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CASE REPORT

SCROFULODERMA IN AN OLDER PATIENT: AN UNDERESTIMATED ENTITY AND UNDERDIAGNOSED CASE

ABSTRACT

Cutaneous tuberculosis (TB) is a rare form of extrapulmonary tuberculosis (EPT) and scrofuloderma is one of the manifestations of cutaneous TB. It is usually caused by fistulization of lymph nodes to the skin. The most common predilection site is the cervical region. A 96 year-old female was admitted with multiple cervical swollen lymph nodes with 10 years duration. The biggest one had fistulized through the skin discharging purulent material. Multiple samples of purulent material were obtained and evaluated by direct light microscopy after Ziehl-Nielsen procedure. Acido-resistant bacilli (ARB) at a level of (++++), were detected in every microscopic field BACTEC and polymerase chain reaction (PCR) analyses were found as positive and microorganism was identified as *Mycobacterium tuberculosis*. The fistulized lymph node was debrided at the bed-side and pathological examination showed granulomatous lymphadenitis. Four-drug anti-tuberculosis regimen was started immediately. However, although clinical improvement was present, the patient died unexpectedly at the first month of the treatment due to cardiac reasons without any therapy-related complications. In conclusion, scrofuloderma should be suspected in any elderly patient with enlarged lymph nodes along with a fistulization to skin especially in countries where TB is endemic like Turkey, and approached appropriately.

Key Words: Skin; Tuberculosis; Scrofuloderma.



OLGU SUNUMU

YAŞLI HASTADA SKROFULODERMA: GÖZARDI EDİLMİŞ ANTİTE VE TANISIZ KALMIŞ OLGU

Öz

Cilt tüberkülozu (TB) ekstrapulmoner tüberkülozun (EPT) ender bir formudur, skrofuloderma ise cilt TB formlarından biridir. En sık servikal bölgede görülmekte ve genellikle lenf nodlarının cilde fistülizasyonu sonucu ortaya çıkmaktadır. 96 yaşında kadın hasta 10 yıldır süren servikal lenf nodları nedeniyle kliniğe yatırıldı. Bu lenf nodlarından en büyüğünün cilde fistülize olduğu ve buradan pürülan bir materyalin drene olduğu izlendi. Lezyondan drene olan pürülan materyal, Erlich-Ziehl-Nielsen ile boyandıktan sonra direkt ışık mikroskopunda incelendi ve (++++), asit-rezistan basil (ARB) saptandı. Ayrıca BACTEC ve PCR yöntemleri ile ARB pozitifliği gösterildi ve mikroorganizma *M. tuberculosis* olarak verifiye edildi. Fistülize lenf nodu yatak başı debride edildi ve patolojik incelemede granülatöz lenfadenit saptandı. Dörtlü rejim içeren anti-tüberkülo tedavi hemen başlandı. Tedavinin birinci ayında ateş düştü, klinik yanıt alındı ancak olgu tedavi ile ilişkili komplikasyonlar gelişmeden ani kardiyak nedenler yüzünden kaybedildi. Sonuç olarak, Türkiye gibi TB açısından endemik bir bölgede cilde fistülize lenf nodu saptanan ileri yaşta olgularda akla hemen skrofuloderma getirilmeli ve buna yönelik yaklaşım yapılmalıdır.

Anahtar Sözcükler: Cilt; Tüberküloz; Skrofuloderma.

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INTRODUCTION

Cutaneous tuberculosis (TB) is a rare form of extra-pulmonary tuberculosis (EPT) and accounts for 0.11 to 2.5 per cent of all patients with skin diseases (1,2). Scrofuloderma, a type of cutaneous TB is the term applied to lesions that develop in the skin from contiguous spread or extension of tuberculosis infection from an underlying or adjacent structure. Most often, the primary focus is lymph nodes but bones and joints may also be the source of infection. Scrofuloderma after Bacille Calmette-Guerin (BCG) vaccination has also been reported (3). Although scrofuloderma has been reported mainly in childhood, it can be seen in all ages. Herein, we are presenting an underdiagnosed case which had been surprisingly left untreated for years.

CASE REPORT

A 96 year-old lady was admitted with 10 years history of multiple swollen cervical lymph nodes in different variety of size and the biggest one (2 x 2.5 cm), fistulized to the skin surface discharging purulent material. She was in poor health without taking enough nutrition or hydration. During 10 years, the lesions had become in different size, some of them disappeared and newly formed. However five months ago, one of them had begun to fistulize to the skin. She had been evaluated in different health care centers multiple times; however, no diagnosis had been established. Before admitting our hospital, a smear from fistula tract was learned to be gained without any specific diagnosis. We were unable to find any note about a search for tuberculosis in her laboratory charts from other centers. Primer hypertension and thoraco-abdominal aortic aneurisms with 7 cm diameter had been diagnosed within past three years and she had been on antihypertensive therapy. On physical examination, the temperature was 38°C, and blood pressure 100/60 mmHg. She had dry mouth, coarse crackles at the both lung bases, and pansystolic murmur at the mitral focus. Moreover, five lymph nodes which were soft like a rubber with smooth surface giving fluctuation were found at the cervical region. A sinus tract was observed on the biggest one, the center of lesion had nodularity and the borders were like crater on her atrophic skin (Figure 1). The white blood cell count was 13400/mm³, erythrocyte sedimentation rate 100mm/h and C-reactive protein was 16.87mg/dL. Blood biochemistry was normal and she was negative for human immunodeficiency virus (HIV) antibody. Posterior-anterior chest x-ray revealed that aortic arc and descendant aorta were widened and mediastinal structures moved to the right

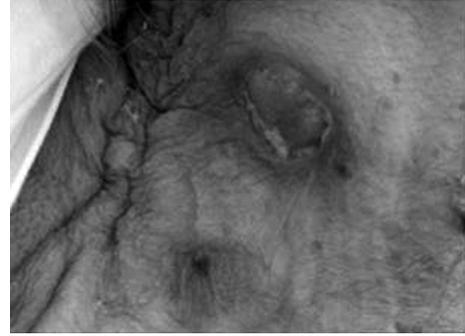


Figure 1— Lymph nodes on the cervical region (one of them was fistulized to the skin by a crater-like mouth) were shown.

hemithorax without paranchimal infiltration. Echocardiogram revealed normal left ventricular function with mild aortic and mitral insufficiency.

Multiple samples of purulent material were evaluated by direct light microscopy following Ziehl-Nielsen staining procedure. Acido-resistant bacilli (ARB) were detected in every microscopic field, revealing four positivity (++++), according to American Thoracic Society (ATS) classification (Figure 2) (4). BACTEC and polymerase chain reaction (PCR) analyses were found positive and microorganism was identified as *Mycobacterium Tuberculosis*. Sputum smear was negative for ARB. Four-drug anti-tuberculosis regimen (rifampicin, isoniazid, pyrazinamide and ethambutol) was started immediately. Nasogastric feeding tube was inserted and medication and nutrition solutions were administered via this route. The fistulized lymph node was debrided bed-side and following debridement, all of the lymph nodes at the right cervical region were extirpated by local anesthesia in operating room. The microscopic examination of these was also positive for

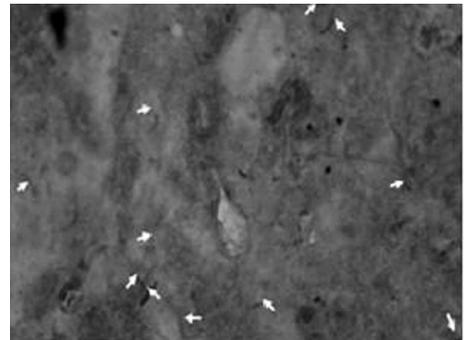


Figure 2— Acido-resistant bacilli four positivity (++++), (white arrows) from the purulent discharge by Ziehl-Nielsen.



ARB. Histopathological examination showed granulomatous lymphadenitis. After the operation, the incision line was clear and dressing was performed in every other day for fifteen days until removing the sutures. Under anti-tuberculosis therapy, the fever began to go down to the normal level in a week. The patient regained her lose appetite and nasogastric tube was removed. However, she suddenly died at the first month of the therapy due to cardiac reasons without any therapy-related complications.

DISCUSSION

Tuberculosis is a major cause of illness and death worldwide, especially in Asia and Africa. Globally, 9.27 million new cases and 1.7 million deaths from TB occurred in 2007 (5). In the era before HIV pandemic and in studies involving immune-competent adults, it has been observed that EPTB constituted about 15 to 20 per cent of all cases of TB (6,7). In Turkey, TB has two peaks: in younger groups (15-24 years of age) and in elder population (65+ years of age) (8).

Cutaneous TB is a rare form of EPT. Our country has high incidence rate of TB with a 27.9/100.000 incidence according to 2007 statistics reported in 2009 (8). In a study from Turkey, it has been reported that EPT constitutes 20.70% of all TB cases and cutaneous TB constitutes 2.2% of EPT (9). Scrofuloderma is one of types of cutaneous TB and usually caused by fistulized lymph nodes to the skin. The most common focus is cervical lymph nodes. ARB can be demonstrated in the discharge of lesion. Fine needle aspiration cytology of underlying structure, demonstrates the tuberculosis etiology of the skin lesion. Biopsy from the edge of the sinus reveals a mixed cell granuloma with areas of necrosis. ARB may be identified and culture may be positive for *Mycobacterium tuberculosis*. In the presented case the diagnosis was established by microscopic examination and PCR evaluation of discharging material. Then the diagnosis was supported by excisional biopsy performed for mainly therapeutic reasons.

Peripheral lymph node involvement is commonest form of EPT and the cervical region is most frequently affected region like in our patient. In HIV-negative patients, isolated cervical lymphadenopathy is seen in about two-thirds of the patients (10,11). Lymph node TB is considered to be the local manifestation of a systemic disease and often affects children and young adults different from presented case. Patients usually present with slowly enlarging lymph nodes and may otherwise be asymptomatic. In the presented case, the duration of illness was extremely long, since the lymph nodes first appeared 10 years ago. Especially elder patients may not ma-

nifest general TB symptoms. The enlarged lymph nodes may be of varying in size, discrete or matted. There are numerous causes of peripheral lymphadenopathy and it is difficult to distinguish clinically from other causes of enlarged nodes, such as reactive and/or HIV-related lymphadenopathy, malignancies and other lymph node infections, which are also common. Hence, excisional biopsy of the lymph nodes should be performed to reach definite diagnosis. Certain hysto-pathological features like granulomatous inflammation with caseating necrosis have been accepted as suggestive for TB. Needle aspiration with cytology and tuberculosis microscopy of aspirated material has a high diagnostic yield, with confirmation of over 85% of patients with tuberculosis lymphadenitis suggesting that the technique may be important. Cytology, can also identify most other important causes of enlarged lymph nodes.

The recommended therapy for cutaneous TB is the use of classical regimens as used for pulmonary TB (2). In a study, using these regimens, the skin lesions of scrofuloderma healed in five to six months (12). Surgical excision, cryotherapy and electrocautery application in addition to anti-tuberculosis therapy may also be performed in selected patients. In presented case, surgical excision of lymph nodes were performed in order to get over TB load in the occupied lymph nodes for contribution of the therapy.

Presented geriatric case is unique, because she remained under diagnosed for approximately 10 years although it was very simple to reach diagnosis. In conclusion, scrofuloderma should be brought into mind in geriatric age when long-standing enlarged lymph nodes fistulize to skin and microscopic examination for ARB should immediately be performed.

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