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Unusual Primary Cutaneous Actinomycosis: A Clinical Enigma

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Abstract

Background: Actinomycosis is a rare subacute to chronic bacterial infectionencountered in the humans.Actinomycosis is most commonly caused by actinomycosis israelii (A. israelii). Males have a higher incidence than females but now the incidence of actinomycosis is decreased.

Methods and Result: A 35-year-old woman presented to outpatient department (OPD) with complaint of a multiple plaque like lesion on the nape of neck. There was no history of trauma, human bite or barefoot walking. FNAC and punch biopsy from these lesions was taken and sent for histopathology. Histopathological features were pathognomic of Cutaneous Actinomycosis.

Conclusions: Clinical features, histopathological findings, and cultures are the cornerstones to prevent misdiagnosis.

Keywords: Actinomycosis, Cutaneous, A. israelii, FNAC, Histopathology.

Introduction

In the literature human actinomycosis was first described in 1857, although in cattle a similar infection had been discovered in 1826. In Israel, first human case was described in 1878, though the first microscopic appearance of actinomyces granules was identified by Hartz in 1879.[1] Actinomycosis is caused by facultative pathogenic commensals. It is a rare sub-acute or chronic suppurative and granulomatous bacterial infection. In the humans A. israelii and A. gerencseriae are the most commonly encountered organisms.[2]

Around More than thirty species of actinomyces have been identified. The most prevalent species isolated in human infections is A. israelii and found in most clinical forms of actinomycosis.[2]The disease occurs worldwide and has no geographic boundaries with a peak incidence in the middle decades. Males have a three times higher incidence than females.[3]

Actinomycosis was once a common and very deadly disease but now, the incidence of actinomycosis is decreased since the introduction of antimicrobial agents,

actinomycosis has become a more diagnostic challenge.[1,4]

Here we present a case report of a primary cutaneous actinomycosis.

Case Report

A 35-year-old woman presented to the Dermatology OPD of a tertiary care hospital with complaint of a multiple plaque like lesion on the nape of neck and back since 5 years. On examination largest lesion was measuring 1.5x1.5 cm over the back and had multiple openings discharging pus (Figure 1). The patient was a housewife from low middle socio-economic status. She was apparently well about 5 years back. Then she noticed a hard swelling over the back which increased in size gradually spreading on back and upwards. The patient had no history of trauma, human bite or barefoot walking. She complained of mild intermittent fever. There was no history of weight loss and no past history of any major medical or surgicalillness. Her past and family history was non-contributory. Based on the history and the clinical findings, a differential diagnosis of tuberculosis due to the endemicity of tuberculosis in this region was considered.

Her laboratory investigations revealed hemoglobin level of 10.2 gm/dl, total leukocyte count was 14,000/ml with differential being of 80% neutrophils. Blood sugar, renal and liver function tests and urine as well as blood cultures were negative. Montoux's test was negative. Other investigations like radiological evaluation of chest, spine, abdomen, pelvis was within normal limits.

Fine needle aspiration from the multiple lesions was done. Slides were prepared from the pus-like material and stained for MGG, AFB, PAS and Gram stain. Cytosmear revealed abundant neutrophils and few macrophages in a proteinacecous background (Figure 2). AFB was non-contributory. In PAS and Gram stain no organism identified. So skin biopsy was suggested for definite diagnosis.

A punch biopsy from these lesions was taken and sent for histopathology examination. Microscopic haematoxylin and eosin-stained sections showed unremarkable epidermis of the skin. The dermis revealed dense chronic inflammatory infiltrate, comprising mainly of lymphocytes, histiocytes, plasma cells, and fibroblasts. Giant cell and granuloma was not observed. The inflammation was involving blood vessels and adnexal structures; and reaching upto the subcutaneous tissue. On further sectioning, a well-defined focus of abscess formation was identified, comprising predominantly of neutrophils. In the center of focus, few filamentous organisms were identified which were PAS positive. Splendore-Hoeppli phenomenon was observed. This bacterium appeared to form clubs at the end/periphery of bacterial colonies. ZN stain using 20% H_2so_4 was non-contributory. Histopathological features were pathognomic of Cutaneous Actinomycosis (Figure 3).

Discussion

Actinomycosis has been called as "the most misdiagnosed disease" even by experienced clinicians. It is listed as a "rare disease" by the office of rare diseases (ORD) of the National Institute of Health (NIH).[5] Incidence of actinomycosis is reducing due to better dental hygiene and antibiotics used for other infections.[1]

It's a worldwide disease seen mainly in tropical regions such as Asia, Africa, Central and South America. Infection mostly spread through the foot of bare footed persons and primary skin infections may develop after human bites. Actinomycosis is most commonly caused by A. israelii, which may also cause endocarditis. Other

less common causes of Actinomycosis include A. odontolyticus, A. naeslundii/viscosus complex, A. meyeri, and A. gerencseriae. Actinomycosis is a subacute to chronic contiguous infection, characterized by suppurative and granulomatous inflammation and formation of multiple abscesses and sinus tracts that may discharge "Sulphur granules".[6,7]

Although, certain species are commensal in the skin flora, oral flora, gut flora and vaginal flora of human.[8]The causative agents are non-virulent, a breech in the mucous membranes and the presence of devitalized tissues lead to invasion into deeper body structures and cause disease.[9] Immunocompromised patients due to HIV, lung and kidney transplantation, steroid and chemotherapy etc are at high risk of developing actinomycosis.[10]

Depending upon the site involved it can be discussed under cervicofacial actinomycosis, respiratory tract actinomycosis, bone and joint actinomycosis, genitourinary actinomycosis, digestive tract actinomycosis, CNS actinomycosis, cutaneous actinomycosis forms.[11]Cervicofacial actinomycosis is the most frequent form and typically causes oral-cervicofacial disease characterized by a painless "lumpy jaw". Lymphadenopathy is uncommon in this disease.[12]

The third most common type is respiratory actinomycosis after cervicofacial and abdominopelvic forms. Actinomycosis of thoracic is often misdiagnosed as neoplasm.[13] Abdominal actinomycosis can form a sinus tract that drains to the abdominal wall or the perianal area, and symptoms include fever, abdominal pain and weight loss.[12] Pelvic actinomycosis is a rare but proven complication of use of intra uterine devices.[14]

Primary actinomycosis of skin and soft tissue is poorly described in literature. Skin disruption may facilitate invasion of Actinomyces species. Most patients may present with an abscess or cold mass or nodular lesions with fistulas that need to be differentiated from chronic inflammatory skin disease, cutaneous mycobacterial infection and sporotrichosis.[15]

Our patient had plaque like lesions predominantly on the back. History of trauma or bite was absent. Similar case was reported by Kara et al where 9-year-old boy had back swelling.[16] Another case was reported by Bose et al. where a 32 year old female patient with a pus-like discharge on the left side of her back and left armpit.[9] Actinomycosis can present as tumors mass and may be misdiagnosed as malignancy especially in cases of Actinomycosis oral cavity. In literature similar cases are reported in literature in which Actinomycosis mimic not only primary malignancy but sometimes even metastasis.[17]

Most of the actinomyces species are present in polymicrobial flora.[18] Hence, making it more difficult to diagnose or isolate Actinomyces unless when the culture is pure and associated with neutrophils.[19] Once the infection is established, the host produces an intense inflammatory response (suppurative, granulomatous) and fibrosis may develop subsequently. Infection mostly spreads contiguously and invades surrounding tissues or organs. Ultimately, the infection produces draining sinus. Lymphatic dissemination is uncommon, whereas hematologous disseminationto distant organs may occur in any stage of infection.[20]

The management of actinomycosis includes clinical examination, histopathology, isolation and culture of the organism. Histopathology is diagnostic, but it is the direct identification or isolation of the infecting organism from a clinical specimen (draining sinuses, deep needle aspirate, or biopsy specimens) with

demonstration of sulfur granules which is definite for its diagnosis.[9]

In our case diagnoses was made on histopathology examination. Gram stain and culture was non-contributory. All kinds of abscess, cellulite, folliculitis, subcutaneous fat necrosis perceived as mass and malignant formations, and tuberculosis that causes nodules with fistula and nocardiosis should be considered in differential diagnosis.[16]

Conclusion:

Primary cutaneous actinomycosis is a rare sub-acute or chronic suppurative and granulomatous bacterial infection caused by facultative pathogenic commensals. Cervicofacial and thoracic forms can mimic as tumor. Therefore, it's important for clinician to keep a differential diagnosis of actinomycosis to arrive at correct diagnosis. Clinical features, histopathological findings, and cultures are the cornerstones to prevent misdiagnosis.

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Legend Figures

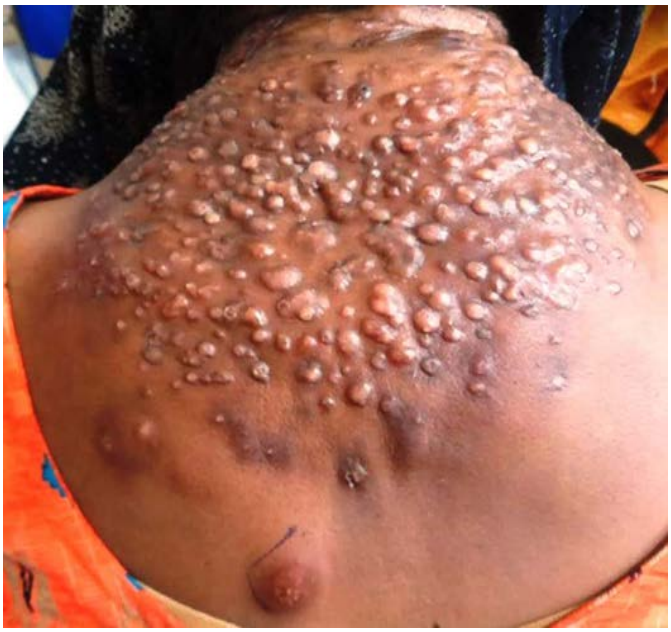

Figure 1: Shows multiple plaque-like lesions.

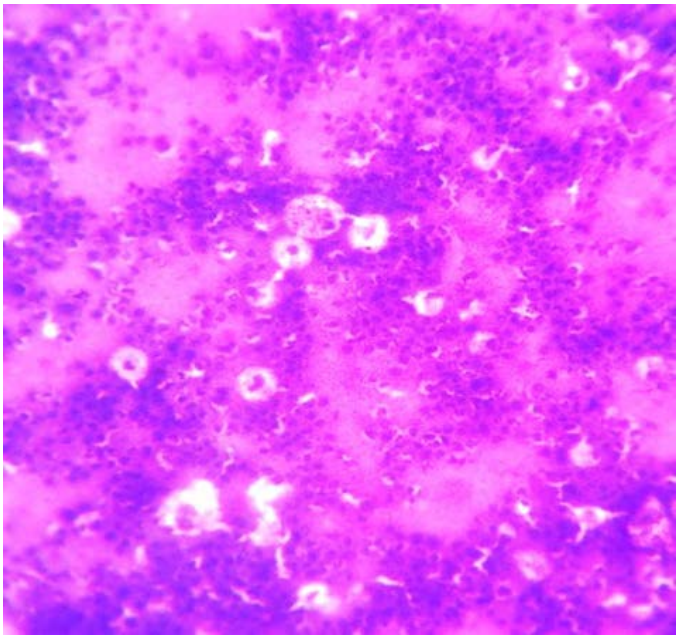

Figure 2: Shows abundant neutrophils with scattered macrophages in a proteinaceous background.

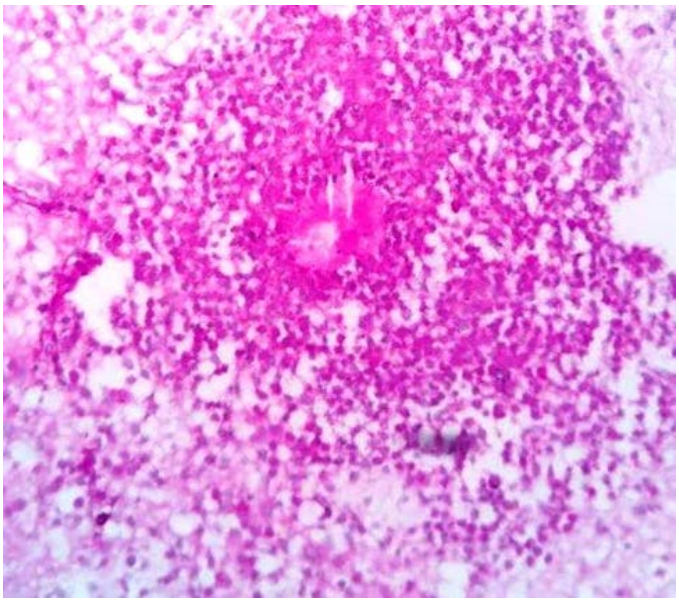

Figure 3: Shows actinomyces colony among purulent exudates.