



Coexistence of a sialolith, a Warthin tumor in the submandibular gland, and sialocutaneous fistula in the neck

Submandibüler bezde sialolit, Warthin tümörü ve boyunda sialokütanöz fistül birlikteliği

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ABSTRACT

Although sialolithiasis is the most common submandibular gland pathology, a Warthin tumor arising from the submandibular gland is rare. In this article, we report a 63-year-old male patient with a swollen right submandibular region, erythematous, and a fistulous area from which a cutaneous fistula with seropurulent exudates located at the submandibular region. Ultrasound revealed a stone and, therefore, sialoadenectomy and excision of the fistulous tract were performed. Besides the sialolith, the pathological examination revealed a Warthin tumor.

Keywords: Cutaneous fistula; cystadenoma lymphomatosum; papillary; salivary gland calculi; Warthin tumor.

ÖZ

Sialolithiazis submandibüler bezde en sık görülen patoloji olmakla birlikte, submandibüler bezden kaynaklanan Warthin tümörü nadirdir. Bu yazıda, sağ submandibüler bölgede şişlik, kızarıklık ve boyunda submandibüler bölgede seropürülan drenajın olduğu kütanöz fistülü olan 63 yaşında bir erkek hasta sunuldu. Ultrasonda taş tespit edildiği için, sialoadenektomi ve fistül trakt eksizyonu yapıldı. Patolojik incelemede, sialolithiazisin yanı sıra, Warthin tümörü tespit edildi.

Anahtar sözcükler: Kütanöz fistül; kistadenoma lenfomatosum; papiller; tükürük bezi kakülleri; Warthin tümörü.

Sialolithiasis is considered to be the most common salivary gland disorder and most commonly develops in the submandibular gland in 80% of cases. The exact etiology of sialolithiasis is unknown but it can impair salivary flow and predispose the patient to acute and chronic sialadenitis.^[1] Papillary cystadenoma lymphomatosum or Warthin tumor (WT) is the second most common salivary gland tumor.^[1,2] Warthin tumor most commonly develops in the parotid gland, and rarely arises in the submandibular gland.^[2] Warthin tumor is a benign epithelial tumor, multicentric in 30% and bilateral in 5 to 14% of patients.^[1,2] Little is known about the etiology of WT but smoking is significant risk factor for its development. Radiation exposure and autoimmune disorders also may be related to WT.^[2] A cutaneous salivary fistula is rare and arises from

various causes such as infection, malignancy, stone, trauma and operative complications. A rare case of cutaneous salivary fistula caused by stone has been reported.^[3] Here, we report the coexistence of the sialolithiasis and WT of the submandibular gland as a possible cause of cutaneous salivary fistula. To the best of our knowledge, the coexistence of WT and sialolithiasis in the submandibular gland has not been defined before.

CASE REPORT

A 63-year-old male presented himself complaining of seropurulent discharging fistula in the right submandibular region for four days. He complained of recurrent pain, swelling and redness in the right

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Figure 1. Intraoperative view showing fistula in the right side opening in neck.

submandibular area for four years but he did not seek professional medical attention at that time. The intensity of the pain increased during mealtimes. His medical history was unremarkable. He smoked one pack of cigarettes a day. Upon inspection, the right submandibular region was swollen and erythematous with a fistulous area from which seropurulent exudate drained (Figure 1). A 5 cm hard tender swelling in the right submandibular region was palpated. Intraoral examination revealed induration with absent salivary flow from the right Wharton's duct. No cervical lymph nodes could be detected. Sonography revealed a 2 cm stone inside the right submandibular gland with hypo and hyper echoic areas of gland parenchyma. Sialadenectomy and excision of the fistulous tract was carried out (Figure 2). The gland was solid and strongly adherent to the neighboring tissues. The postoperative course was uneventful. Besides the



Figure 2. Removed right submandibular gland and sialoliths.

sialolith, histopathological examination revealed the existence of WT (Figure 3). Two weeks after surgery, the patient underwent a cervical magnetic resonance imaging that did not reveal any pathologic findings. Written informed consent was obtained to publish this case report.

DISCUSSION

This case report might suggest a causative relationship between sialolithiasis and WT. There are few reports on other types of epithelial tumors, multiple myeloma and lymphoma concomitant with sialolithiasis.^[4,5] Although WT occurred with no clinical evidence of concurrent or previous sialolithiasis, based on current knowledge, it is best to accept a coincidental occurrence considering the high incidence of sialolithiasis in the submandibular gland. To our knowledge, there is also no reported sialocutaneous fistula with WT, but a few in sialolithiasis cases.^[3] In our case the coexistence of WT and sialolithiasis may have played additive roles on the deterioration of salivary drainage and subsequent sialocutaneous fistula.

Among current imaging procedures, ultrasonography has the best diagnostic predictive capability for detection of salivary gland diseases because it is noninvasive, has low cost and no radiation exposure. The accuracy of sonography in assessment of sialolithiasis is approximately 90%. Salivary gland neoplasms may be confused with sialolithiasis.^[1] Chronic obstructive sialadenitis can cause progressive paranchymal inflammation, atrophy and fibrosis and possibly be a reason for confusion

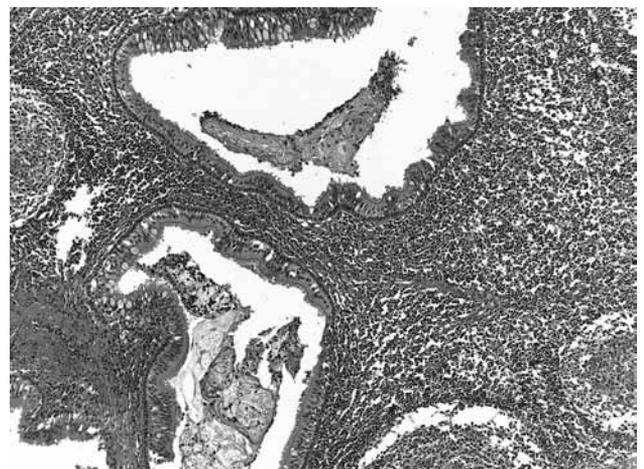


Figure 3. Histopathological findings revealed Warthin tumor (oncocyctic cells lining the cystic space and a dense population of lymphocytes in the stroma) (H-E x 100).

in differentiating from tumor for a radiologist. This brings up the question of whether we need full radiographic examinations to rule out suspicion of tumor in long lasting sialolithiasis. It has been thought that excessive use of medical radiographs may cause increased risk of salivary gland carcinoma. Magnetic resonance imaging is recommended if the degree of suspicion is high based on sonographic findings in patients with sialolithiasis.^[6]

The treatment of submandibular gland WT is sialadenectomy due to multi centric features. Sialadenectomy is the surgery of choice for recurrent obstructive sialadenitis also if a sialolith with a substantial mass is located within the gland and intraoral surgical access is not possible.^[6]

In the present paper we have a new case of coexisting WT with sialolithiasis. We recommend that long-standing sialolithiasis cases should be carefully examined to exclude suspicions of WT before surgery.

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