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Internal Medicine

PRIMARY CUTANEOUS ACTINOMYCOSIS IN SLE—A RARE OCCURRENCE

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ABSTRACT Actinomycosis is caused by filamentous, gram positive, anaerobic to microaerophilic bacteria belonging to actinomyces species. The disease usually manifests as a systemic illness such as cervico-facial, thoracic or abdominal disease. Primary cutaneous actinomycosis is relatively rare and occurs in setting of imunocompromised states such as HIV. Occurrence of actinomycosis in autoimmune diseases such as systemic lupus erythematosus (SLE) has not been previously described. We present herewith a rare case of primary cutaneous actinomycosis in a patient with SLE.

KEYWORDS : Actinomycosis, Cutaneous, SLE, Autoimmune Conditions

INTRODUCTION

Actinomycosis is a sub-acute to chronic infection caused by filamentous, gram positive, non spore forming anaerobic to microaerophilic bacteria belonging to actinomyces spp[1,2]. They normally colonize oral cavity, gastrointestinal tract, and female genital tract[1,2] . While the most common species infecting humans is Actinomyces israelii, more than 30 other species such as Actinomyces meyeri, Actinomyces odontolyticus or Actinomyces viscosus are also reported to cause infection[1,2] . The disease is characterized by contiguous spread and suppurative and granulomatous inflammation which ultimately results in formation of abscesses and sinus tracts which discharge sulfur granules. The disease usually manifests as cervico-facial, thoracic or abdominal disease[2]. Primary cutaneous actinomycosis is relatively rare and occurs in setting of imunocompromised states such as HIV[3].

Occurrence of actinomycosis in autoimmune diseases is not previously described. We present herewith a case of primary cutaneous actinomycosis in a patient with systemic lupus erythematosus (SLE).

CASE REPORT

A 20-year-old male, a known case of SLE with lupus nephritis on immunosuppressive drugs for 10 years presented with a localized swelling on dorsal aspect of right foot.

The swelling had progressively increased in size over one and half months. It was associated with pain. There was no erythema or discharging sinus. There was no history of fever, trauma, cough, dental caries or weight loss. His general and systemic examination was normal. The patient was in complete remission of lupus nephritis on tapering doses of immunosuppressive medications (prednisolone 20 mg per day and azathioprine 100 mg per day).

On local examination, a swelling was present on dorsal aspect of right foot, approximately 6x4cms, non-tender, and firm to hard in consistency. His detailed biochemical and hematological work up was within normal limits. Chest X ray was normal.

Based on above findings, a provisional diagnosis of giant cell tumor was made and a decision for surgical excision of the tumor was made. Post excision the tumor tissue was sent for histopathological examination, which revealed multiple suppurative granulomas enclosed by fibro collagenous tissue. The center of granuloma showed actinomycotic colony.

Based on above findings, the patient was diagnosed as actinomycosis of right foot with SLE. As the diagnosis was made post operatively from the histopathological specimen, microbiological confirmation could not be made. The patient was treated with amoxicillin therapy, initially intravenous followed by oral, which is planned to be continued for a long course of about 6 months or more. While the patient initially had discharge of sulfur granules from the surgical incision site, he responded well to antibiotic therapy subsequently. Presently the wound has completely healed, and patient is asymptomatic. There is no evidence of systemic dissemination of the infection till date.

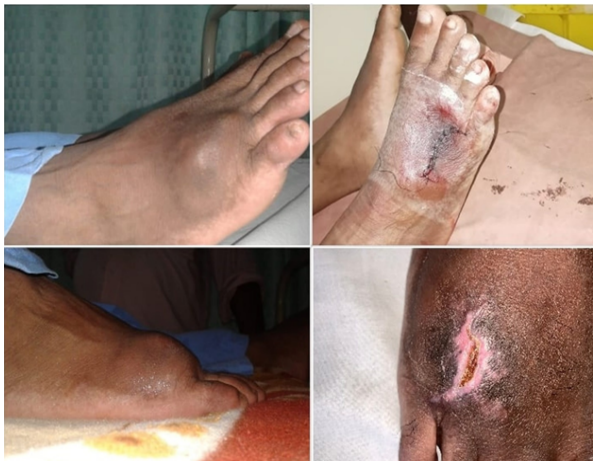

Image 1: photograph showing soft tissue swelling on foot (Left: preoperative, Right: post operative)

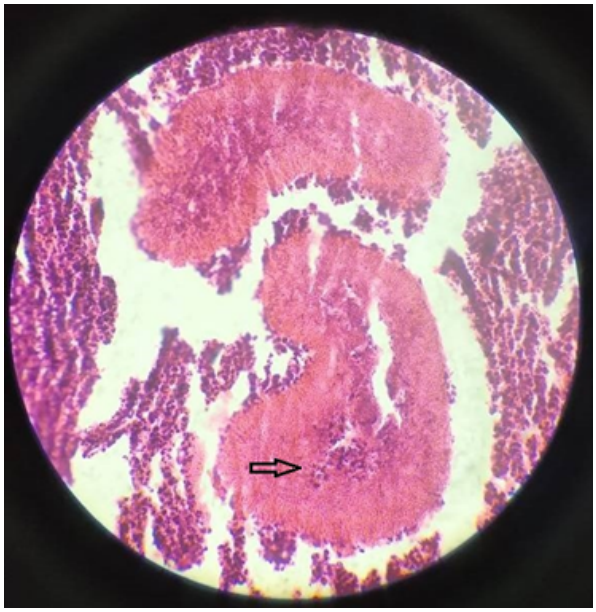

Image 2: photomicrograph of the biopsy specimen showing actinomycotic colony (arrow)

DISCUSSION

The present case is unique for the following reasons

1. It is a case of primary cutaneous actinomycosis.
2. Occurrence of the cutaneous disease in absence of trauma.
3. Occurrence in SLE.

Cutaneous actinomycosis generally occurs as a secondary infection due to dissemination from other sites such as thoracic or abdominal cavity[2]. Primary cutaneous actinomycosis is rare[3-5] and occurs following trauma, which leads to inoculation of the organism at the site[6]. In our patient there was no evidence of any systemic involvement, thus suggesting that it was a case of primary actinomycosis. The patient denied any history of trauma, however possibility of minor injury cannot be ruled out, as the patient was wearing tight shoes at his work place which itself could have caused some cutaneous injury.

Cutaneous actinomycosis is often difficult to diagnose, it is usually mistaken for other chronic skin conditions like cutaneous tuberculosis, soft tissue tumors or neoplasms [1, 7]. Our patient was also initially mistaken to be a case of giant cell tumor of the right foot. However the unusual finding of non adherence of the tumor to underlying skin and bone made us consider alternative diagnosis. The diagnosis of actinomycosis was subsequently made on biopsy specimen.

Actinomycosis is reported to occur with increased frequency in immunocompromised patients or those on immunosuppressive therapy[1, 2]. The disease has been reported earlier in PLHA (persons living with HIV/AIDS) [3,8-10]. However, occurrence of actinomycosis in SLE has not been previously described, hence the present case assumes significance. Our patient was a case of SLE diagnosed 9 years back and was started on maintenance steroid treatment. Subsequently he had a flare of lupus nephritis and was treated with immunosuppressive treatment. The patient is currently in remission on prednisolone 20 mg per day and azathioprine 100 mg per day.

Systemic lupus erythematosus(SLE) is multisystem autoimmune disease resulting in tissue damage due to antibody and complement fixing immune complex deposition[2]. The disease is associated with multiple primary and drug related immunological defects such as lymphocytopenia, low production of interleukins, hypocomplementemia and low cytotoxic T cell response, which predispose patients to infections[2]. It is likely that our patient developed cutaneous infection in the absence of obvious trauma due to his immunocompromised state. Our patient is further unique, as SLE has female preponderance; its occurrence in a male patient is itself less common.

Actinomycosis is treated by prolonged course of antibiotics such as penicillin or amoxicillin for a prolonged period of 6-12 months[1,2]. Our patient responded very well on amoxycillin therapy and is currently asymptomatic on oral treatment. We plan to continue treatment for at least 6 months to avoid recurrence.

In short, our case points to a rare case of actinomycosis occurring in immunocompromised state other than HIV/ AIDS. Thus, in clinical practice, though actinomycosis poses diagnostic challenge, it should be considered in immunocompromised states with appropriate clinical features.

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