Primary actinomycosis of vulva with inguinal lymphadenopathy

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ABSTRACT

Actinomycosis is a chronic suppurrative granulomatous infection of subcutaneous tissues caused by bacterium Actinomyces israelii. It is a normal commensal of the oral cavity, gastrointestinal tract, respiratory tract, and vagina. Infection is first established locally by breach of mucosal barrier during various procedures, aspiration, trauma, or human bite. Rarely, it may spread through hematogenous and lymphatic system. We present a case of actinomycosis involving the vulva, extending to the inguinal region along with inguinal lymphadenopathy. Involvement of vulva by actinomycosis is uncommon in literature.

Key words: Actinomycosis, lymphadenopathy, vulva

INTRODUCTION

Actinomycosis was first described as a clinical entity 100 years ago. Isreal and Ponfick, in 1891, defined the anaerobic nature of actinomycetes and isolated them in man. Of the five species causing infection in man, Actinomyces israelii is the most frequently isolated. The usual sites of infection are head, neck, thorax, abdomen, and pelvis. Primary actinomycosis of skin is rare and has been reported over upper and lower extremities, gluteal region, face, and breast following trauma or human bite.

CASE REPORT

A postmenopausal 56-year old woman, mother of three children, presented with swelling and discharging lesions on the right side of vulva and groin since past 14 months. The lesion first appeared 14 months ago on the right side of vulva and gradually extended to the right groin. There was no history of any kind of bite, trauma, operative procedure, or insertion of intrauterine device in the past. She had normal vaginal deliveries without any operative intervention. There was a history of occasional sexual intercourse, in the past 10–15 years. General examination revealed no physical abnormality. On local examination, multiple nodules were seen on the right vulva extending up to the right inguinal region [Figure 1]. There was a mild serosanguinous discharge in the inguinal area. Closer examination revealed yellowish white granular deposit on top of the nodule, which on further examination, turned out to be sulfur granules [Figure 2]. There was edema over groin, right thigh, and leg [Figure 3]. Inguinal lymph nodes were enlarged on the right side. Routine gynaecological examination...
was within normal limits. Investigations including hemogram, urinalysis, kidney and liver function tests, ELISA for HIV, and Mantoux test were within normal limits. Chest radiography and ultrasonography of abdomen and pelvis were normal. Cervical Pap smear did not show any evidence of malignancy. Acid fast staining of the purulent material was negative. Crushed sulphur granules on staining with haematoxylin and eosin showed multiple active colonies with surrounding neutrophilic infiltration [Figure 4]. Each colony on staining with MGG stain showed basophilic filamentous centre, and radiating peripheral projections in a sunray appearance (Gupta bodies) [Figure 5]. Fine-needle aspiration cytology from an inguinal nodule on staining showed similar findings along with giant cells and neutrophils. Actinomyces colonies could not be demonstrated in aerobic as well as anaerobic culture media.

Thus the diagnosis of actinomycosis involving subcutaneous tissue of vulva and inguinal region along with spread to inguinal lymph nodes was kept. Crystalline penicillin was started. The patient showed significant clinical improvement within 6 weeks. There was reduction in pus discharge and induration at the site of lesion as well as edema of right limb after 6 weeks of treatment. The patient was shifted to oral doxycycline 100mg twice daily along with oral cotrimoxazole 960 mg daily. The edema decreased significantly and nodules showed healing with scarring after 6 months of treatment [Figures 6 and 7].

DISCUSSION

Common sites of infection are cervicofacial, pulmonary, thoracic, and abdominopelvic regions. Infection has been reported at unusual sites such as soft tissues and metacarpals of hand following punch injuries, and lower
extremities, gluteal region,[4] and face following trauma or human bite. Breast infection presenting as sinus tracts or mass has been reported to occur due to spread of infection following breast feeding, kissing, or bite.[5] Yi reported a case of actinomycosis involving inguinal region in a male patient.[6] Rarely, infection due to actinomycetes may present as lymphadenitis of submandibular region,[7] mediastinum[8] and inguinal region. Our case represents a rare case of actinomycosis involving the vulva, extending to the inguinal region along with inguinal lymphadenopathy. Actinomycotic infection of vulva is a rare occurrence represented by very limited case reports. It may present as nodular mass[9] or multiple sinuses. Although it is known that Actinomyces is a commensal of vagina and infection occurs after breach of the mucosal barrier, the exact cause of infection in our patient could not be ascertained. Minor trauma such as bruising during sexual contact or oral sex may be assumed to cause implantation of the organism.

Quick response to treatment is uncommon in actinomycosis due to fibrosis. In our case, edema of left lower limb was observed, which might be due to chronic inflammation and fibrosis of inguinal lymph nodes. Diagnosis was confirmed on the basis of finding sulfur granules in pus and demonstration of actinomycotic colonies along with Gupta bodies or actinomycotic drusen. Culture is the least reliable method for confirmation, being positive only in 35% of cases.[10] Prolonged antibiotic therapy with penicillin is the treatment of choice. Alternative drugs such as doxycycline, minocycline, and rifampicin can also be used for long-term therapy.

CONCLUSION

The diagnosis of actinomycosis requires a high index of clinical suspicion. Actinomycosis has to be differentiated clinically from other chronic inflammatory skin diseases, such as cutaneous tuberculosis, sporotrichosis, nocardiosis, botromycosis, lymphogranuloma venereum, cat scratch disease, and malignancy. Diagnosis is difficult because of low success rate of culture, as a result of its fastidious nature.

Financial support and sponsorship
Nil.

Conflicts of interest
There are no conflicts of interest.

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