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Case Report

Primary Actinomycosis of the Foot: A Case Report and Literature Review

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Abstract: Actinomycosis is an indolent, slowly progressive, suppurative infection caused by gram-positive branching bacteria of the genus *Actinomyces*. The organism is a part of the oral and gastrointestinal microflora of humans. The most common parts of body involved in actinomycosis are cervicofacial (55% of patients), abdominopelvic (20%) and pulmonothoracic (15%). Involvement of other parts of the body is uncommon and is usually secondary to a lesion in one of the above sites. Extremity involvement can occur secondarily through direct extension or hematogenous spread. Primary actinomycosis of an extremity is very rare. Here we present a case of cauliflower growth over the foot thought to be squamous cell carcinoma but later proved to be primary actinomycosis on biopsy.
Keywords: primary Actinomycosis, microflora, cervicofacial, biopsy

INTRODUCTION

Actinomycosis is an indolent slowly progressive suppurative infection caused by gram positive branching bacteria of genus actinomycosis. These organism are part of natural oral and gastrointestinal flora. The most common parts involved are cervicofacial (55% of patients), abdominopelvic (20%), and pulmonothoracic (15%)[1]. Involvement of other parts are usually secondary to infection in one or the other above sites. Extremities are involved secondarily via direct extension or hematogenous spread. Primary involvement of an extremity is very rare. Here we present a case of actinomycosis of foot which was earlier thought to be squamous cell carcinoma but proved otherwise on histopathology.

CASE HISTORY

A 52 year old male farmer with no comorbidities hailing from eastern Maharashtra came to the out patient department with a cauliflower shaped growth over right foot. He noticed it 2 years back when the swelling was small and associated with itching. There was no history of any discharge from the mass. The mass progressively grew in size over the period of 2 years. Patient had received treatment in the form of oral antibiotics in the past with no benefits.

On examination, the patient was vitally stable. Local examination showed a cauliflower shaped growth over the right foot on the medial aspect, about 15 x 15 cm in size with central ulceration, everted edges and minimal serous discharge [Figure 1]. The surrouding skin was indurated and hyperpigmented. There were no lymphatic streaks, discharging granules, subcutaneous crepitus or palpable inguinal adenopathy. Pulsations were normal.

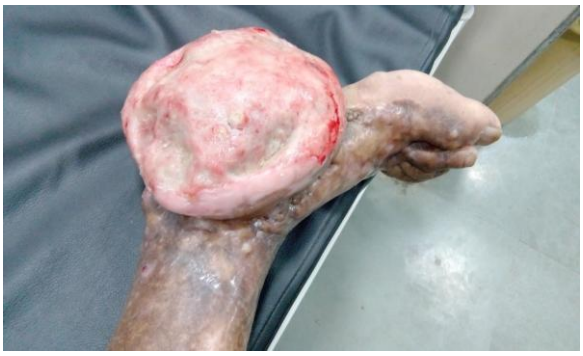


Fig-1

Routine blood investigations revealed a hemoglobin of 5g%, normal leucocyte count and an ESR of 70. X ray of the foot showed features osteomyelitis involving the talus of the foot and medial malleolus four quadrant wedge biopsy was taken and sent for histopathological examination which was suggestive of multiple dermal abscesses with presence of eosinophilic granular material with thin delicate fibrous septae around it resembling actinomycosis [Figure 2-5]. Because of extensive osteomyelitis a

below knee amputation was done after taking consent, stump was closed with a long posterior flap. Patient was given a 21 day course of clindamycin and double strength cotrimoxazole along with hyperbaric oxygen therapy. Stump was healthy and sutures were removed on day 15 and patient discharged with advice to follow up for prosthesis placement. Histopathology of the specimen confirmed the diagnosis of actinomycosis. Patient on follow up after 3 months showed no residual disease and healthy stump.

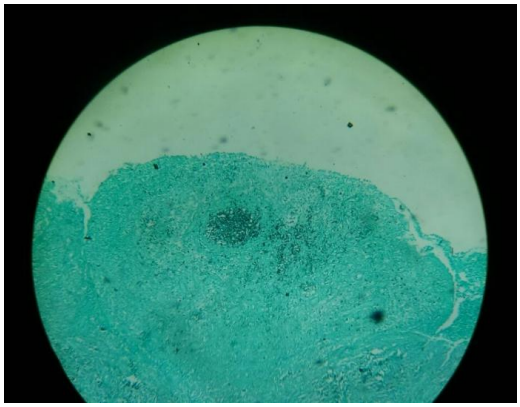
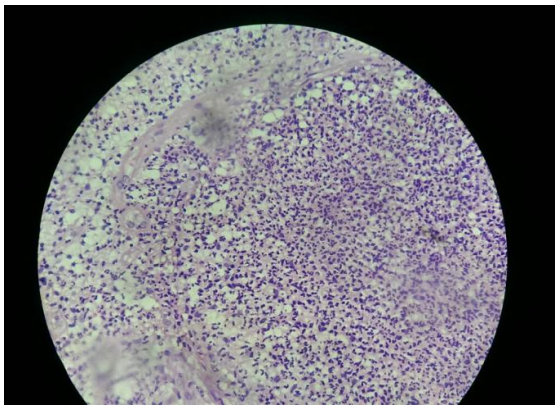


Fig-2



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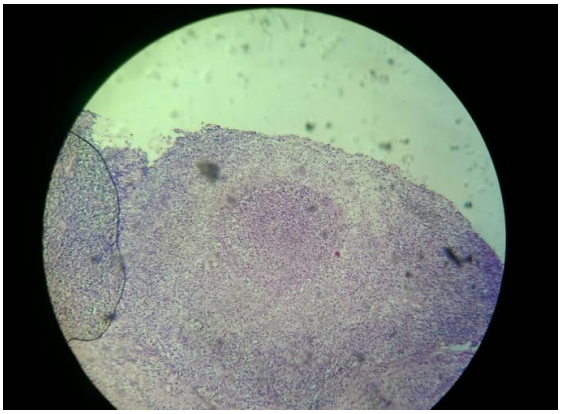

Fig-4

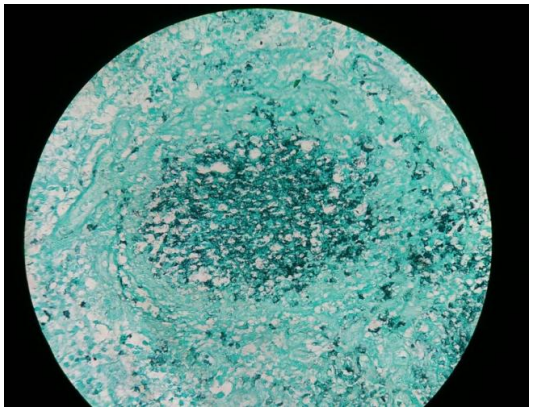

Fig-5

DISCUSSION AND REVIEW OF LITERATURE

Actinomycosis was previously a common disease in humans with the most common etiology being A. *Israelii*. However, with improvement in oral care and antibiotics the disease has become rare [1]. Actinomycosis is an indolent slowly progressive suppurative infection caused by gram positive branching bacteria of genus actinomycosis. These organism are part of natural oral and gastrointestinal flora. Actinomycosis can occur in all ages, with a peak incidenc in the middle decades. Males are infected more frequently than females, at a ratio of approximately 3 to 1 [1, 3-6]. The organism becomes pathological when there is disruption of the mucosal barrier and it gets inoculated into soft tissues where there is impaired blood supply [1, 7]. Spread of the organism occurs through direct extension via tissue planes and rarely via hematogenous route. Thus, systemic disease, extremity involvement, and regional adenopathy are uncommon manifestations [1]. Pathogenesis involves the ability of these organisms to suppress some of the immune functions of the host. When phagocytized by host defense cells, they cannot be destroyed and thus are defined as facultative intracellular parasites similar to *Mycoplasma tuberculosis* in their disease-causing role [1]. This explains the tendency to cause chronic granulomatous inflammation of soft tissues and external discharging sinuses, although the factor responsible for this unique pathogenesis has not been determined [7].

The lesions usually appear as solitary or multiple indurated swellings. As the swellings mature, purulent loculations develop, and central suppuration causes the swellings to become soft and produce discharging sinuses. Microscopically, lesions consist of a dense outer zone of granulation tissue made up of collagen and fibroblasts. The granulation tissue forms multiple loculations with a purulent center containing sulfur granules surrounded by neutrophils, microcolonies of actinomyces, cellular debris, and other associated microorganisms. The sulfur granules are diagnostic. They range in size from microscopic to macroscopic, and each loculation contains 1 to 6 sulfur granules [2].

As stated above, actinomycosis is most commonly reported in 3 body regions: cervicofacial, abdominopelvic, and pulmonothoracic; however, other rare presentations have also been reported. Hematogenous dissemination can occur from the common primary sites and is reported to occur in 3% of cases [1, 2]. The most frequent source of hematogenous spread is reported to be from initial pulmonary infection, following aspiration of contents [9].

Pang *et al* [8] noted that since the first report of anaerobic osteomyelitis by Von Langenbeck, 850 cases have been reported. One percent of all reported cases had osteomyelitis with actinomyces. Only 28 cases of osteomyelitis of the extremity have been reported in the literature, with 11 occurring in the hand and foot [10]. Pang et al also noted only 1 reported case [11] in the English literature of osteomyelitis of the foot confirmed by culture as *Actinomyces israelii* [8].

A correct diagnosis requires a combination of microbiological, molecular, and pathologic studies. A clinical diagnosis starts with obtaining a sample of suppurative exudate, tissue, or sulfur granules. The literature has described many ways to obtain a sample, depending on the location, such as computed tomography-guided core biopsies of the pelvis [12] and ultrasound-guided fine needle aspiration of the thorax [13]. It is extremely helpful if the sample demonstrates grains in the purulent drainage or histologic section of a surgical specimen [2] The grains represent infection that occurs in vivo and are merely a conglomeration of microorganisms. A hematoxylin and eosin stain is used to demonstrate the grain, but a Gram or silver stain is used to show that the grain is composed of branching bacteria. It is imperative that antibiotic therapy be withheld until the sample specimen is obtained; otherwise, actinomyces may not be isolated. If antibiotics have been given already, Gram stain is usually more sensitive than culture. A diagnosis of actinomycosis is considered whenever a direct Gram stain of the suppurative exudate shows gram-positive, non–acid-fast rods in diphtheroidal arrangements with or without branching [2].

Actinomyces can be cultured on a rich medium, such as brain heart infusion blood agar, and incubated anaerobically with added carbon dioxide. The source of actinomyces used for culture are taken from crushed granules or well-mixed pus in the absence of granules [7]. The growth usually appears within 5 to 7 days, but primary isolation may take up to 2 to 4 weeks. Traditional identification depends on the combination of isolates: morphological grown patterns resembling actinomyces species; the tests for urease, catalase, gelatin hydrolysis; and fermentation of cellobiose, trehalose, and arabinose [2].

The drug of choice in the treatment of actinomycosis continues to be penicillin. The principle of therapy began with Peabody and Seabury back in 1960. They have shown that because of the tendency of actinomyces to recur following apparently successful treatment, penicillin is necessary in high doses and for prolonged periods of time. This allows the antibiotic to penetrate the avascular fibrotic walls of the lesion and reach the core of the sulfur granules [14]. Therapy must be individualized, although complicated infections usually require a 2-to-6-week course of 18 million to 24 million units parenteral penicillin G followed by a prolonged course of oral penicillin V or amoxicillin for 6 to 12 months. It is a reasonable approach to also include antibiotic coverage for companion microbes in the initial treatment because many are pathogens on their own.Many different antimicrobials and antifungal drugs have been used with varying degrees of success. Trimethoprim-sulfamethoxazole alone or together with diamino-diphenyl-sulfone is the treatment of choice for actinomycetoma. Amikacin is used for severe cases, unresponsive to previous treatment, and for those in danger of dissemination to adjacent organs.

For penicillin-allergic patients, alternatives such as tetracycline, doxycyline, erythromycin, and clindamycin may be given.Newer agents such as imipenem, ceftriaxone,and ciprofloxacin have been used with anecdotal success. Antimicrobial agents that are not effective against actinomyces are oxacillin, dicloxacillin, cephalexin, metronidazole, and aminoglycosides [15].

A combination of antimicrobial and surgical treatments is used in the treatment of actinomyces infection. Some data indicate that medical therapy alone may be all that is needed, even with extensive disease [16, 17]. Treatment response can by monitored by radiological imaging like CT scan and MRI.In the case of a well-defined abscess, percutaneous drainage is a reasonable option along with medical therapy [18]. few case reports suggest hyperbaric oxygen therapy may be used as an adjunct with antimicrobial and surgical treatment in refractory actinomycosis infections [19,20].

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