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Case report

Persistent synovial actinomycosis in a native knee joint: A case report

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ABSTRACT

Introduction and importance: Actinomycosis is a chronic granulomatous disease associated with the *Actinomyces* species. This unusual condition, especially in the musculoskeletal system, has been considered a diagnostic challenge due to its initial non-specific symptoms requiring high clinical suspicion and an adequate diagnostic approach for its identification.

Case presentation: We present the case of a 39-year-old Hispanic female with right knee pain and associated purulent secretions for the past four years, who demonstrated persistent synovial actinomycosis despite arthrotomy with cleansing and debridement plus a long-term antibiotic regime.

Clinical discussion: Actinomyces species remain a rare cause of musculoskeletal disease. Its presentation could include localized swelling, tissue fibrosis, sinus tracts, or an abscess, yet these symptoms are not specific, requiring high clinical suspicion to avoid a potential misdiagnosis. Culture in an anaerobic media and pathologic specimens are vital diagnostic tools. Among the treatment alternatives, antimicrobial therapy and surgery are usually required to manage bone and joint infections. Adequate antibiotic selection is crucial, as suboptimal treatment could promote the development of a persistent infection.

Conclusion: This case highlights the diagnostic challenge of synovial actinomycosis, a rarely reported condition in native knee joints. High clinical suspicion is critical as early diagnosis, and adequate management is essential to avoid a persistent infectious process.

1. Introduction and importance

Actinomyces species are slow-growing, gram-positive, filamentous bacillus commonly found in the intestinal and oropharyngeal tract [1]. These bacteria might invade deeper tissues through breaches in the mucosa, causing an insidious and indolent chronic granulomatous disease known as Actinomycosis [2,3]. There are no recent estimates of its incidence due to limited epidemiologic data, especially in developing countries [4,5]. This granulomatous condition has been generally classified by its anatomic involvement with common sites, including oro-cervicofacial, thoracic, and abdominopelvic [2,5]. Cutaneous manifestations, musculoskeletal involvement, central nervous system infection, and disseminated disease have also been described [5].

Nowadays, *Actinomyces* species remain a rare cause of bone and joint infections [2]. We present the case of a healthy middle-aged woman that showed persistent actinomycosis in a native knee joint despite multiple antibiotic regimes and surgical procedures.

This case is reported according to the updated consensus-based surgical case report (SCARE) guidelines [6].

2. Case presentation

A healthy 39-year-old G3P2A1 Hispanic female had presented right knee pain and sporadic purulent drainage for the past four years. The symptoms started after she felt a prick while kneeling on a plastic surface at home. No puncture or wound was noticed. She experienced a warm sensation, progressive swelling, and pustules with purulent oozing on her right knee a few days later. A primary care physician (PCP) prescribed oral antibiotics [i.e., Ciprofloxacin and Amoxicillin-Clavulanic acid for ten days] under the impression of purulent cellulitis.

Abbreviations: PCP, primary care physician; MRI, Magnetic Resonance Imaging; WBC, White Blood Cell; ESR, erythrocyte sedimentation rate; CRP, C-Reactive Protein; ID, Infectious Disease; MUA, manipulation under anesthesia.

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Symptoms persisted despite antimicrobial therapy reason for which she was referred to a general orthopaedic surgeon for further evaluation. She was scheduled for an arthrotomy with cleansing and debridement under the impression of a Septic Prepatellar Bursitis.

Intraoperative cultures show no growth, but the specimen report was remarkable for actinomycosis with associated neutrophilic exudate. Directed antibiotic therapy (i.e., high dose Penicillin G for six weeks) was administered with a temporary resolution of symptoms. A few months later, she started with occasional pustules and drainage managed with different antibiotics regimes over three years. Considering the persistent nature of the symptoms, Magnetic Resonance Imaging (MRI) of the knee was ordered by the PCP. The study revealed *an extensive phlegmon with inflammatory changes, synovitis in Hoffa's fat pad, and inferior joint capsule irregularity* (Fig. 1). Based on these findings, she was referred to our Musculoskeletal Oncology Clinic due to concerns of malignancy within the differential of a suspected recurrent infectious process.

At the musculoskeletal oncology clinic, she reported persistent knee discomfort with intermittent purulent drainage. Constitutional symptoms such as fever, chills, fatigue, weight loss, or malaise were denied. There was no history of surgical procedures before the arthrotomy with cleansing and debridement. Social history was negative for toxic habits (i.e., tobacco, alcohol, or illicit drugs). Physical examination revealed a warm and tender right knee with multiple sinus tracts in the anterior-inferior aspect with serosanguineous drainage. Range of motion and neurovascular function was preserved. Laboratory values showed: White Blood Cell Count (WBC) 5.8 $\times$ 10^3 ml, Erythrocyte Sedimentation Rate (ESR) 63 mm/h and C-Reactive Protein (CRP) 19.8 mg/l. Imaging review in the context of the clinical presentation was suggestive of Persistent Right Knee Infection. Surgery was scheduled for mass (i.e., phlegmon) excision with cleansing and debridement.

The surgical intervention performed by the senior author proceeds with a medial arthrotomy that allows direct visualization of the mass for removal. Cultures were taken, and the joint was irrigated with normal saline and Irrisept®. Then, a combination of 10 cm^3 of Calcium Sulfate and 4 g of Piperacillin-Tazobactam was deposited at the surgical site before a layered closure. The excised mass was sent to pathology for evaluation. Intraoperative cultures show no growth in five days, but the pathologic report revealed *basophilic filamentous structures surrounded by acute inflammatory cells consistent with Actinomyces species* (Fig. 2). Infectious Diseases (ID) service was consulted, and the patient was started on Penicillin G 5 million units IV every 6 h based on their recommendations. On postoperative day eight, the patient was discharged to continue therapy under home IV service to complete 42 days of treatment.

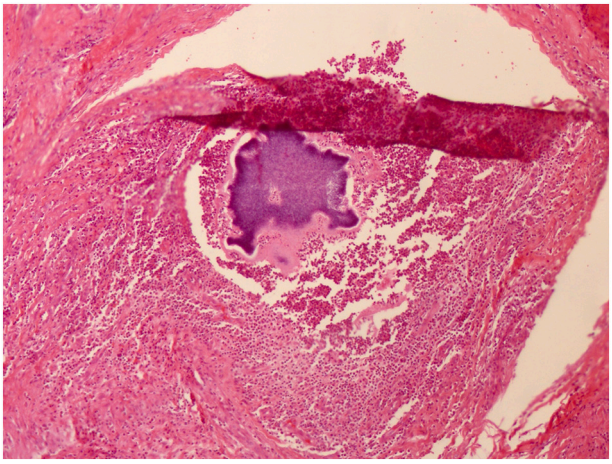

Fig. 2. Surgical specimen on hematoxylin & eosin stain showing basophilic filamentous structures with a configuration consistent with *Actinomyces* species.

She demonstrated improvement in redness, swelling, and purulence on the right knee; inflammatory markers persist on descending trend. Based on clinical response, the therapy was transitioned to Penicillin VK 1 g oral every 6 h up to a year. However, the orthopaedic service was recontacted due to right knee stiffness. The evaluation showed a flexion-extension arc between 15 and 70°. She was scheduled for manipulation under anesthesia (MUA) due to contracture secondary to infectious arthritis.

The procedure improved the arc of motion to 10–100°. She continued physical rehabilitation and antibiotic therapy. ID follow-up demonstrates clinical improvement, preserved range of motion, and inflammatory markers within normal limits. A follow-up knee MRI seven months after MUA displays *persistent inflammatory changes surrounding the patellar tendon and synovitis changes in Hoffa's fat pad with no abscess or fluid collection* (Fig. 3). These findings are indicative of a persistent infectious process despite clinical improvement. After twelve months from surgery, she continued antibiotic and hyperbaric oxygen therapy.

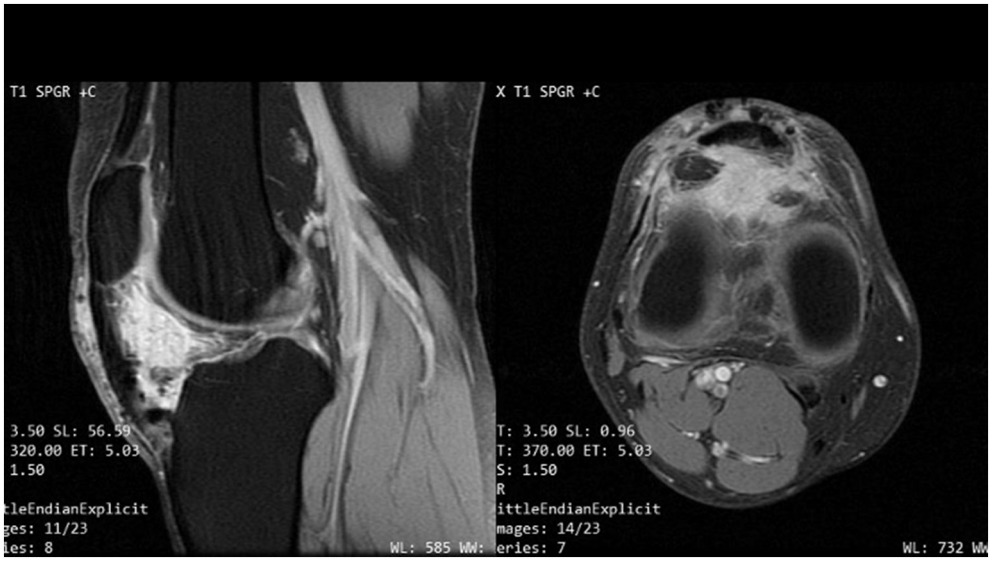

Fig. 1. Contrast-enhanced right knee Magnetic Resonance Imaging (MRI) demonstrating extensive inflammatory changes, a phlegmon, and enhancement in the Hoffa's fat pad compatible with an infectious process.

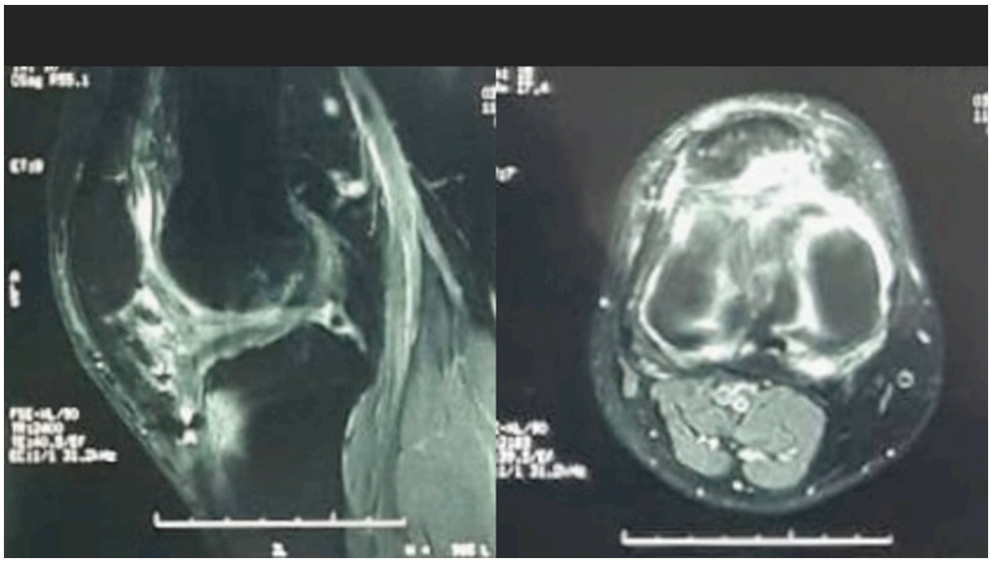

Fig. 3. Contrast-enhanced right knee Magnetic Resonance Imaging (MRI) demonstrating phlegmonous inflammatory changes in the Hoffa's fat pad suggestive of a persistent infectious process.

3. Clinical discussion

Actinomycosis, a condition that induces suppurative and granulomatous inflammation, has been recognized for over 150 years [7]. This disease is usually misdiagnosed due to its capacity to mimic other conditions such as malignancy or tuberculosis, requiring a high level of suspicion for an adequate diagnosis [4]. Its presentation could include localized swelling with suppuration, sinus tracts formation, tissue fibrosis, or abscess formation [7]. Currently, *Actinomyces* species remain a rare cause of musculoskeletal disease [2,3], with most cases reporting osteomyelitis or prosthetic joint infections. This case presented a healthy middle-aged woman who showed persistent actinomycosis in a native knee to contribute to the scarce literature regarding its clinical course, management, and outcomes in native joints.

Early diagnosis of actinomycosis is often challenging due to the non-specific nature of its symptoms [5]. Our patient showed signs (e.g., swelling, warmness, and purulent secretions) suggestive of an infectious process. Consequently, this presentation was initially misdiagnosed as cellulitis, and therefore, the initial treatment regime does not target *Actinomyces* species.

Appropriate cultures and pathologic evaluation are essential for diagnosis. An anaerobic media is crucial, and pus is the preferred specimen for microbiologic sampling [7]. Conventional biochemical tests are available for most species but are usually demanding. Strains frequently demonstrate indifferent growth leading to false-negative results and poor reproducibility [5]. Additionally, prior empirical treatment further reduces the sensitivity of the culture [3,8]. In tissue samples, *Actinomyces* species are infrequently visible, generally requiring special staining techniques [7,9]. Nonetheless, sampling from different tissue levels could improve the histopathologic diagnosis [7].

Among the antimicrobial alternatives, Penicillin G remains the drug of choice for *Actinomyces* species [7]. It is prescribed in high doses (10–20 million units/day) over prolonged periods (2–6 weeks), as the infection tends to recur [7]. Those cases with initial sub-optimal therapy (i.e., short treatment length or poor antibiotic selection), as in this case, could potentially lead to antibiotic resistance and, consequently, a persistent infection [7,8,10].

A combined approach with antibiotic therapy and surgical resection is commonly required, especially on extensive or recurrent disease [8]. Most cases involving prosthetic joint infections undergo a two-stage procedure for prosthesis exchange, using an antibiotic spacer followed by reimplantation of a permanent prosthesis after infection clearance [11]. Concerning native joints, few cases are reported [12–15]. Bose et al. reported the first case of actinomycoses in the knee joint synovium in which the patient underwent long-term treatment with penicillin successfully [12]. Previous literature about bone and joint manifestations recommended surgical debridement if evidence of bone exposition or abscess formation [3]. However, Kundu et al. reported a case in which the patient had to undergo an above-the-knee amputation as the infection significantly worsened despite therapy [13]. Our patient exhibited a persistent illness despite clinical improvement with no functional limitations or signs of disease spreading. Thus, further surgical management is unnecessary, and the patient remains on antibiotic therapy.

This case highlights the diagnostic challenge of synovial actinomycosis, a rarely reported condition in native knee joints. High clinical suspicion is critical as early diagnosis, and adequate treatment is essential to avoid a persistent infectious process.

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Ethical approval

This study is exempt from ethical approval in our institution.

Informed consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Author contribution

Manuel Ramírez-González, MD: Conceptualization, investigation, data curation, and writing.

Norberto J. Torres-Lugo, MD: Investigation, data curation, visualization, writing, and editing.

Luis Bisbal-Matos, MD: Conceptualization, investigation, data curation, and writing.

Gerardo Olivella, MD, MPH: Investigation, Data curation and writing.

Hector Sánchez-Fernández, MD: writing, reviewing, and editing.

Juan Bibiloni-Rodríguez, MD: Investigation, supervision, reviewing, and editing.

Research registration

Not applicable.

Guarantor

Manuel Ramírez-González, MD and Norberto J. Torres Lugo, MD.

Declaration of competing interest

The authors have no conflict to disclose.

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