

An unusual giant subacute necrotizing sialadenitis as an emergency case of otolaryngology

Kulak Burun Boğaz acili olarak başvuran nadir büyüklükte subakut nekrotizan siyaladenit olgusu

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Subacute necrotizing sialadenitis is an inflammatory necrotizing lesion occurring in minor salivary glands. Most cases occur in the palatal region. In this article, we reported a 36-year-old man referred to our clinic as an emergency with the complaints of excessive bleeding, airway obstruction and hipovolemia. Intraorally, there was a hemorrhagic, protruding giant mass in the palatal region. Following the first biopsy, which was not diagnostic, a second biopsy was performed. Histopathologic examination showed acinar cell necrosis and dense inflammation of the affected minor salivary glands in the second biopsy. The diagnosis of subacute necrotizing sialadenitis was made on the basis of clinical and histologic features of the lesion. Subacute necrotizing sialadenitis is a rare lesion, and admittance to the otolaryngology clinic as an emergency case is much rarer. To avoid unnecessary surgical intervention, it is necessary to diagnose subacute necrotizing sialadenitis correctly, which can be confused with malignant diseases of the salivary glands.

Key Words: Carcinoma; salivary glands; subacute necrotizing sialadenitis.

Subakut nekrotizan siyaladenit, küçük tükürük bezlerinde görülen enflamatuvar nekrotizan bir hastalıktır. Olguların çoğu üst damak yerleşimlidir. Bu yazıda, aşırı kanama, üst solunum yolu tıkanıklığı ve hipovolemi bulgularıyla kliniğimize acilen refere edilen 36 yaşında erkek hasta sunuldu. Oral kavite ortaya konduğunda üst damakta protrude olmuş kanamalı büyük bir kitle varlığı görüldü. Tanısal olmayan birinci biyopsiden sonra ikinci biyopsi alındı ve histopatolojik olarak tükürük bezi asiner hücrelerinde yoğun enflamasyon ve nekroz görüldü. Lezyonun histolojik özellikleri ve mevcut kliniğine göre subakut nekrotizan siyaladenit tanısı kondu. Nadir bir lezyon olan subakut nekrotizan siyaladenit olgusunun, acil olarak kulak burun boğaz kliniğine gelmiş olması çok daha nadirdir. Tükürük bezinin malign hastalıklarıyla karıştırılabilen subakut nekrotizan siyaladenitte gereksiz cerrahiden kaçınmak için doğru tanı konması esastır.

Anahtar Sözcükler: Karsinom; tükürük bezleri; subakut nekrotizan siyaladenit.

Necrotizing sialometaplasia (NS) and subacute necrotizing sialadenitis (SANS) are self-limiting benign inflammatory lesions of the salivary glands.

The etiology of these lesions is unknown, but trauma, ischaemia, infections and allergies have been suggested as possible etiologies.^[1,2] The reported

cases predominantly occurred in the palatal minor salivary glands but rarely involved the major salivary glands. The lesions typically present as localized and often erythematous palatal swellings accompanied by an abrupt onset of pain.

Microscopically, SANS is characterised by a sub-acute inflammation of the affected minor salivary glands. The inflammatory infiltrate is composed of neutrophils, lymphocytes, histiocytes and eosinophils and there is a loss of the acinar cells.^[3]

As far as we know, there is no case in literature that reaches a size of 5x7 cm and necessitates an emergency intervention because of excessive bleeding and airway obstruction.

Therefore, here we present a giant SANS as an emergency case of otolaryngology together with the treatment protocol and the evaluation of the patient.

CASE REPORT

A 36-year-old man presented with excessive bleeding, airway obstruction and hypovolemia. During the intraoral examination, a hemorrhagic, protruding mass was observed in the palatal region. Macroscopically, it was initially thought to be a malignant lesion. There was tonsillitis in the patient history, and a mild bleeding had occurred a week ago. In addition to these, the lesion had rapidly increased within five days.

The patient was taken directly to the operating theatre and general anaesthesia was induced. On the clinical examination, there was a, fragile, tender mass 5x7 cm in size in the left soft and hard

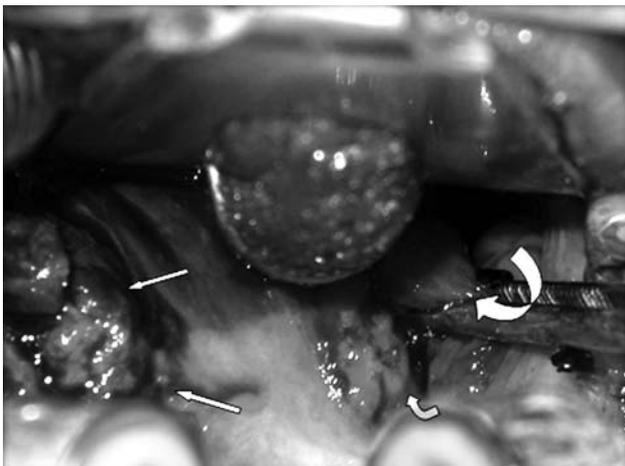


Fig. 1. Clinical appearance of the protruding mass from the soft palate. The mass involves left tonsillary fossa and the lesion is indicated by arrows at the left side.

palate region (Fig. 1). The magnetic resonance imaging showed that the mass blocked the airway passage in such a way as to include the tonsils (Fig. 2, 3). The excessive bleeding was controlled with ice-packs and an hemostatic agent (surgicel). Angiography could not be performed due to the unfavorable conditions at that time. A tracheostomy was performed to secure the airway and a nasogastric tube was applied for feeding. The hematologic examination of the patient revealed that the hemoglobin concentration was 10.7 gr/dl, the white cell count 35000 and the hematocrit were 33%. Therefore, he was given a course of amoxicillin trihydrate + potassium clavulanate 1.2 gr intravenously (i.v.) twice a day and metronidazole 500 mg i.v. three times a day for seven days. Hemorrhagic diathesis was excluded because the coagulation tests were negative. Five days later, the white cell count decreased to 12300, but the erythrocyte sedimentation rate was 125 mm/h. Therefore, an autoimmune vasculitis was considered, but the cytoplasmic antineutrophil cytoplasmic antibody (c-ANCA) and perinuclear antineutrophil cytoplasmic antibody (p-ANCA) were negative. An incisional biopsy was taken from the lesion. Regression was observed in the mass five days later and the ulceration was seen in the palate one week later. In two weeks, the lesion was completely healed. The histopathologic examination primarily showed a non-diagnostic necrotic tissue. In the second biopsy, it turned out to be an

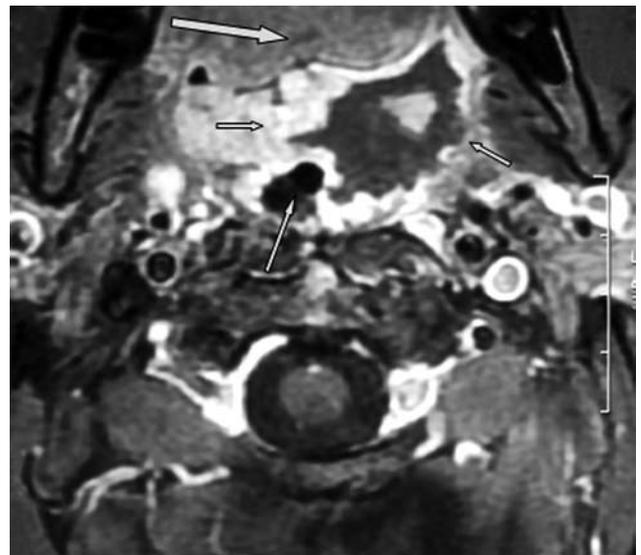


Fig. 2. Axial magnetic resonance imaging of the lesion indicated by arrows. The mass blocked the airway passage (thin arrow) in such a way as to include tonsillary fossa and left soft palate (small arrow).

acinar cell necrosis and dense inflammation of the affected minor salivary glands (Fig. 4). The diagnosis of SANS was reached on the basis of the clinical and histologic features of the lesion after repeated biopsies. The patient was discharged from the hospital two weeks later and the follow-up period was uneventful.

DISCUSSION

Subacute necrotizing sialadenitis is a nonspecific inflammatory condition of the oral minor salivary glands. The typical clinical presentation of SANS is a painful, localised erythematous, non-ulcerated swelling with a history of recent rapid increase in size ranging from 0.3 to 1.5 cm. The duration of the swelling is about a week in most of the cases. Subacute necrotizing sialadenitis has a male to female ratio of 3:1 and the majority of the cases occur in the second and third decades of life. Similarly, our patient was a 36-year-old man and had a palatal lesion, which was progressively enlarged in five days.^[2]

Some authors consider SANS to be an unusual subtype or early stage of NS, while some others think that it is a distinct and specific entity of infec-

tious or allergic origin. A current or recent upper respiratory tract infection is a clinical feature supporting an infectious origin for SANS.^[3-5] Our patient also had a history of tonsillitis a week ago.

The differential diagnosis of SANS includes NS, chronic sialadenitis, squamous cell carcinoma and mucoepidermoid carcinoma. Subacute necrotizing sialadenitis and NS share a similar sex and site predilection. The clinical differences between SANS and NS are related to the duration and the type of symptoms.^[3,6-8] The average duration before diagnosis in NS is approximately three weeks, whereas for SANS it is usually less than one week. Pain is almost always present in SANS, but it is a variable finding in NS. Subacute necrotizing sialadenitis presents as an erythematous swelling whereas NS presents as an ulcer. The average healing time for NS is five to six weeks, where most cases of SANS resolve within two weeks after diagnosis. In our case, we initially suspected a malignant lesion or NS on the basis of the clinical appearance, but the lesion completely healed with medical treatment in two weeks. The clinical features of our case were consistent with SANS.

According to Fowler and Brannon,^[3] the inflammation appears to be the earliest event in SANS, whereas in NS, The ischaemia induced acinar necrosis is the earliest event, followed by an inflammatory response. Only a focal acinar cell necrosis is typically observed in SANS, although lobular acinar cell necrosis and ductal squamous metaplasia are key diagnostic criteria of NS. There was a dense inflammation of the affected minor salivary glands with necrotic areas but without



Fig. 3. Coronal magnetic resonance imaging showed the lesion extending from the left median pterygoid muscle (curved arrow) to the right palatine tonsil (tall arrow) and inferiorly the epiglottis (small curved arrow) by involving left tonsillar fossa (small arrow).

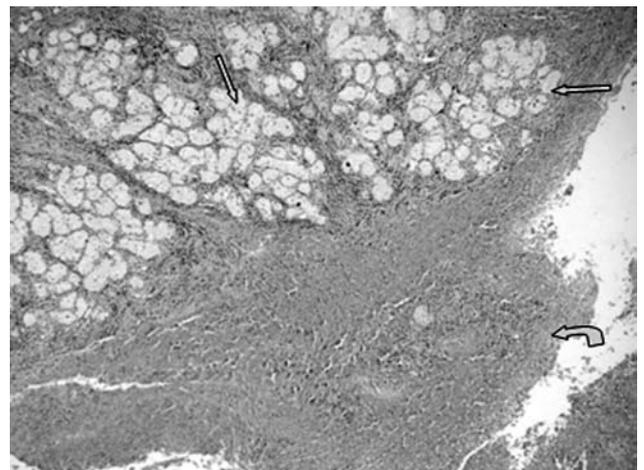


Fig. 4. Large geographic necrosis adjacent to mucous glands (curved arrow) and dense inflammatory cell infiltrate (straight arrow) in the biopsy taken from the palatal region (H-E x 6).

ametaplasia in the present case, which are also consistent with the diagnosis of SANS.

In conclusion, SANS is a rare lesion and an admittance as an otolaryngology emergency case is much rarer. Unfortunately, this case has been misdiagnosed as a malignant neoplasm, resulting in unnecessary surgery: a high cost for a self-limiting lesion. An incisional biopsy is the sensible way to confirm the histologic diagnosis and to rule out more serious disease processes.

This case report illustrates that SANS may present as a giant mass and necessitate an emergency intervention because of excessive bleeding and respiratory obstruction. Since it may be misdiagnosed as a malignancy, it is vital to make the differential diagnosis of SANS correctly to avoid a surgical intervention based on the assumption of a malignant lesion.

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