available diagnostic modalities and treatment options.*

**Keywords:** Large, Sialolithiasis, Sublingual, Treatment, Salivary Gland

## Introduction

Sialolithiasis is the most common disease of the salivary glands and is accepted as the most frequent cause of acute or chronic infection of the salivary gland. While sialoliths can occur in any salivary gland or at any age, they generally develop after the age of 25, with prevalence in adult population of 12/1000.<sup>1</sup> Patients with sublingual sialoliths can experience pain, swelling and/or speech difficulty, but dysphagia is normally present only in extended lesions.<sup>2</sup> It may be palpated in the salivary duct only when it is of considerable size.

As the Wharton duct is longer and with larger caliber and lies against gravity, the saliva, which has high mucin and calcium content, flows with slow rates. This, in addition to the higher alkalinity of the saliva found there, facilitates stone formation in the sub-mandibular gland, but it is rarely seen in the sublingual gland.<sup>3</sup>

We review the literature and report a case of a sialolith in the sublingual gland measuring 23 mm in length with review of the various available diagnostic modalities and treatment options.

## Case Report

A 41-year-old female with a previous submandibular gland excision due to left submandibular sialolithiasis, described a left side pain related with eating, which began 5-6 months prior to her visit and increased in a stepwise fashion in the postoperative period. The patient began to feel stiffness in tongue movement over the preceding few weeks. Per-oral examination and bimanual palpation revealed a large, tender, yellowish and firm mass, which had displaced the tongue laterally, in the left floor of the mouth over the sublingual region. The ductal opening was normal and no discharge was detected, however the sialolith was seen protruding from ductal opening. On the basis of clinical findings, a diagnosis of left sublingual gland sialolith was made.

This left sublingual gland sialolithiasis was treated with excision of both the left sublingual gland and stone via a transoral approach under general anesthesia (Figure 1). The sialolith recovered measured approximately 23 mm in length. (Figure 2) No postoperative complications were noted. This study

features human subjects, we have read the Helsinki Declaration and have followed the guidelines in this investigation. We explained the scientific importance of the patient's disease to her, and she provided consent for publication of her case in scientific literature.



Figure 1: The intraoral view of sialolith in submandibular duct.



Figure 2: The sialolith was delivered out through the duct. The calculus was oval, tan in color and had a granular surface. The calculus was surrounded by white fibrous tissue.

## Discussion

The etiology and pathogenesis of the sialolithiasis are still unclear.<sup>4</sup> The defects in mucin formation, electrolyte concentration disorder, hyposalivation, increase the viscosity of saliva, ductal dilatation, metabolic disorder, infection and rise in the mucin part of secretion are all contributing factors to the etiology. It is thought that the sialoliths are formed as a result of accumulation of the calcium salt around the organic core, which is composed of mucin, bacteria and epithelial cells.<sup>5-6</sup>

Sialolithiasis is frequently seen in submandibular gland, but

sublingual sialoliths are very rarely reported in the literature.<sup>7</sup> Clarence P. et al reported an unusual case of sialolithiasis of the sublingual gland presenting in a 35-year-old female. They removed the right sublingual salivary gland, together with the five stones (the largest measuring 7x6x4 mm)<sup>7</sup>. Liao LJ et al studied a case of a 50-year-old male with left sublingual gland sialolithiasis who was treated with excision of the left sublingual gland and the stone via a transoral approach.<sup>8</sup> Ki Hwan Hong et al evaluated a case of multiple sialoliths arising in the sublingual gland in a 14-year-old female; the diameter of the largest one was 9 mm.<sup>9</sup> Güngörmüş et al reported the largest single sublingual gland calculus in the literature. In this case, the giant sialolith was completely encased in the glandular substance, and the stone was a single calcified mass about 32 mm in diameter.<sup>10</sup>

Sialoliths may consist of a single or multiple stones in any major salivary gland, however 70–80% of reported patients present with a single stone sialolith. Lustmann et al indicated that one sialolith was present in 75.3% of cases, two in 15.6%, three in 2.9% and four to eight in 6.2%.<sup>5</sup> In our study, the patient had one single sialolith measuring 23 mm in size.

Salivary gland stones become symptomatic when the growing stone causes increasing obstruction of salivary secretion.<sup>11</sup> Our patient described a left side pain, related with eating, which began 5-6 months ago and increased stepwise.

The treatment of sialolithiasis varies depending on the location of stone, duration of symptoms, frequency of the recurrence and size of the sialolith. Some conservative methods like proper hydration of the patient, moist warm heat application and massaging the gland in combination with sialogogues can be performed for treatment of small sialoliths.<sup>12</sup> Whenever the stone can be palpated intraorally, it is best to remove it through an intraoral approach from the duct.<sup>12,13</sup> The duct should be isolated and a longitudinal incision made into the duct (over the stone) to retrieve it. The choice of surgical approach to access the sialolith and the consideration for preserving the salivary gland requires careful evaluation when dealing with large sialoliths of a remarkable size.<sup>14</sup> Newer treatment modalities such as extracorporeal shock wave lithotripsy and more recently the use of endoscopic intracorporeal shockwa-

ve lithotripsy are effective alternatives to conventional surgical excision for smaller sialoliths.<sup>15</sup> The sialolith of our case was also treated with excision of the left sublingual gland and the stone via transoral approach under general anesthesia.

### Conclusion

In general, sublingual gland sialoliths are considered rare. Patients with sublingual sialolith experience pain, swelling and/or speech difficulty commonly but dysphagia is present in extended lesions. To our knowledge, our patient is the second case of a single large sialolith 23 mm size in sublingual gland reported in literature.

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