International Journal of Otorhinolaryngology and Head and Neck Surgery
*Çeliker M et al. Int J Otorhinolaryngol Head Neck Surg. 2017 Jul;3(3):728-731*
http://www.ijorl.com
pISSN 2454-5929 | eISSN 2454-5937

DOI: http://dx.doi.org/10.18203/issn.2454-5929.ijohns20173056

**Case Report**

# Sialolithiasis and deep neck infection after the submandibular gland excision

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**Received:** 04 April 2017
**Accepted:** 10 May 2017

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## ABSTRACT

Sialolithiasis is a common disease and mainly affects the submandibular glands and the Wharton's duct. Recurrent sialolithiasis is a rare condition. Despite submandibular gland excision, sialolithiasis which requires recurrent sialolithectomy causing also deep neck infections is even rarer. Herein, a 57-year-old female patient, who had recurrent sialolithiasis in Wharton's duct despite submandibular gland excision and sialolithectomy performed 10 years ago and sialolithectomy for Wharton's duct calculi performed 2 years and one year ago via transoral approach, is presented. The patient had also deep neck infection requiring hospitalization and underwent transoral sialolithectomy and marsupialization of the duct after medical treatment. The present case study aimed to present with radiological modalities the recurrent sialolithiasis also causing deep neck infections despite submandibular gland excision, and to indicate the causes of recurrence and the points to be remembered for prevention.

**Keywords:** Sialolithiasis, Sialolithectomy, Deep neck infection

## INTRODUCTION

Sialoliths are calcified concrements that develop in the parenchyma or ducts of the salivary glands and are among the most common causes of obstructive sialadenitis. The majority of sialoliths (84%) are located in the submandibular gland, particularly (90%) in the Wharton's duct.[1] Sialoliths become symptomatic when they block salivary flow or cause an infection. Most patients present with swelling and pain of the salivary gland during meals.[1]

The diagnosis of sialolithiasis is made based on clinical and radiological examinations. The majority of stones is radio-opaque and can be demonstrated by radiography in approximately 80% of patients.[1] Ultrasound imaging, as an easy and accessible method, is also preferred in the diagnosis of sialoliths. In addition, magnetic resonance (MR) sialography provides more definite information on the location and width of stenosis, while sialendoscopy enables direct visualization of the ductal system.[2]

The treatment of choice for sialoliths is the removal of calculi (sialolithectomy) by preserving the functions of the salivary gland. Most of the sialoliths can be removed by simple intraoral approach under local anesthesia. Total excision of the salivary gland is required in some instances depending on the location and size of the calculi.[1]

The present paper reports a 57-year-old female patient, who had recurrent sialolithiasis in Wharton's duct despite submandibular gland excision and sialolithectomy performed 10 years ago and sialolithectomy for Wharton's duct calculi performed 2 years and one year ago via transoral approach. The patient had also deep

neck infection requiring hospitalization and underwent transoral sialolithectomy and marsupialization of the duct after medical treatment.

## CASE REPORT

A 57-year-old female patient, who presented to our clinic in December 2015 with difficulty in swallowing and opening her mouth as well as a painful swelling in the sublingual region, was hospitalized with a pre-diagnosis of Wharton's duct abscess. Her medical treatment, including intravenous infusion of Ampicillin sulbactam (3x1 g/day) and metronidazole (3x500 mg/day) for 7 days, was started.

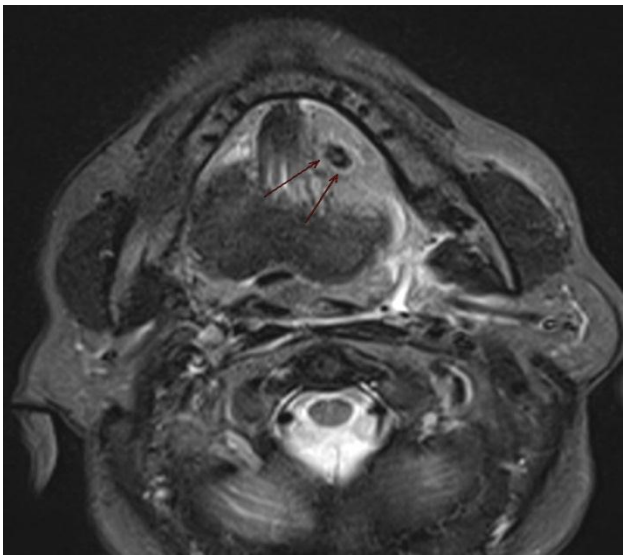

**Figure 1: T1-weighted and T2-weighted axial images showing a hypo intense focus on the left side consistent with the distal part of the Wharton's duct.**

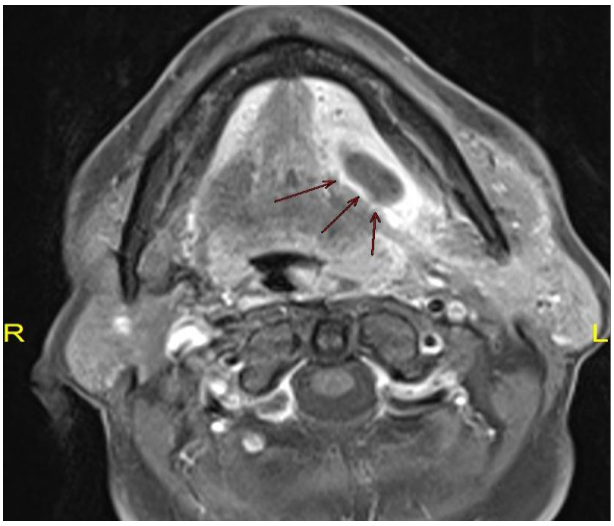


**Figure 2: Post-contrast axial and sagittal T1-weighted MR images of an abscess formation proximal to the calculus with peripheral contrast uptake at the level of Wharton's duct.**

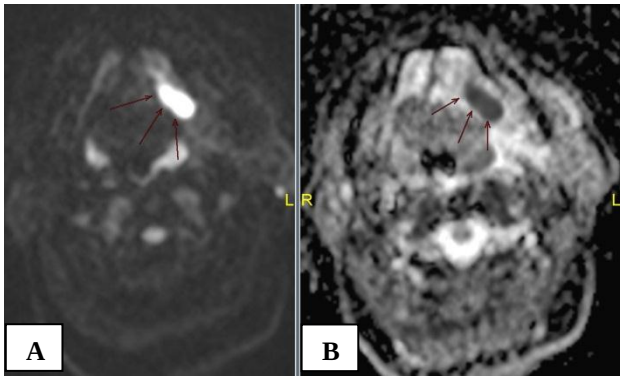


**Figure 3: Echo-planar imaging (A) and apparent diffusion coefficient images (B) showing diffusion restriction in the lesion consistent with abscess.**

Her medical history revealed submandibular gland excision and sialolithectomy performed 10 years ago and transoral sialolithectomy performed 2 years and one year ago. On her physical examination, a hard mass was palpated close to the distal portion of the Wharton's duct on the left side of the floor of the mouth. Left submandibular gland could not be detected on her ultrasound examination as it was removed before. Wharton's duct was dilated on the left side and the lumen showed a heterogeneous echo pattern with a 13 mm echogenic calculus at the distal part of the long axis. Adjacent sublingual soft tissue planes also showed a heterogeneous echo secondary to inflammation. With the prediagnosis of deep neck infection in the patient, who had submental and submandibular lesions, MRI was performed to determine the extensiveness of the infection and to evaluate abscess development in detail (Figures 1, 2, 3). MRI demonstrated hypointensity consistent with calculus in the Wharton's duct and, with a prediagnosis of sialolithiasis, non-contrast-enhanced computed tomography (CT) was performed (Figure 4).

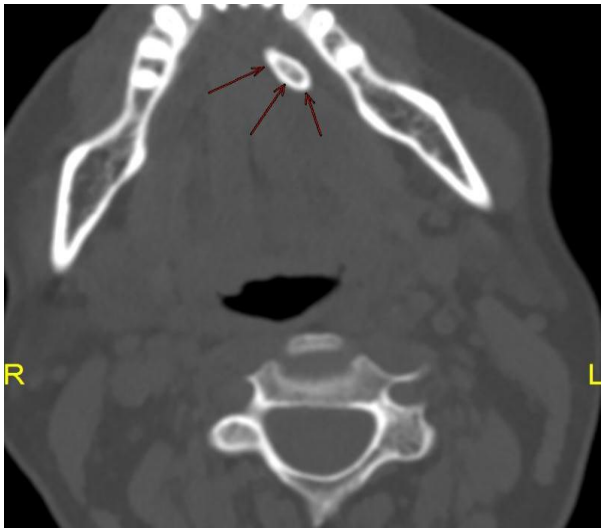


**Figure 4: Non-contrast-enhanced axial CT image of the dense calculus at the floor of the mouth.**

The patient underwent surgery under general anesthesia for recurrent stone formation in the Wharton's duct. Wharton's duct was accessed via transoral incision on the mucosa of the hard mass, which was palpated in the distal portion of the duct. The duct was dissected and a 13 mm calculus was excised. The duct was marsupialized and re-implanted in the floor of the mouth using 4/0 vicryl. Patency of the duct was maintained by periodic irrigation after the surgery. Hydration and sour foods were recommended to the patient after hospital discharge.

This study was approved by the Ethics Committee of Recep Tayyip Erdoğan University in accordance with the Decleration of Helsinki. The study subject had given informed consent and the data of the subject after receiving permission.

## DISCUSSION

Higher incidence of sialoliths in the submandibular gland is attributed to anatomic and physiological factors. Anatomically, the Wharton's duct is the longest among all salivary gland ducts, it has a curved course and it is narrower. Sharp curves of the duct form a basis particular for the development of calculus.

The sublingual gland is connected with the Wharton's duct via the Bartholin's duct or directly into the floor of the mouth.[3] Therefore, the connection between the sublingual gland and the Wharton's duct is hypothesized to provide an anatomical basis for sialolith formation. In addition, the sublingual gland is predominantly a mucus secreting gland, and the viscous saliva in the relatively stagnant environment within the residual part of the Wharton's duct further facilitates sialolith formation.[4]

Additionally, submandibular gland saliva is more viscous, with an alkaline pH and a higher concentration of calcium. Although the etiology of sialoliths is unclear, viscosity of saliva and slow salivary flow are thought to contribute to the development of sialoliths.[1] Mucous plugs or salivary organic material acting as a nidus has been reported for some sialoliths. Boynton and Lieblich reported a case with sialolith in the distal end of the Wharton's duct in which facial hair acted as a nidus to initiate the formation of the sialolith.[5] It is estimated that sialoliths recur in 1-10% of the patients.[1] Sialolithiasis recurred for three times in the present case. Sialolith has been reported as the reason for surgery in nearly half (46%) of the patients who underwent submandibular gland excision.[6] Medical history of the present case revealed submandibular gland excision due to sialolithiasis 10 years ago. Despite this, recurrent sialolith formation was observed in the patient. One of the reasons for recurrent sialoliths despite submandibular gland excision may be some ductal calculi being overlooked during excision.[7] Therefore, in order to avoid this situation, detailed examination using imaging techniques such as ultrasound and CT should be performed for the detection of ductal calculi before the surgery. In addition, dilatation of the submandibular duct punctum, as well as irrigation for the removal of calculi that may be left behind, are recommended at the end of surgical procedure while performing submandibular excision.[8]

Infection of the surrounding soft tissues may accompany sialoliths. Medina et al reported a case with a sialolith and sialadenitis in the Wharton's duct complicated with submandibular soft tissue infection.[9] The present case had deep neck infection involving the sublingual soft tissue. Gallego et al reported a case with small calculi in the submandibular gland and obstructive sialadenitis, of whom histopathological examination revealed carcinoma in situ of Wharton's duct, and suggested that chronic inflammation might have been a probable factor for the development of malignancy.[10]

In conclusion, the case presented herein is important, as the patient developed recurrent sialolithiasis despite submandibular gland excision and had also deep neck infection. As the Wharton's duct can promote the sublingual gland drainage, it is important for the prevention of sialadenitis of the sublingual gland to preserve a part of the Wharton's duct after the excision of the mandibular gland for sialolithiasis. A careful massage of the duct from the bottom to the top may help to remove any undetected calculi; and combination of the massage with sialodochoplasty of the duct of the submandibular gland may reduce the possibility of sialolith and sialadenitis recurrence in the sublingual gland.

In this context, it should be taken into consideration that sialolith can occur even in cases that previously underwent submandibular gland excision and it should also keep sialolith in mind in the differential diagnosis of deep neck infection.

*Funding: No funding sources*
*Conflict of interest: None declared*
*Ethical approval: Not required*

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**Cite this article as:** Çeliker M, Çeliker FB, Terzi S, Dursun E. Sialolithiasis and deep neck infection after the submandibular gland excision. Int J Otorhinolaryngol Head Neck Surg 2017;3:728-31.