

Rare coexistence of sialolithiasis and actinomycosis in the submandibular gland

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Abstract

Sialolithiasis is a condition characterized by the obstruction of salivary gland or its excretory duct by a calculus or sialolith. This condition provokes swelling, pain, and infection of affected gland leading to salivary ectasia and even causing the subsequent dilatation of the salivary gland. The aim of this case report is to present a rare condition of sialolithiasis of the submandibular gland with actinomycosis. In this report, we presented a 35-year-old male patient having coexistence of submandibular sialolithiasis and actinomycosis with a literature review. Patient underwent excision of the right submandibular gland due to sialolithiasis. Pathologic examination revealed chronic sialadenitis, sialolithiasis, actinomyces which all necessitate the excision of right submandibular gland with stones with 1.5 cm in diameter. It should be kept in mind that sialolithiasis may be a predisposing factor for submandibular actinomycosis and removal of the sialolith or the entire gland is of particular importance.

Keywords: Submandibular, sialolithiasis, actinomycosis.

Özet: Submandibüler bezde siyalolityaz ve aktinomikozun seyrek görülen birlikteliği

Siyalolityaz, tükürük bezi veya boşaltım kanalının bir taş veya siyalolit ile tıkanması ile karakterizedir. Bu durum etkilenmiş bezin şişmesi, ağrması ve enfeksiyonunu teşvik ederek tükürük bezi ektazisine, hatta daha sonra tükürük bezinin dilatasyonuna neden olmaktadır. Bu olgu raporunun amacı submandibüler bezde siyalolit ile birlikte aktinomikozun görüldüğü nadir bir siyalolityaz olgusunu sunmaktır. Bu raporda submandibüler siyalolit ve aktinomikozu olan 35 yaşında erkek hasta literatür taraması eşliğinde raporlanmıştır. Siyalolityaz nedeniyle sağ submandibüler bez eksize edilmiştir. Patolojik incelemede gözlemlenen kronik siyaladenit, siyalolityaz ve aktinomikoz nedeniyle çapı 1.5 cm taşlarla dolu sağ submandibüler bezin eksizyonunun gerektiği görülmüştür. Siyalolityazın submandibüler aktinomikozun predispozan faktörü olabileceği, siyalolit veya bezin tümüyle çıkarılmasının son derece önemli olduğu akıldan çıkartılmamalıdır.

Anahtar sözcükler: Submandibüler, siyalolityaz, aktinomikoz.

Actinomycosis is a chronic suppurative infection. This infection is characterized by formation of multiple abscesses, draining sinuses, and granulation tissue. The three major clinical presentations of actinomycosis include the cervicofacial region, pulmothoracic region, and abdominopelvic regions.^[1] In cervicofacial actinomycosis, the submandibular area, parotid gland and buccal space are affected. Primary involvement of the submandibular gland is not frequent. Actinomycosis with sialolithiasis of the submandibular gland is a rare situation.^[1] Actinomyces are found as commensal organisms in the human oral cavity, respiratory and digestive tracts. The bacteria become

invasive when a mucosal lesion gains access to the subcutaneous tissue.^[2]

Sialolithiasis usually appears between the age of 30 and 60 years.^[3] It most commonly involves the submandibular gland (80% to 95%) and less frequently the parotid (5% to 20%). The sublingual gland and the minor salivary glands rarely (1% to 2%) have sialolithiasis.^[4] The stones may reach from 0.1 to 30 mm.^[5] While 85% of submandibular gland stones are located in Wharton's duct, the remaining 15% are in gland parenchyma.^[6] However, giant sialoliths (>15 mm) in the submandibular duct have rarely been reported.^[7]

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Different aetiological hypotheses have been put about salivary gland calculi. These are mechanical, inflammatory, chemical, neurogenic, infectious, strange bodies, etc. Submandibular gland is more susceptible to the development of the stone compared to parotid gland. Wharton's duct is longer and wider than the Stensen's duct. Submandibular salivary flow is against gravity; and salivary pH is more alkaline; and mucin proteins, calcium and phosphates are contained in greater amount than serous secretion of parotid saliva.^[8]

In this report, we presented a 35-year-old male patient having coexistence of submandibular sialolithiasis and actinomycosis with a literature survey.

Case Report

Thirty-five-year-old male patient admitted to Şehit Kamil Government Hospital complaining of swelling in the right submandibular region. He had a history of episodes of right submandibular swelling occurring during meals and gradually decreased by resting. Physical examination showed increased sensation and pain on the right submandibular gland. By intraoral examination, saliva flow from the right Wharton's duct was not observed. The ear, nose and throat examination findings were normal. Submandibular neck ultrasonography was reported as "The right submandibular gland diameter increased; ducts dilated in gland; and right sialoadenitis was present". Neck

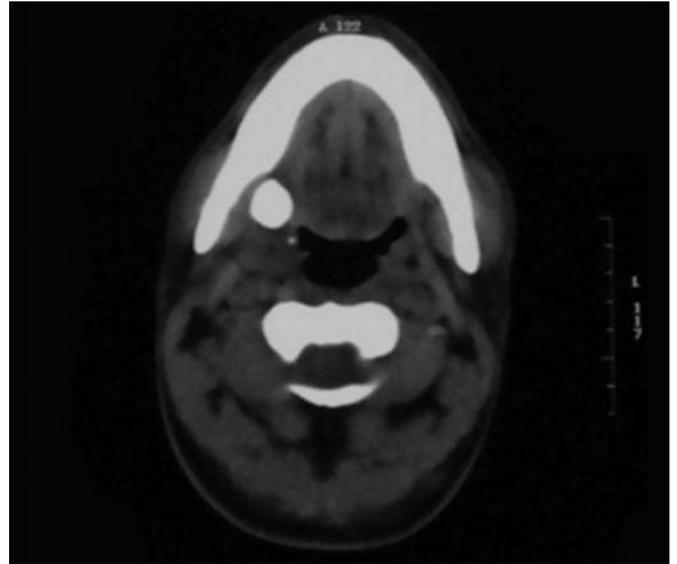


Fig. 1. Computed tomography scan of the right submandibular gland showing calculus.

Computerized Tomography examination showed bone density structure which was 1.5 cm of diameter in the right submandibular gland parenchyma (Fig. 1). Magnetic resonance imaging (MRI) of the neck also demonstrated 1.5 cm structure in the parenchyma (Figs. 2 and 3).

Patient underwent excision of the right submandibular gland. Cephalosporin antibiotic was administered and the

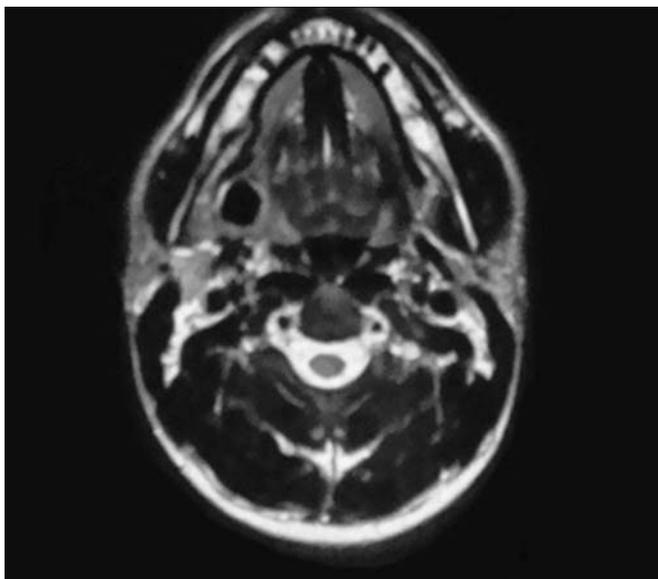


Fig. 2. Axial MRI of the right submandibular gland.

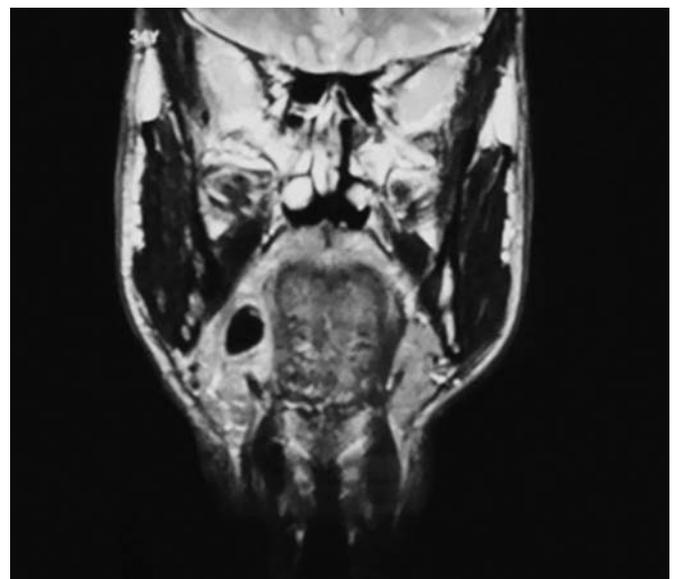


Fig. 3. Coronal MRI of the right submandibular gland.

patient was discharged without problems. On the third day of the patient's discharge, he admitted to the hospital with complaints of pain, swelling and discharge at the operation region. He was hospitalized, and pathology result of the operation was reported simultaneously as "Chronic sialadenitis, sialolithiasis and actinomyces necessitate the right submandibular gland excision with stones with 1.5 cm in diameter". Postoperative infection was thought to be due to infection with Actinomyces. The patient was administered amoxicillin and clavulanic acid 1 g i.v (twice a day) for 10 days; and later, 1 g p.o (twice a day) for 21 days. The patient's symptoms improved without the need for another surgical intervention. The patient gave informed consent for using his data for scientific purposes.

Discussion

Actinomycosis is an *Actinomyces israeli* infection. This commensal organism exists at high amounts in tonsil tissue and carious teeth. Poor oral hygiene, diabetes, immune suppression, malnutrition and local tissue damage are predisposing factors. These conditions may lead to infection and subsequent invasion of subcutaneous tissues.^[8] Actinomycosis infection usually spreads into nearby soft tissues without regard for tissue planes or lymphatic drainage.^[2]

Cervicofacial actinomycosis is frequently the result of oro-maxillofacial trauma, dental manipulation or dental caries. Cervicofacial actinomycosis mostly presents as a firm, painless, slow-growing swelling and a multiloculated abscess which frequently progresses to multiple discharging sinuses.^[3] The infection, most commonly, presents as a chronic, fluctuant mass, commonly positioned at the border of the mandible, becoming progressively larger within weeks or months. The colonies may be visible to the naked eye as 'sulphur granules', which are pathognomonic. When this is identified, the appropriate therapy should be initiated.^[9]

Diagnosis of actinomycosis infection is difficult, as it is hard to isolate *A. israeli*. When actinomycosis is suspected, fine needle aspiration, smears of freshly obtained pus, standard culture swabs, and special stains for fungi are required.^[3] Furthermore, imaging techniques of CT and MRI usually provide non-specific findings.^[4] In this case, the patient's diagnosis was established by the presence of clusters in pathology specimen, but actinomycosis did not grow in culture. Additionally, preoperative CT and MR images did not comply with actinomycosis.

Disease is more rarely seen in the submandibular region. Actinomycosis and salivary gland stones rarely occur. It

should be kept in mind that sialolithiasis may be a predisposing factor for submandibular actinomycosis, and removal of the sialolith or the entire gland is of particular importance. In our case, we thought that actinomycosis infection was present with the submandibular stone, and to be induced by surgery.

Actinomycosis in the head and neck has a better prognosis than other sites since this region is more responsible to surgery and antibiotic therapy. Surgery plays an important role in diagnosis and treatment. High-dose intravenous antibiotics for 2–4 weeks are a fundamental part of treatment and 3–6 months of treatment with oral antibiotics is recommended. Tetracycline and erythromycin are used in patients with penicillin allergy.^[10] In our case, infection has been treated with proper therapy after the diagnosis without invasive surgery.

The instrumental diagnosis of sialolithiasis is based on several imaging techniques. Ultrasonography represents an excellent first-level diagnostic technique because it reveals ductal and highly mineralized stones with a diameter of at least 1.5 mm with an accuracy of 99%.^[11] In this case, USG examination did not give definitive diagnosis but CT established a definitive diagnosis.

The ultimate aim of giant sialolith treatment is to restore a normal salivary flow. When the stone can be palpated intraorally, the best option is to remove it through an intraoral approach. The choice of a surgical approach to access the sialolith with submandibular gland preservation requires careful imaging evaluation, minimal invasive removal and transoral sialolithotomy. After surgical calculi removal, the patients show asymptomatic and normally functioning glands in a short time.^[12] In our case, we removed the huge stone in parenchyma with salivary gland.

Our case showed that giant stone in submandibular gland may be coexistent with actinomycosis. The appropriate use of antibiotics should be recommended to treat these infections.

Conflict of Interest: No conflicts declared.

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