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A rare case of actinomycotic osteomyelitis of calcaneum: Presented with subtalar arthritis

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Abstract

We are reporting an unusual case of actinomycosis infection which presented with subtalar arthritis. Actinomycosis is a chronic granulomatous disease caused by Gram-positive anaerobic bacteria. Actinomycosis is very rare and of the cases reported Cervicofacial infection is the most frequent clinical presentation. Hematogenous osteomyelitis at distant sites can occur in the rare instance in immunocompromised or pediatric patients, only a few cases have been reported in healthy patients.

Case Report: A 39years, female patient presented with pain, swelling around the right ankle for 4 years with healed sinuses over the lateral aspect. She was treated for the discharging sinuses of right ankle and foot 5 years back with antibiotics and surgery elsewhere. She had a history of taking treatment (ATT for 6months) for pulmonary TB, 10years back. X-rays and Magnetic resonance imaging showed a distorted subtalar joint with multifocal intraosseous lesions in the calcaneum. Curettage and subtalar arthrodesis with Cannulated cancellous screws was done and supported with short-term immobilization. The tissue culture, fungus culture, acid-fast bacillus (AFB) culture, AFB smear, and tuberculosis polymerase chain reaction test results of intraop samples were negative. A pathologic examination confirmed the presence of actinomycosis. The patient was treated with intravenous penicillin for 2 weeks followed by Oral Doxycycline for 6 weeks, given no evidence of active infection intraoperatively. After 1year follow-up, the patient had no signs of recurring infection or complications and she is ambulant pain-free.

Conclusion: Actinomycosis of the foot, though rare, can mimic tuberculosis in areas with a high prevalence of tuberculosis. In patients with recurrent discharging sinuses, a high index of suspicion is warranted to early diagnose it.

Keywords: Subtalar joint arthrodesis, Actinomycosis, ATT

Introduction

Actinomycosis is a rare, subacute to chronic infection caused by a group of filamentous, Gram-positive, and anaerobic to microaerophilic bacteria, resembling fungi that are not acid-fast. Actinomycosis israelii is the species associated frequently with human infection. The most common clinical presentation of Actinomycosis is of cervicofacial ('lumpy jaw'), thoracic, abdominal, and pelvic in women. Actinomyces exist naturally in the mucous lining of the nose, throat, mouth, digestive tract, and female reproductive tract and are not normally harmful. Any injury, trauma, or surgical procedure can cause the bacterial cells to enter deeper tissues without oxygen, resulting in infection. Typically there will be suppurative and granulomatous inflammation accompanied by the formation of multiple bone abscesses and sinus tracts that may discharge. Orthopedic and dermatological manifestations predominate in disseminated Actinomycosis. We are reporting an unusual case of Actinomycosis affecting the Calcaneum, which presented as subtalar arthritis in a patient with a history of treated pulmonary tuberculosis which leads to diagnostic confusion and prompted me to report the case.

Case Report: A 39-year-old female presented with pain around the right ankle which increases while walking, especially over uneven surfaces for the past 4years. Clinical examination revealed diffuse tender swelling all around the hindfoot and healed sinuses over the lateral aspect with markedly restricted painful subtalar joint movements (Fig.1. Healed sinuses on the lateral aspect of the right ankle). She was treated for the discharging sinuses of right ankle and foot with antibiotics and surgery elsewhere 5years back. She is an Asthmatic on inhalers for 5years.



Fig 1: Healed sinuses on the lateral aspect of the right ankle

Her radiograph showed a reduction of subtalar joint space with a lytic lesion in the anterior calcaneum (Fig.2. Radiograph showing the gross reduction in subtalar joint space and lytic lesion). All blood investigations were within normal limits. MRI showed a distorted Subtalar joint with multiple cavitory lesions in the Calcaneum (Fig.3. MRI

showing distorted Subtalar joint with multiple cavitory lesions in the Calcaneum). She was diagnosed as a case of subtalar joint arthritis secondary to calcaneal osteomyelitis.



Fig 2: Radiograph showing the gross reduction in subtalar joint space and lytic lesion in calcaneum

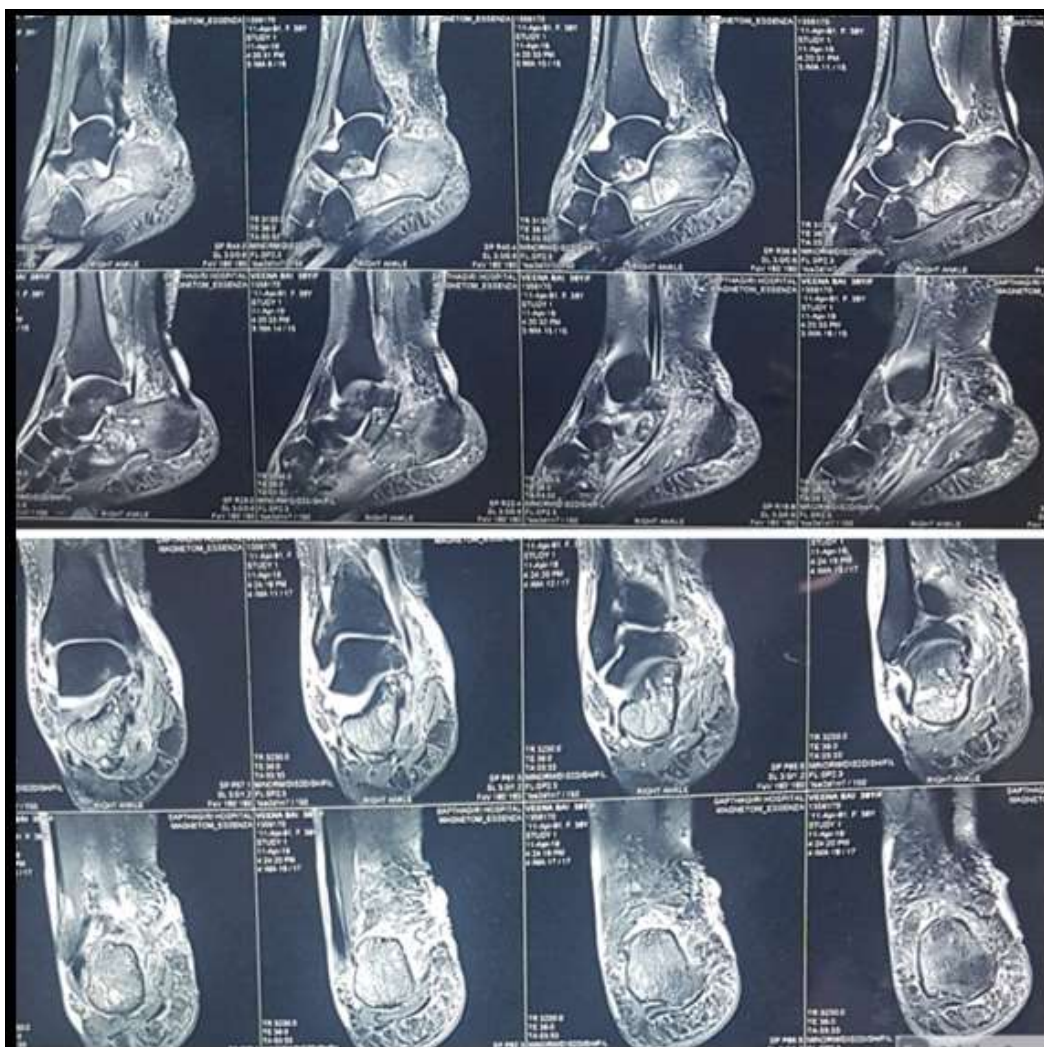


Fig 3: MRI showing distorted Subtalar joint with multiple cavitory lesions in the Calcaneum

She underwent open subtalar joint arthrodesis with a sinus tarsi approach. Articular surfaces of calcaneum were found to be sclerosed. All posterior, anterior, and medial facets of subtalar joints were prepared for sound fusion. Two 6.5mm cannulated cancellous screws were used in compression mode for fixation. Screws were passed from the non-weight bearing

aspect of the heel up to the neck of the talus. (Fig.4. Postoperative radiograph showing fixation). No pus was noticed in the cavities or the subtalar joint. Tissues were scrapped from the cavity, and the material was sent for a histopathology examination.



Fig 4: Postoperative radiograph of right ankle showing fixation

Postoperatively, the patient was immobilized with below-knee plaster splints. She was empirically started on broad-spectrum intravenous antibiotics. Her postoperative course was uneventful. After removal of suture, a below-knee plaster cast was applied for 12 weeks, and the patient was kept non-weight bearing. The tissue culture, fungus culture, acid-fast bacillus (AFB) culture, AFB smear, and tuberculosis polymerase chain reaction test results of intraoperative samples were negative. Histopathology examination showed the presence of deeply basophilic bacterial colonies surrounded by bright eosinophilic material, no necrosis or atypical cells, morphologically suggestive of Actinomycotic colonies (Fig.5. Actinomycotic colonies on HPE).

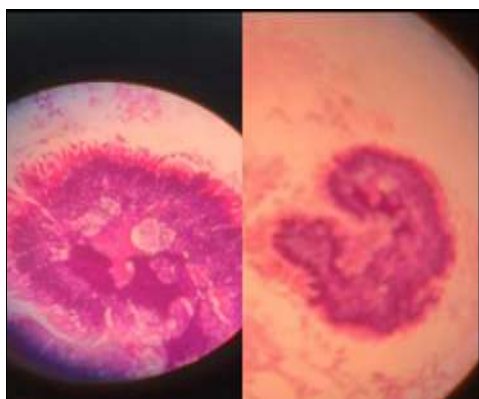


Fig 5: Actinomycotic colonies on HPE

She was treated with Intravenous penicillin for 2 weeks and Oral Doxycycline for 4 weeks. X-rays at 12 weeks post-surgery showed union (Fig.6. Follow-up radiographs at 12 weeks showing union). The patient was advised progressive weight-bearing after 3months and she is symptom-free without recurrence at 1year follow-up.



Fig 6: Follow-up radiographs at 12 weeks showing union

Discussion

Actinomycosis of bone and joints is a rare entity in comparison to tuberculosis. Actinomycosis is caused by gram-positive Actinomyces species, which is the natural flora of the oral cavity, gastrointestinal tract, and urogenital tract [1, 2, 5, 9]. Almost 30 species have been isolated, of which the most common microorganisms causing infections are *A. israelii*, *A. naeslundii*, *A. odontolyticus*, *A. viscosus*, *A. meyeri*, and *A. gerencseriae* [9]. These infections usually involve the cervicofacial, thoracic, and abdominopelvic regions, and no person-to-person transmission has yet been documented [5]. Actinomycosis infection of the bone is mainly due to adjacent tissue infection, but it can also be seen in some fractures or hematogenous spread [7]. The diagnosis of infection is based on characteristic histological and/or microbiological findings. Histopathology is characterized by the presence of Grampositive filamentous structures and sulfuric granules. Gomori methenamine-silver and Giemsa stains are commonly used, and a rapid direct immunofluorescence method has also been developed to identify a species-specific antibody [5, 9]. Identification of the microorganism is successful in less than 50% of cases, owing to the recent use of antibiotics, overgrowth of other microorganisms, and inadequate diagnostic methodology. Gram staining is thus considered a more sensitive technique than is culture. Culturing Actinomyces requires a selective anaerobe agar maintained at 37°C and a growth time of 3 weeks [7].

Antibiotic treatment duration depends on infection localization and severity, and concomitant surgical excision. Actinomyces species are known to be susceptible to penicillins and other β -lactams, except oxacillin, dicloxacillin, and cephalixin. They are also susceptible to doxycycline, clindamycin, erythromycin and clarithromycin, linezolid, and tigecycline [3, 5, 8, 9].

Although the organism is multi susceptible, in most cases of infection, initial intravenous treatment with antibiotics for 2 to 6 weeks (preferably with penicillin G) with subsequent prolonged oral treatment for 6 to 12 months is required [1-6, 8-13].

Actinomycosis of the Calcaneum and subtalar joint as a reason was never thought of because of the absence of typical signs and symptoms of disease and rarity of location. It was only the histopathological study of retrieved material showed the presence of Actinomycosis. In developing countries, tuberculosis of the foot and ankle is the diagnosis most commonly taught. With the patient history of treatment for pulmonary tuberculosis and the absence of a typical history of discharge of sulfur granules, the diagnosis of Actinomycosis is uncertain.

Conclusion

Actinomycosis is a rare disease encountered by an orthopedic surgeon and that too of the lower limb. Because of its varied presentations reported in the literature and its ability to mimic different diseases, it leads to diagnostic confusion and delayed appropriate medical therapy. We emphasize the importance of actinomycosis as a differential diagnosis and early diagnosis with simple gram stain to avoid delayed treatment which can lead to increased morbidity.

Consent: The patient has given their informed consent for the case report to be published.

Competing interests: The author declares that they have no competing interests.

Clinical Message

An unusual case of Actinomycosis affecting the Calcaneum, which presented as subtalar arthritis in a patient with a history of treated pulmonary tuberculosis which leads to diagnostic confusion and prompted me to report the case.

Authors Corner: The presentation of the patient with symptoms of subtalar joint arthritis, and giving a history of treated pulmonary tuberculosis and chronic discharging sinuses, prompted me to think about tuberculosis rather than Actinomycosis. After the histopathological confirmation, reviewing the references about a similar case made me publish this case report.

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