Bilateral postauricular Kimura's disease: a case report

Bilateral postauricular Kimura hastalığı: bir olgu sunumu

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Abstract

Kimura's Disease is a chronic inflammatory disorder of unknown etiology commonly seen among people who live in the Middle and Far East countries; characterized lymphatic follicles, vascular proliferation, and marked eosinophilic infiltration by histologically. It is mainly seen on the head and neck region. The lesion is benign but might be confused with malignant lesions. Kimura's Disease is often seen in the second and third decades of life. This disease is characterized by a blood and tissue eosinophilia, markedly elevated serum IgE levels, painless subcutaneous mass and regional lymhpadenophaty on the head and neck region. In this report, we present a patient who admitted to our hospital with bilateral postauricular swelling for a one month history. Bilateral postauricular masses were excised under local anesthesia and Kimura's disease was diagnosed by histopathological examination of these lesions.

Özet

Kimura hastalığı orta ve uzak doğu ülkelerinde yaşayan insanlar arasında yaygın olan, histolojik olarak lenfatik folliküller, vasküler proliferasyon ve belirgin eozinofilk infiltrasyon ile karakterize nedeni bilinmeyen kronik bir inflamatuar hastalıktır. Daha çok baş boyun bölgesinde görülür. Lezyon benigndir fakat malign lezyonlarla karıştırılabilir. Kimura hastalığı çoğunlukla yaşamın ikinci ve üçüncü on yılında görülür. Hastalık ağrısız subkutan şişlik, baş boyun bölgesinde bölgesel lenfadenopati, kan ve doku eozinofilisi ve belirgin artmış serum IgE düzeyleri ile karakterizedir. Bu makalede her iki kulak arkasında bir aydan beri mevcut olan şişlik şikâyetiyle başvuran bir hastayı sunduk. Her iki kulak arkasındaki kitleler lokal anestezi altında eksize edildi ve bu lezyonların histopatolojik incelemelerinde Kimura hastalığı tanısı konuldu.

Introduction

Kimura et al in 1948 described a condition entitled “On the unusual granulation combined with hyperplastic changes of lymphatic tissues. It is a chronic inflammatory condition, often producing subcutaneous tumor like nodules with a predilection for the head and neck region. It is prevalent among eastern people and is often associated with major salivary gland enlargement and regional lymphadenopathy (1). Although its etiology is unclear, the clinical and histopathological features of this disease are consistent with allergic or autoimmune. Peripheral blood eosinophil counts and serum immunoglobulin E levels are commonly increased (2). The lesions mainly occur on the head and neck region and may be recurrent after the treatment. Histologically, these conditions are characterized by lymphoid infiltration, vascular proliferation and tissue eosinophilia (3). Herein, we aimed to present a patient with a one month history of bilateral postauricular swelling diagnosed as Kimuras's disease.

Case Report

A 13-year-old Turkish male was admitted to our hospital with asymptomatic masses behind both of his ears for a one month history. Clinical examination revealed a 3x2 cm painless subcutaneous masses which localized to postauricular regions. The salivary glands were not enlarged and lymph nodes were not palpable on the physical examination. Laboratory findings were found within the normal limits except for peripheral blood eosinophilia and elevated serum total IgE level. Excisional biopsies of the bilateral postauricular masses were performed, and the specimens were stained with hematoxylin and eosin (H&E). Histological examination revealed multiple lymphoid follicles with distinct germinal centers (Figure 1-2). Kimura's disease was diagnosed by histopathologically and the patient had no complaints during the two months follow-up while being monitored from the outpatient clinic.

Discussion

Kimura's disease is also called eosinophilic hyperplastic lymphogranuloma. It is a rare, chronic

inflammatory condition with a predilection for the head and neck region involving subcutaneous tissues, lymph nodes, parotid glands, submandibular glands, and lacrimal glands (1). Kimura's disease is an unknown etiology, and self-limited. Allergic or autoimmune reactions may play a role for this disease. It has been speculated that candida albicans, viruses, parasites or neoplasms may alter T-cell immunoregulation or induce IgE-mediated type I hypersensitivity resulting in the release of eosinophiliotrophic cytokines (4, 5). It is characterized histologically by distinct lymphocytic infiltration of subcutaneous tissue with vascular proliferation and fibrosis. This disease is seen commonly among eastern people and is usually seen in the second and third decades of life, and more seen in men (1). Contrary to what has been previously reported from the literature, this disease was seen in the second decade and the lesions were bilateral in our patient.

Angiolymphoid hyperplasia with eosinophilia (ALHE) was thought to be a similar lesion with Kimura's disease. The clinical appearance of the lesions in ALHE and Kimura's disease are similar with increased erythematous skin lesions and fibrous subcutaneous nodules (3). Several researchers have suggested that ALHE is a stage of histiocytoid or epithelioid hemangioma, which is a real neoplasm, whereas Kimura's disease is a localized chronic inflammatory condition which is a consequence of systemic immunologic reaction.

Nodal involvement in Kimura's disease is characterized by prominent eosinophilic infiltration with occasional formation of eosinophilic microabscesses, an increased number of small blood vessels, eosinophilic folliculitis, and increased number of small bleed vessels, eosinophilic folliculitis and perinodal eosinophilic infiltration (1). The differential diagnoses for Kimura's disease include reactive lymphadenopathy, lymphoma, nodal metastasis, salivary gland tumor and Mikulicz's disease. Fine-needle aspiration cytology may have an important role to play in the diagnosis of recurrent Kimura's disease. Other possible differential diagnoses based on the cytological features include reactive lymphadenopathy with eosinophilia, parasitic infection and, in particular, lymphoma. Thus, for the initial diagnosis, it is advisable to perform an excision biopsy for confirmation. Fine-needle aspiration cytology is reserved for the diagnosis of recurrent lesions in Kimura's disease and may spare the patient repeated biopsy examination. Various treatment modalities have been tried with variable success in the case of Kimura's disease, including surgical excision, radiotherapy, corticosteroids (intralesional and oral), pentoxifyslline, cyclosporine, nonsteroidal anti-inflammatory drugs and oral retinoids (3). Messina-Doucet et al have suggested surgical excision as the treatment of choice for definitive diagnosis and initial management (6). Localized initial recurrence can be managed by surgical excision. If recurrence becomes frequent, oral corticosteroids should be initiated. If all else fails, radiotherapy can be considered, but side-effects limits it's usage for such a benign condition (3).

In conclusion, Kimura's disease of the bilaterally postauricular subcutaneous tissue is rare. It is a chronic inflammatory disorder that can be confused with a malignant tumor. Therapeutic surgical excision, radiation therapy, steroids or a combination of these may be useable. Spontaneous recovery is rarely seen in some cases.

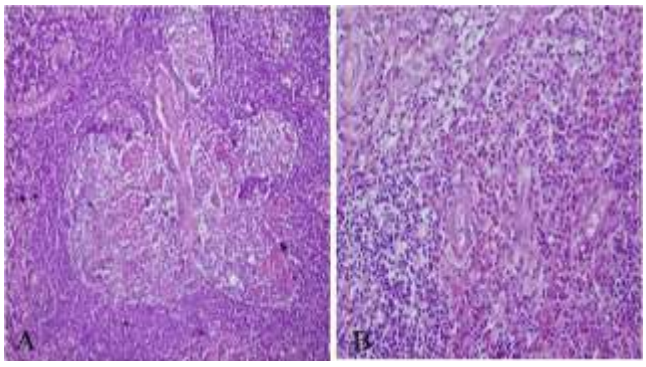

Figure 1: *Follicular hyperplasia of germinal centers (A), mature eosinophils in hyaline vessels (B)*

Yazarlarla ilgili bildirilmesi gereken konular (Conflict of interest statement) : Yok (None)

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