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RESEARCH ARTICLE

RARE CASE REPORTING OF ACTINOMYCOSIS AND MYCETOMA OSTEOMYELITIS OF DISTAL RADIUS IN TWO DIFFERENT CASES: RARE CASE SERIES

Dr. Jayashree Tamatam[1], Dr. Hemant Sharma[2], Dr. Rajesh Kumar Verma[3], Dr. Asgar Ali[4], Dr. Lalit Kumar[5], Dr. Guruditta Khurana[6], Dr. Shallini Mittal[7] and Dr. Nikita Jajodia[8,*]

[1]Fellow, Joint Replacement, Sports Injury and Trauma, Department of Orthopaedics, Marengo Asia Hospitals, Gurugram, Haryana, India – 122001; [2]Chairman, Joint Replacement, Sports Injury and Trauma, Department of Orthopaedics, Marengo Asia Hospitals, Gurugram, Haryana, India – 122001; [3]Director, Orthopaedics, Trauma & Spine Surgery, Department of Orthopaedics, Marengo Asia Hospitals, Gurugram, Haryana, India – 122001; [4]Consultant, Orthopaedics and Spine Surgery, Department of Orthopaedics, Marengo Asia Hospitals, Gurugram, Haryana, India – 122001; [5]Senior Consultant, Orthopaedics & Spine Surgery, Department of Orthopaedics, Marengo Asia Hospitals, Gurugram, Haryana, India – 122001; [6]Senior Resident, Orthopaedics and Joint Replacement, Department of Orthopaedics, Marengo Asia Hospitals, Gurugram, Haryana, India – 122001; [7]Senior Consultant, Department of Radiology, Marengo Asia Hospitals, Gurugram, Haryana, India – 122001; [8]Research Manager, Orthopaedics and Spine Surgery, Department of Orthopaedics, Marengo Asia Hospitals, Gurugram, Haryana, India - 122001

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***Corresponding author:**
Dr. Nikita Jajodia

ABSTRACT

Actinomyces and Eumyectoma is characterized by contiguous spread, suppurative and granulomatous inflammation and formation of multiple abscesses and sinus tracts that may discharge sulfur granules. It is relatively uncommon and it is rarely affects the upper limb bony structure. To our knowledge there is a dearth of literature where these affect the Radius bone. Here we describe a follow up case series of osteomyelitis of distal end radius caused by Actinomycosis & Mycetoma who were immunocompetent having no other co-morbidity or medical conditions.

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INTRODUCTION

Eumycetoma mainly infects younger male field workers in rural area. It usually results from small traumatic implantation of the causative agent in the subcutaneous tissue and the most common site of infection is the foot, followed by the hand, but sometimes other body parts may also be infected. While, the Actinomycosis commonly involves the respiratory tract (oropharynx), genitourinary tract, digestive tract, central nervous system, skin, and soft tissue structures. The clinical presentation of Mycetoma can sometimes be misleading causing delay in the diagnosis. We present 2 cases of young males with Mycetoma of the distal end of the radius that had been erroneously diagnosed as soft tissue tumors. The patients were successfully treated with surgical excision followed by treatment with antibiotic beading, antibiotics and antifungals with a follow up of 3 years.

CASE 1

20-year-old male patientfrom Nepal, working as a white collar worker presented with non-radiating on and off pain with gradual developing swelling over his left wrist. His symptoms began with pain and swelling without any history of trauma 7 months ago from his date of visit to the doctor. The swelling was immobile in all directions and was fixed to the underlying structures. He self-medicated himself for the pain without seeking any opinion. He gave a past history of external fixation

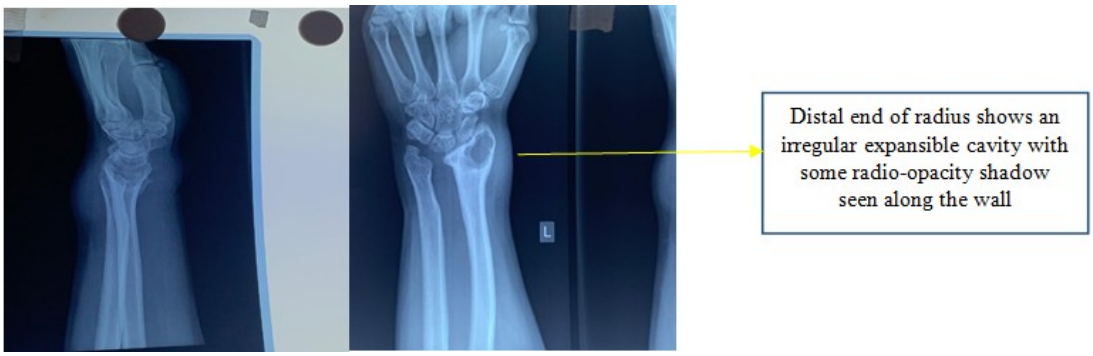

Fig. 1. Pre-operative XRAY of left forearm showing infective site at the distal end of Radius (Lateral and AP View)

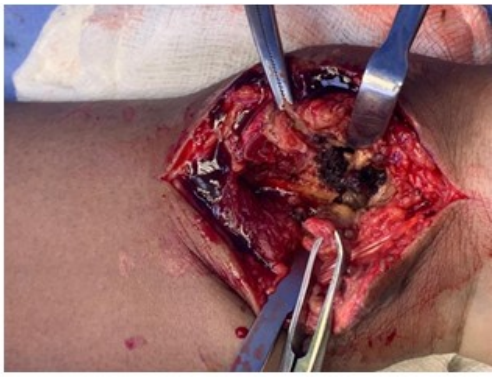

Fig 2. Site of Osteomyelitis.

Fig 3. Microbiology Sample showing Sulphur granules

Fig 4. Post-Surgical site (Volar & Dorsal aspects)- Intact sutures

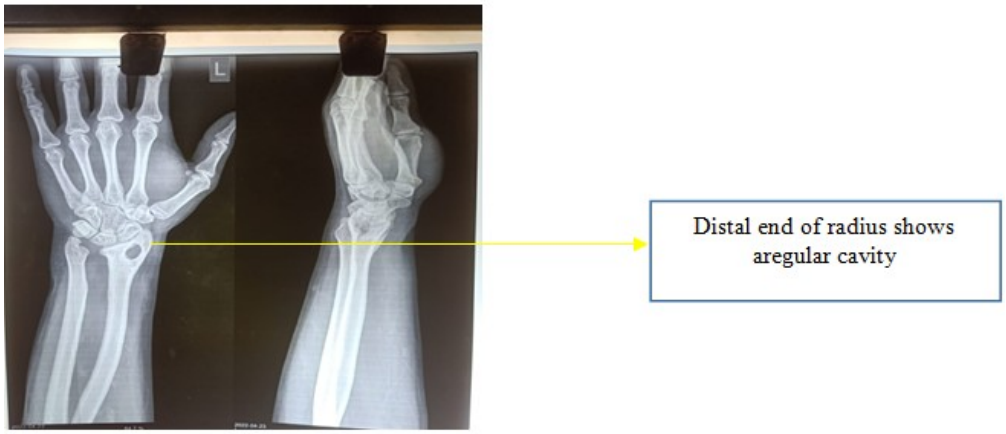

Fig 5. Post- Op XRAY of left forearm (AP and Lateral View)

of the left distal radius 7 years back for compound distal radius fracture. He presented with no fever and his personal history was insignificant. Upon his visit to the hospital, local examination revealed a swelling size 4*4 cm over distal end left forearm with a healed scar mark and a scab of size 1* 0.5cm over volar aspect. There was no raise in temperature, the distal left forearm showed no tenderness, there was no motor or sensory deficit. All Range of Motion's were normal. Hence, the patient was admitted for further management, where he was diagnosed with Actinomycosis Osteomyelitis of the distal end of the radius via the Fine Needle Aspiration. He then underwent medical therapy, surgical debridement, biopsy, curettage & anti-biotic beads insertion. His biopsy revealed multiple dark brown soft tissue pieces measuring 1.5*1*0.5cm. The tissue section examination showed tightly packed fungal hyphae with inflammatory exudate and cell debris with no evidence of Malignancy in the tissue samples. His AFB cultures showed no Acid Fast Bacillus. Biopsy specimens from the tissues confirmed the diagnoses of chronic Actinomyces Israelii. The routine biochemical and hematological tests including erythrocyte sedimentation rate were within the normal range. The X-ray of the upper extremity revealed no significant abnormalities following which an antibiotic course of Ampiciline/Salbactum for a period of one month was initiated. The patient was followed up for 2 years where the patient reported complete resolution of the infection and no hindrance in any activity.

The surgical intervention included taking the patient in Supine position, incision given at the volar aspect interval which was elated between Palmaris longus and Flexor Carpi Radialis. The pus from the wound was taken and sent for culture and sensitivity. Black necrotic tissue was collected from the lesion which was additively sent for culture, sensitivity and histopathology. In the dorsal aspect, after the longitudinal incision, sac along with the unhealthy tissue was excised and sent for culture, sensitivity and histopathology. A cavity was noted that was seen communicating to the volar aspect. The wound was washed thoroughly with Betadine and hydrogen peroxide. The dorsal lesion was sealed with bone wax. The Lesion was filled with 10CC Stimulac after the wound was cleared both at volar and dorsal region. The surgery included wound debridement, biopsy, curettage & Anti-biotic beads insertion in the lytic lesion under General Anesthesia. Post-surgical course was uneventful. The length of stay was one day. Patient was discharged in a stable condition. The medical management included Voriconazole 200mg, thrice daily for 3 days followed by twice daily for 27 days.

CASE 2:

A 31 year old male patient from Sudan, working as a daily wager presented with pain and swelling over right wrist sine 6 months. Pain was throbbing in sensation, non radiating, intermittent mildly relieved with medication. Swelling was non progressive, hard in consistency, non-mobile. Patient had surgical scar over the swelling due to some unknown procedure done at his place.

Upon his visit to hospital, examination revealed 3*0.5cm surgical scar over the dorsal aspect of right wrist with discharging sinus and tenderness present over distal radius. Swelling was hard in consistency, non-mobile with pinchable skin over the swelling. There was local rise of temperature. There was no distal neurovascular deficit. All the range of motion was normal. Patient was then admitted and further investigations were done. Xray of right wrist showed irregular multilocular cystic lesion with radio-opaque shadow along the wall was seen. Patient was counseled for surgical intervention. After obtaining valid informed consent, patient was taken up for thorough debridement, curettage and antibiotic stimulant insertion. The surgical intervention included taking the patient in Supine position, incision given at the dorsal aspect over the previous surgical scar. Bone was exposed and de-roofing of the cavity was done. Debridement and curettage was done. The wound was washed thoroughly with Betadine, normal saline and hydrogen peroxide. The dorsal lesion was sealed with bone wax. The Lesion was filled with 10CC Stimulan with vancomycin after the wound was cleared. Closure done after inserting 8 no drain.

Intra- operatively, the curettage showed casseous material with black sulphur granules which was sent for biopsy, AFB staining, culture and histopathology. His biopsy revealed multiple grey-white to dark brown soft tissue pieces measuring 2*1.2*0.3cm. The tissue section examination showed tightly packed fungal hyphae with inflammatory exudate and cell debris with no evidence of Malignancy in the tissue samples. His AFB cultures showed no Acid Fast Bacillus. Biopsy specimens from the tissues confirmed the diagnoses of acute on chronic eumycetoma. The routine biochemical and hematological tests including erythrocyte sedimentation rate were within the normal range. A post-operative Xray showed no remnant cysts and abnormalities following which an antibiotic course of Ampiciline/Salbactum for a period of one month was initiated. The patient was followed up for 2 years where the patient reported complete resolution of the infection and no hindrance in any activity. Patient had uneventful stay in hospital which was for one day. Patient was discharged in a stable condition. The medical management included Voriconazole 200mg, thrice daily for 3 days followed by twice daily for 27 days.

DISCUSSION

To our knowledge, involvement of the upper extremities is very rare. Cultures of the surface of ulcers/draining sinuses are often misleading making bone cultures absolute necessary to determine the true pathogens of the infection. Umut Hatay Gölge Mah et al. reported a case of 50-year old patient with complaints of pain, swelling, purulent discharge, multiple nodules, and deformity of his right hand. Most patient often have a history of trauma to the infected site[10]and in our case too, patient reports a previous history of trauma of 7 years back to the left forearm. Patients often require prolonged (6- to 12-month) high dose penicillin G or amoxicillin treatment. The duration of antibiotics could probably be shortened to 3 months in those where optimal surgical resection of infected tissues has been performed. It is often discussed that delay in diagnosis, delay in reporting, prolonged disease, poor patient compliance in continuing the treatment makes amputation as the last resort in preventing the infection from spreading. It is therefore advisable that clinicians/surgeons should be aware of the full spectrum of this disease where presentation is usually insidious, having diverse manifestations, including nodular lesions, subcutaneous abscess, or even mass lesion mimicking tumor. An early recognition will lead to minimizationin surgical interventions and morbidity. Drug resistance is not a

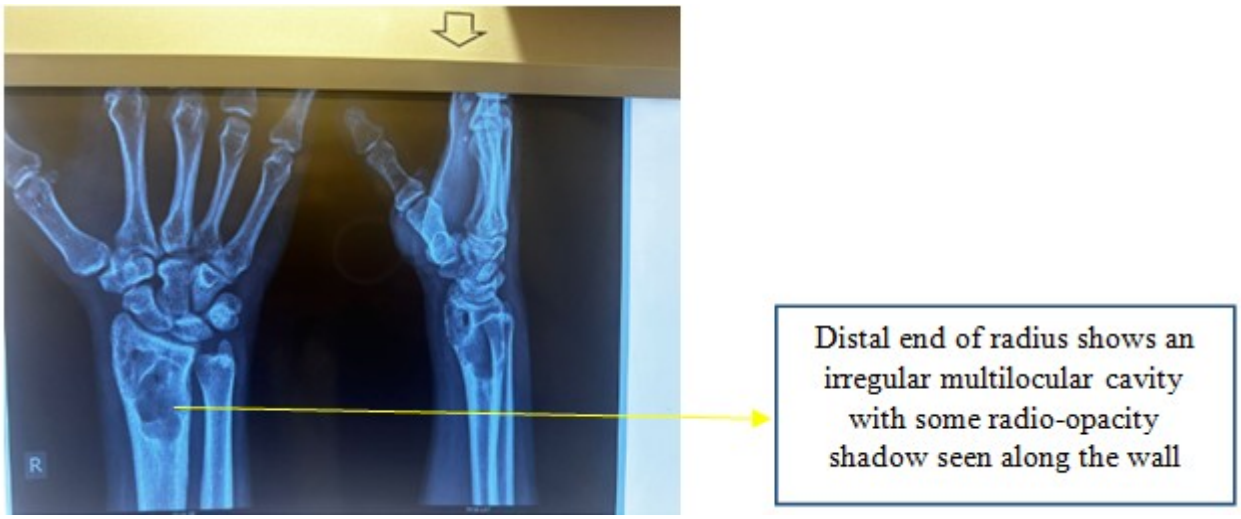

Fig. 6. Pre-op Xray of Wrist Joint (Right side)

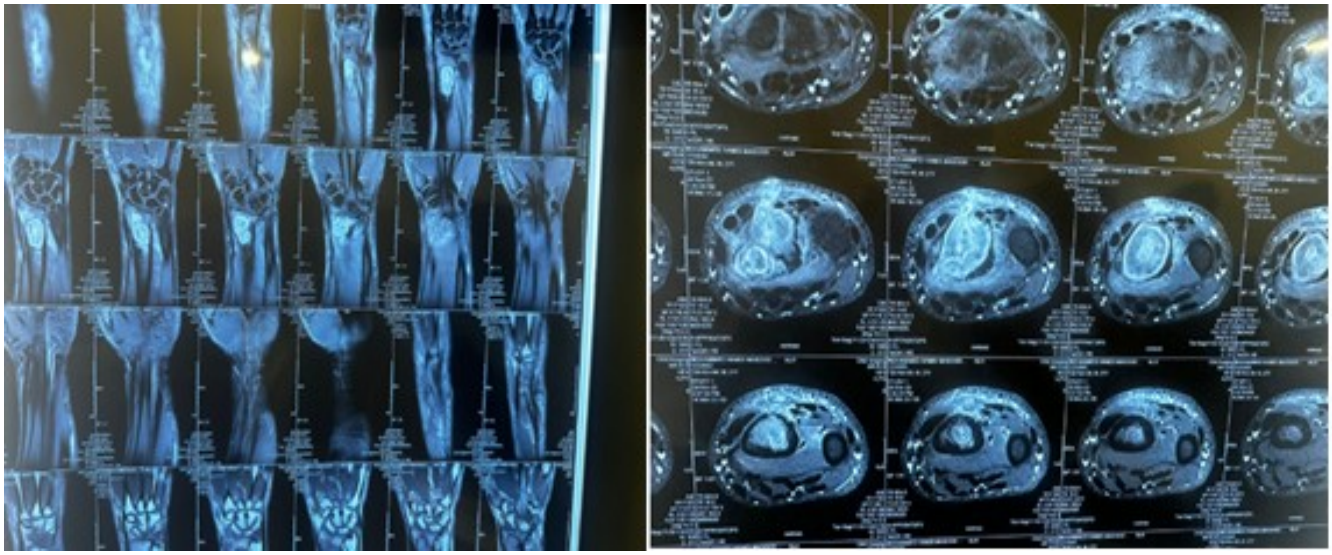

Fig 7: Pre-op MRI of Wrist Joint (Right side) showing the lesion

Fig 8. Intra-op Image of the cavity

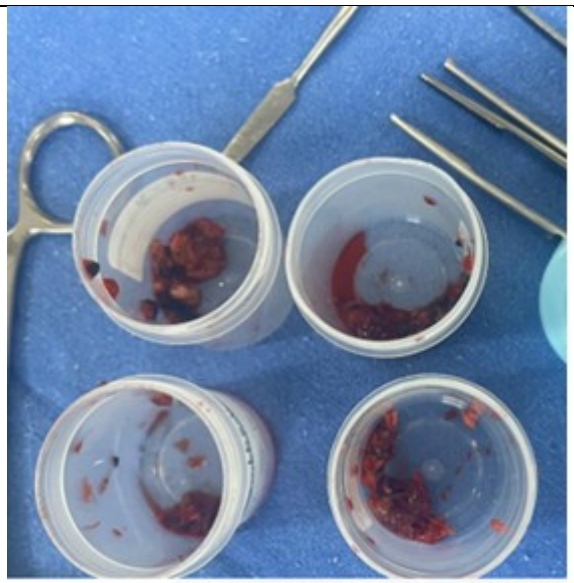

Fig 9. Granules curettage from the site

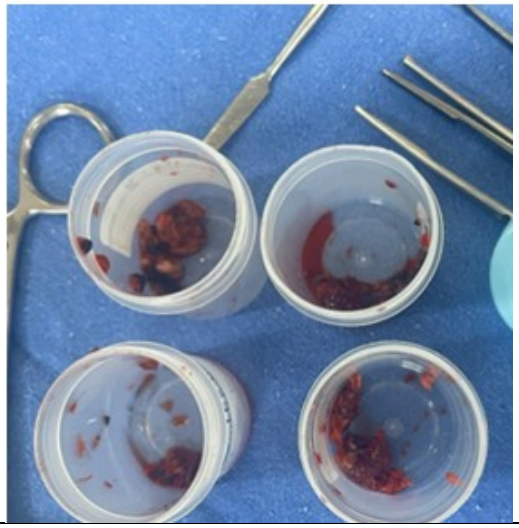

Fig 10. Post curettage image

Figure 11. Post Op XRay (AP & Lateral View)

threat in mycetoma treatment. *Actinomyces Spp*. are usually extremely susceptible to beta-lactams (especially penicillin G or amoxicillin). The main implication for physicians is to be aware of the different clinical forms of mycetomaby using specific biopsies and cultures if required.

CONCLUSION

Mycetoma is often difficult to diagnose clinically and well as radiographically. Given the rarity of the disease in the long bones, it is important to consider Fine Needle aspiration in these cases along with thorough debridement and prophylactic antibiotic beads. Post-operative use of anti-fungals are to be used for longer periods to prevent further recurrence of the disease.

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