Primary cutaneous actinomycosis of scrotal skin: A rare entity often misdiagnosed

Varna Indushekar, I. L. Jeswani, Shikha Goyal, Mukesh Punjabi, Chetan B. Patil
Departments of Pathology and Respiratory Medicine, JLN Medical College, Department of Pathology, Dayalveena Charitable Diagnostic and Research Centre, Ajmer, Rajasthan, India

Address for correspondence:
Dr. Varna Indushekar, Suraj Vihar Colony, Ajmer, Rajasthan, India. E-mail: var9colors@gmail.com

Actinomycosis is one of the most misdiagnosed diseases even by experienced clinicians and listed as a “rare disease” by the office of rare diseases of the National Institutes of Health.[1] It is a chronic suppurative granulomatous lesion producing sinuses with discharge showing characteristic sulfur granules. The primary cutaneous form of actinomycosis is a rare manifestation. We report a case of cutaneous actinomycosis presenting with a chronic nonhealing ulcer on the scrotal skin without involving any extracutaneous sites.

A 60-year-old male presented with 1-year history of chronic nonhealing ulcer of scrotal skin with the recent increase in size since a month associated with pain and discharge since 3–5 days. On examination noduloulcerative mass measuring 5 cm × 3 cm × 1 cm was seen at the base of the scrotum with associated sinus. He was previously treated with antibiotics but showed no improvement. The scrotal mass was excised and sent for histopathology examination.

Grossly the specimen was single soft tissue mass measuring 6 cm × 4 cm × 3 cm, covered with skin showing ulcer with everted margin, covered with fibrinopurulent discharge and firm to hard base. Cut section showed a well‑defined mass with variegated appearance and blackish white nodular lesion extending deep into muscular tissue.

Sections studied were lined by keratinized squamous epithelium. Deep dermis showed granuloma formed by few actinomycosis colonies (sulfur granules) surrounded by an acute inflammatory cells comprising predominantly polymorphs infiltrating deeply into the muscular layer [Figure 1]. Subepithelial tissue showed numerous congested blood vessels.

The acid fast stain was negative with PAS positivity and showed no evidence of malignancy. The final diagnosis of actinomycosis (mycetoma) of scrotal skin was made. Postsurgery, the patient, was treated with intravenous penicillin G for 4 weeks along with supportive treatment.

Actinomyces are a group of filamentous, anaerobic Gram-positive bacteria. Most of the pathogenic actinomycetes occur in soil saprophytic flora. Pathogenic actinomycetes are normal inhabitants of the human mouth, respiratory, intestinal, and genitourinary flora. Hence, acquired endogenously.[2]

Actinomycosis was first reported in humans by Israel in 1878 and in 1891 Wolff and Israel[3] successfully cultured the microorganism. Species are known to cause disease in humans include Actinomyces israelii and less often by Actinomyces naeslundii, Actinomyces gerencseriae, Actinomyces viscosus, Actinomyces odontolyticus, and Actinomyces meyeri.[4]

Actinomycosis occurs worldwide with the higher prevalence rates in areas with low socioeconomic status and poor dental hygiene. Male gender and diabetes are the predominant risk factors, but pathogenesis remains speculative. Males are more commonly affected with male to female ratio 3:1.[4]

Actinomycosis can affect any organ or tissue with five main clinical types cervicofacial, thoracic, abdominal, primary cutaneous, and pelvic presentations. Cervical actinomycosis is the most frequent form of the disease, commonly caused by A. israelii. Disseminated and primary cutaneous cases have also been reported.

Primary cutaneous actinomycosis is least common variety and usually occurs on
the exposed skin. The lesions are in the form of subcutaneous nodules that enlarge into an abscess with discharging sinuses and regional lymphadenopathy. Lesions from the skin may spread to contiguous structures, including subcutaneous tissue, muscle and bone mimicking malignancy. It has an association with preceding trauma, tissue ischemia, and oral-cutaneous contact.

Clinical features that should increase the suspicion of actinomycosis include the presence of multiple discharging sinuses with transient improvement with short courses of empirical antibiotics, and the presence of sulfur granules.

The reasons for the lower culture rate for *Actinomyces* are: (1) *Actinomyces* is very sensitive to a wide variety of antimicrobials which can interfere with their isolation. (2) Strict anaerobic processing required.

In this reported case, the diagnosis of actinomycosis was based on the histopathology with characteristic colonies (sulfur granules) formed by tangled mass of filamentous aggregates surrounded by radiating organisms [Figure 2] which were PAS stain positive and negative on Ziehl–Neelsen acid-fast staining. The typical appearance and staining characteristics of the sulfur granules help to differentiate actinomycosis from cutaneous tuberculosis, sporotrichosis, nocardiosis, and botryomycosis.[5]

The diagnosis is a challenge, and standardized treatment of actinomycosis is not available. Treatment for actinomycosis consists of surgical intervention and appropriate antimicrobial therapy. Penicillin G was most commonly used in the management to cure such lesions and other drugs such as ampicillin/amoxicillin, sulphonamides have also been used.[6] Surgery is indicated for chronic lesions and prognosis is excellent with extended complete antimicrobial therapy.

Rarity of cutaneous actinomycosis of scrotal skin with single discharging sinus mimics other cutaneous lesions hence there is a significant delay in the diagnosis. Hence, the role of surgical excision and histopathological examination had an important role. All the cutaneous lesions with sinus formation at the pelvic and perineal regions have to be examined extensively as actinomycosis most commonly presents with multiple discharging sinuses, because of this fact it is always undiagnosed in a single lesion. Hence, an exclusively single cutaneous lesion requires a high degree of suspicion to diagnose actinomycosis and appropriate extended antimicrobial therapy indicated to prevent recurrence of lesions at other sites.

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