

Refractory actinomycosis of the humerus

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Abstract

Actinomycosis is a chronic, opportunistic infection caused by *Actinomyces* species, such as *Actinomyces bacillus*. Actinomycosis in long bones is very rare. To the best of our knowledge, isolated primary actinomycosis of the humerus is rarely reported in literature. We present a rare case of a refractory primary actinomycosis of the humerus. A 66-year-old man with no history of concomitant conditions was admitted to our hospital with a history of a tumour on the distal third of the left arm as a result of a closed trauma without fracture 20 years before. Pathological anatomy samples showed the presence of *Actinomyces*. Cultures were subjected to a prolonged incubation of 21 days under aerobic and anaerobic conditions and were always negative. He underwent several surgical procedures and received long-term antibiotic therapy with poor outcome. Primary actinomycosis in long bones is uncommon. Diagnosis may be challenging: considering the small number of case studies reported in the literature, symptoms are not specific, and the organism is difficult to isolate. Antibiotic treatment may not be sufficient to improve the clinical condition, and surgical alternatives should be considered

Keywords

Orthopaedics/Rehabilitation/Occupational therapy, surgery, infectious diseases

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Introduction

Actinomycosis is a chronic, opportunistic infection caused by *Actinomyces* species, such as *Actinomyces bacillus*. First described in 1878 and originally classified as a fungus due to its branching character, today it is known to be an anaerobic bacterium which is part of the normal flora of the oropharyngeal, gastrointestinal and vaginal tracts. It causes cellular ‘hypersensitivity type IV’, which generates a granulomatous inflammatory disease¹ whose clinical symptoms may be similar to those of other infectious diseases or even mimic malignant processes in different anatomic areas.^{2,3}

At the osseous level, it may be confused with tuberculosis or multiple myeloma.⁴ Actinomycosis in long bones is very rare.⁵ To the best of our knowledge, isolated primary actinomycosis of the humerus is rarely reported in literature.⁶

The purpose of this study is to present a rare case of a refractory primary actinomycosis of the humerus.

Case report

A 66-year-old man with no history of concomitant conditions was admitted to our hospital. He had been operated on

due to a tumour on the distal third of the left arm as a result of a closed trauma without fracture 20 years before. Several debridement surgeries were also performed on his lesion, but no detailed information is available. Physical examination revealed new lumps with seropurulent discharge 7 years after the first surgery. Subsequent blood analysis did not show alteration in acute phase reactants, and hematimetric indexes were within normal ranges. For the next 20 years, he continued to experience similar episodes of sporadic reactivation of the lumps.

Before 4 years, he was admitted to our hospital due to painless lumps fixed on the subcutaneous tissue of the left arm (Figure 1). Radiographs of the left humerus revealed an alteration of the bone structure with distal diaphyseal involvement and thickening and cortical irregularity, which

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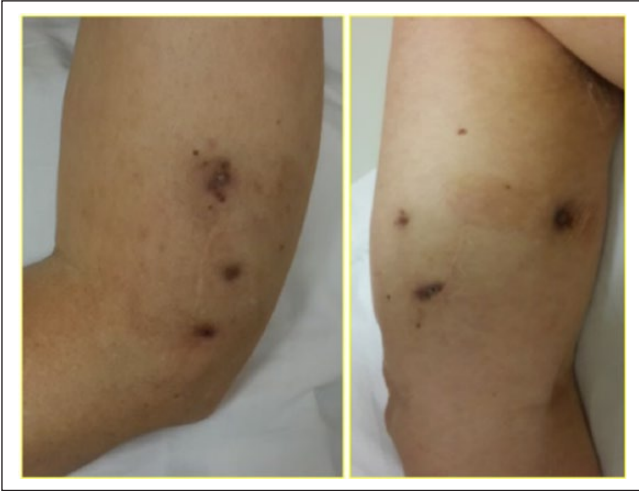


Figure 1. First admission to our institution, 4 years ago. Lumps fixed on the subcutaneous tissue of the left arm.



Figure 2. First X-rays showing (AP and lateral view) distal diaphyseal involvement. The arrows show the thickening and cortical irregularity.

extended up to the metaphysis (Figure 2). A computed tomography (CT) scan disclosed an alteration in the osseous morphology of the metaphysis–diaphysis in the distal third of the humerus at an intramedullary and cortical level, with osteolysis, periosteal reaction, diffuse involvement of adjacent soft tissues and several lower density, superficial, poorly defined liquid areas (Figure 3). A magnetic resonance imaging (MRI) scan confirmed the multiple liquid deposits on the thick walls of the soft tissues and muscular oedema related to joint effusion (Figure 4).

Aspiration was performed, and the extended coloured (Diff-Quick) haematoxylin–eosin, pap-test showed lymphocytes, leukocytes, macrophages, multinucleated giant cells and accumulations of bacillary structures (*Actinomyces*),

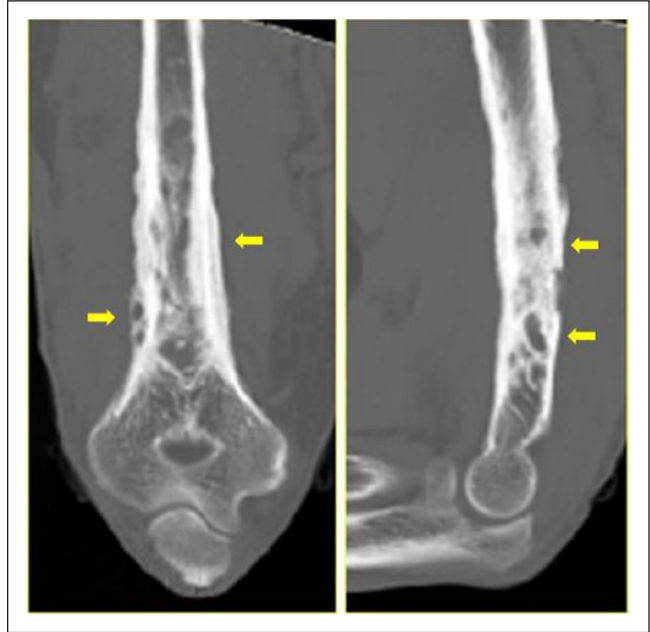


Figure 3. CT scan, arrows showing osteolysis and periosteal reaction. Diffuse involvement of adjacent soft tissues.

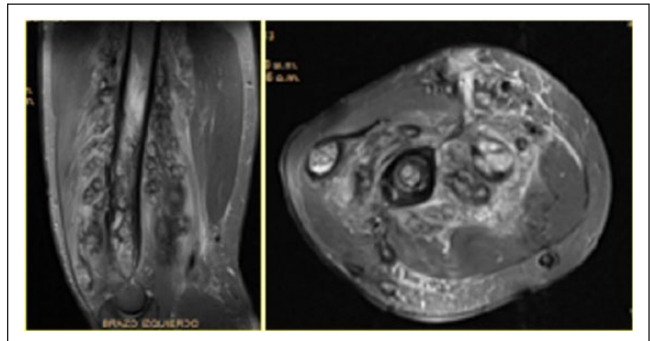


Figure 4. MRI showing multiple liquid deposits and muscular oedema related to joint effusion.

consistent with a granulomatous inflammatory process (Figure 5). Cultures (glucose asparagine agar, arginine-glycerol and tyrosine agar mediums) were subjected to a prolonged incubation of 21 days under aerobic and anaerobic conditions and were negative.

The patient underwent a surgical resection of necrotic tissue and insertion of an antibiotic cemented rod (vancomycin) through a lateral approach of the arm (Figure 6).

Samples were cultured and sent to pathological anatomy. The patient developed a radial palsy, which resolved spontaneously in 6 weeks. Cultures were also negative, and pathological anatomy revealed the presence of gram-positive filamentous bacilli consistent with bacterial colonies of *Actinomyces*. At the osseous level, histological sections evidenced important morphological changes in the fibrofatty medullary bone with lymphocytes, plasma cells and

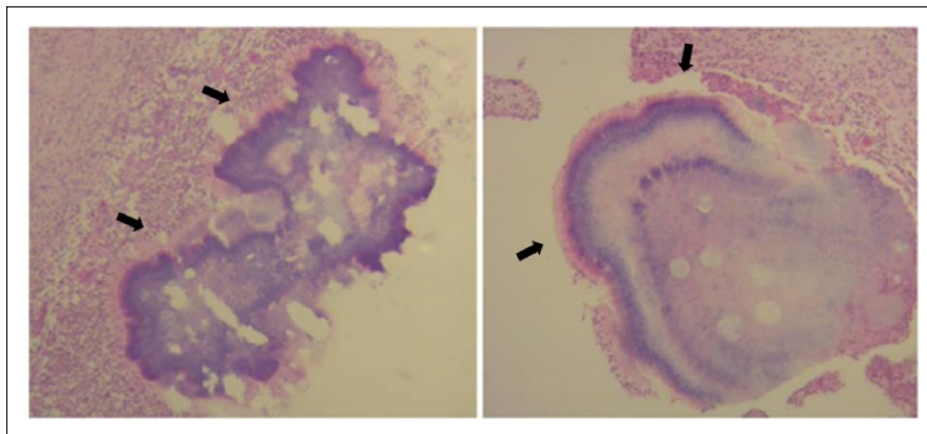


Figure 5. Digital anatomic pathology images (haematoxylin–eosin). The arrows indicate colonies of *Actinomyces*.



Figure 6. Immediate postoperative X-rays (AP and lateral view) after introducing first cement spacer with antibiotics.

polymorphonuclear cells. The patient received intramuscular penicillin G, 4,000,000 IU every 6 h for 6 weeks, followed by oral amoxicillin for 6 months. Clinically, his symptoms improved, but new active cutaneous fistulas reappeared after 2 years.

He underwent a reoperation, and the antibiotic cement was replaced, followed by excision of the necrotic cortical tissue. The antibiotic therapy consisted of 2 g of intravenous ceftriaxone every 24 h for 4 months.

Despite this last procedure, fistula formation continued a year later. Radiology studies and an MRI scan evidenced that the disease had spread to the metaphysis of the proximal humerus (Figure 7).

Consequently, wide surgical resection of the humerus was performed, in which the proximal metaphysis was preserved. An antibiotic cemented rod with vancomycin was inserted (Figures 8 and 9). The new results from pathological anatomy continued to disclose the presence of colonies of *Actinomyces*. Cultures were still negative.

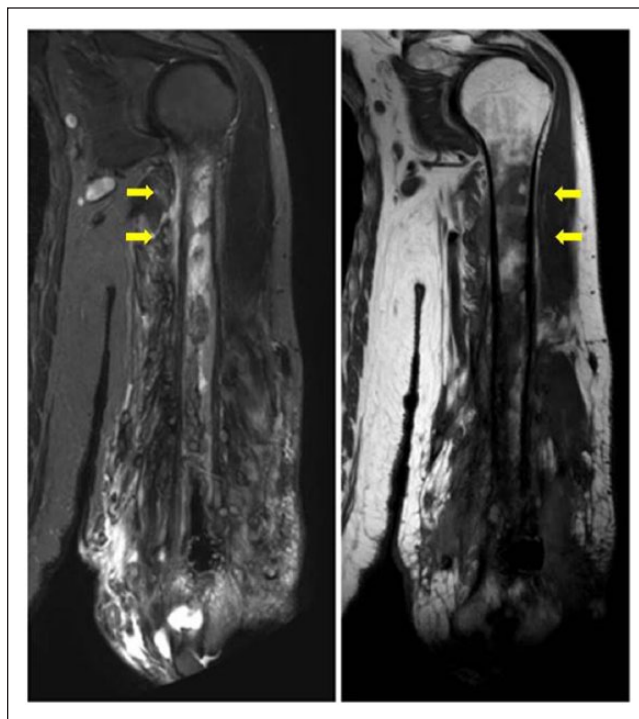


Figure 7. MRI at 3-year follow-up after first index procedure. Disease progression and spreading to the metaphysis of the proximal humerus (arrows).

Subsequently, two new debridement surgeries of the fistulas were carried out. After 6 months, symptoms continued, the rod was replaced with a new one and antibiotic beads were inserted in the fistular areas (Figure 10). Cultures were negative for *Actinomyces*, but the patient developed a *Staphylococcus epidermidis* infection, which further complicated his clinical condition. The new samples sent to pathological anatomy continued to disclose the presence of colonies of *Actinomyces*. The future therapeutic plan includes eventual total humerus reconstruction with prosthesis, once the infection has been completely eradicated.



Figure 8. Wide surgical resection of the humerus. Proximal metaphysis was preserved.



Figure 9. X-ray showing first antibiotic-coated rod.

Discussion

The prevalence of *actinomyces* is estimated at 1 in 119,000 inhabitants.⁷ The *Actinomyces* bacillus builds up, forming inflammatory granulomas surrounded by a pseudocapsule, which gives it the microscopic appearance of sulphur granules.⁸ It is most commonly found at the cervicofacial, abdominal and chest levels.⁹ Approximately 5% of cases occur in the limbs.¹⁰ There are few case studies reported in literature regarding isolated actinomycosis in the upper limbs.

The most frequently isolated variety in humans is *Actinomyces israelii*; however, there are also records of *Actinomyces naeslundii* and *Actinomyces meyeri*.¹¹ Only 50% of cases show positive cultures, and specific conditions are required for bacterial growth, including a minimum of 21 days' incubation.⁹ Open biopsy may be an important diagnostic tool



Figure 10. X-rays showing last cemented rod with antibiotic beads.

and must be considered in difficult cases.⁵ In this case, the samples analysed never revealed *Actinomyces* growth. However, histologic findings always confirmed the diagnosis.

Treatment guides are based on small case series and there is no consensus. Given the absence of an antibiogram, treatment becomes more difficult. Identification of the species is critical, due to the existence of resistant strains. In vitro studies establish that the *Actinomyces* is sensitive to a wide range of antibiotics.¹² Although empirical treatment usually provides good results, this can cause bacterial decapitation. However, the duration of the standard treatment is not well defined, and it may continue for years.¹³ Long-term antibiotic therapy is recommended based on clinical experience. Lack of correct initial treatment is the main predictor of morbidity.¹⁴ In this index case, the initial treatment was unknown, and we may only confirm that the first antibiotic therapy was received in our institution.

Regarding physiopathology, direct inoculation is the usual cause of limb involvement. At the osseous level, the infection depends on its contiguous spread to adjacent tissues, and it is rarely seen in fractures. However, in some cases, it may be due to haematogenous spread.¹⁵ Other forms of transmission described are those following odontogenic outbreaks in people with periodontal infections or in women with intrauterine devices. Direct disruption of the mucus will act as an entry point. These types of infections are usually associated with immunosuppressed patients. Regarding our (immunocompetent) patient, the origin of the infection remains uncertain, even though it may be attributable to the trauma suffered 20 years ago.

Radiological features of actinomycosis include both destruction and formation of bone. It does not discriminate between cortical and cancellous bone, and it even attacks the subchondral bone. Despite multiple lytic lesions, its spheroid nature distinguishes it from bone tumours.¹⁶

Diagnosis may be challenging: considering the small number of case studies reported in the literature, symptoms are not specific, and the organism is difficult to isolate.

Conclusion

The purpose of this study is to report a rare pathology in the upper extremity and its torpid development, refractory to conventional treatment. Primary actinomycosis in long bones is uncommon. Antibiotic treatment may not be sufficient to improve the clinical condition, and surgical alternatives should be considered.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship and/or publication of this article.

Ethical approval

Our institution does not require ethical approval for reporting individual cases.

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Informed consent

Written informed consent was obtained from the patient for their anonymized information to be published in this article.

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