



Actinomycotic Mycetoma on The Wrist: an Intriguing Lesion on An Uncommon Location

KEYWORDS

ACTINOMYCOSIS, ANAEROBIC BACTERIA, HAND, EXCISION BIOPSY

DR. JAI VINOD KUMAR

M.S., ASSISTANT PROFESSOR, DEPARTMENT OF GENERAL SURGERY, SRI MUTHUKUMARAN MEDICAL COLLEGE, HOSPITAL & RESEARCH INSTITUTE, CHENNAI – 87

PROF. K. SIVA PRAKASAM

M.S., PROFESSOR, DEPARTMENT OF GENERAL SURGERY, SRI MUTHUKUMARAN MEDICAL COLLEGE, HOSPITAL & RESEARCH INSTITUTE, CHENNAI – 87

ABSTRACT ACTINOMYCOSIS IS A CHRONIC GRANULOMATOUS DISEASE WITH THE CHARACTER OF EXTENSION TO THE CONTAGIOUS TISSUE FORMING MULTIPLE DISCHARGING SINUS TRACTS. PRIMARY ACTINOMYCOSIS OF EXTREMITY IS NOT A COMMON CLINICAL ENTITY AND IS USUALLY CONSIDERED AS A SOFT-TISSUE INFECTION. WE REPORT HERE A CASE OF ACTINOMYCOTIC MYCETOMA OF THE HAND FOR ITS RARITY, CLINICAL INTERVENTION THAT WAS DONE AND THE GOOD OUTCOME FOLLOWING PROLONGED ANTIMICROBIAL TREATMENT.

INTRODUCTION

ACTINOMYCOSIS IS AN INDOLENT, SLOWLY PROGRESSIVE INFECTION CAUSED BY ANAEROBIC OR MICROAEROPHILIC BACTERIA PRIMARILY OF THE GENUS ACTINOMYCES THAT COLONIZE THE MOUTH, COLON AND VAGINA. MUCOSAL DISRUPTION LEADS TO INFECTION AND SMALL ABSCESSSES AND PUS FILLED SINUS TRACTS ARE FORMED FROM WHICH PUS CONTAINING COLONIES OF ORGANISMS CALLED SULPHUR GRANULES ARE DISCHARGED [1]. ACTINOMYCOSIS HAS A PEAK INCIDENCE IN THE MIDDLE DECADE WITH THREE-FOLD HIGHER INCIDENCE IN MALES. THE COMMON SITES OF INFECTION ARE CERVICOFACIAL, THORACIC, ABDOMINAL, PRIMARY CUTANEOUS AND PELVIC [2].

PRIMARY ACTINOMYCOSIS OF THE HAND & WRIST ARE UNCOMMON AND VERY FEW CASES HAVE BEEN PUBLISHED IN LITERATURE. THIS REPORT PRESENTS THE CLINICAL, MICROBIOLOGICAL, HISTOLOGICAL FINDINGS AND THE OUTCOME OF A PATIENT WHO VISITED THE SURGICAL OUTPATIENT DEPARTMENT WITH MULTIPLE SINUSES ON HIS WRIST WITHOUT ANY HISTORY OF TRAUMA [3].

CASE REPORT

A 50-YEAR-OLD MALE PATIENT PRESENTED WITH NO PREVIOUS HISTORY OF ANY RELATED ILLNESS OR IMMUNODEFICIENCY CAME TO THE SURGICAL OPD WITH A LESION OF MULTIPLE NODULES PRESENTING AS A SWELLING ON THE RIGHT WRIST AND HAND FOR THE PAST 6 YEARS (FIGURE 1). THE LESION STARTED AS A SINGLE PAINLESS NODULE OVER THE FLEXOR ASPECT OF RIGHT WRIST AND WITHIN A PERIOD OF 6 MONTHS PROGRESSED TO FORM MULTIPLE NODULES WITH SEROPURULENT DISCHARGE EXTENDING TO THE HAND REGION. THE PATIENT GAVE NO HISTORY OF TRAUMA TO THE HAND. THE PATIENT WAS NOT DIABETIC OR HYPERTENSIVE AND HAD NO HISTORY OF TUBERCULOSIS. CLINICALLY MULTIPLE ERYTHEMATOUS NODULES WITH DISCHARGING SINUSES OVER THE FLEXOR ASPECT OF RIGHT WRIST AND HAND WITH INDURATION AND SWELLING WERE NOTED. (FIGURE 1). THE MOBILITY OF THE WRIST JOINT WAS NOT RESTRICTED. THE EXAMINATION OF THE DISCHARGE AND THE SCRAPING FROM THE SINUSES DID NOT REVEAL ANY GRANULES. HAEMATOLOGICAL, BIOCHEMISTRY AND URINE ANALYSIS REPORTS WERE ALL WITHIN NORMAL LIMITS. MANTOUX TEST TURNED OUT NEGATIVE AND CHEST X-RAY WAS GROSSLY NORMAL. X RAY OF THE RIGHT HAND SHOWED SOFT TISSUE SWELLING AROUND THE RIGHT WRIST JOINT AND NO BONY INVOLVEMENT TO SUGGEST OSTEOMYELITIS (FIGURE 2). FNAC DONE ON THE WRIST JOINT SHOWED SCATTERED HISTIOCYTES AND OCCASIONAL MULTINUCLEATED GIANT CELLS IN A SEEMINGLY NECROTIC BACKGROUND. AN EXCISION BIOPSY WAS PLANNED AND TAKEN FROM A REPRESENTATIVE NODULE UNDER

ASEPTIC PRECAUTIONS IN THE OPERATING ROOM AND SENT FOR HISTOPATHOLOGICAL EXAMINATION. THE PATHOLOGIST REPORTED THAT THERE WERE BITS OF FIBROUS CONNECTIVE TISSUE WITH A TRACT COVERED BY GRANULATION TISSUE, WHICH FOCALLY SHOWED A PALE BLUE COLONY WITH SURROUNDING SPLENDOR-HOEPLI REACTION AND NUMEROUS ACUTE ON CHRONIC INFLAMMATORY CELLS SUGGESTIVE OF MYCETOMA (FIGURE 3). GRAM STAIN WAS PERFORMED ON TISSUE SECTION AND FOUND POSITIVE. AFB STAINING TURNED OUT TO BE NEGATIVE. AS THERE WAS NO EVIDENCE OF ANY LUNG DISEASE OR GINGIVITIS IN PATIENT, A DIAGNOSIS OF PRIMARY ACTINOMYCOSIS OF HAND WAS MADE FOR WHICH HE WAS TREATED WITH ORAL PENICILLIN THERAPY FOR 6 MONTHS TO WHICH PATIENT RESPONDED VERY WELL. (FIGURE 4)

FIGURE1. INITIAL CLINICAL VIEW OF THE LESION



FIGURE2. X-RAY OF THE WRIST JOINT SHOWS NO BONY INVOLVEMENT.



FIGURE3. ACTINOMYCETOMA NEUTROPHILIC INFILTRATE SURROUNDING THE ACTINOMYCOTIC COLONY.(H&E 40x)

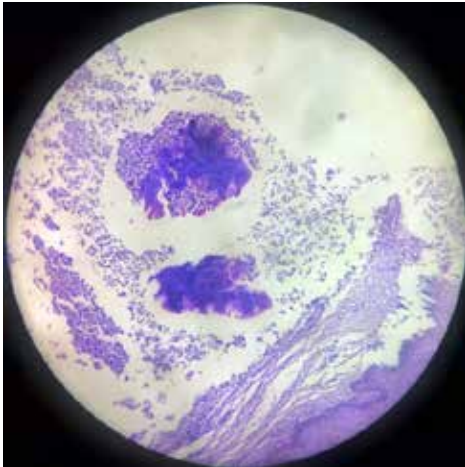


FIGURE4. CLINICAL PICTURE FOLLOWING THE COURSE OF TREATMENT



DISCUSSION

ACTINOMYCOSIS IS A RARE INFECTION CAUSED BY A GRAM-POSITIVE, NON-SPORE FORMING AND ANAEROBIC BACILLI BELONGING TO THE ACTINOMYCES SPP. WHICH IS PRIMARILY A COMMENSAL BACTERIA FOUND IN THE NORMAL ORAL CAVITIES, INCLUDING TONSILLAR CRYPTS, DENTAL PLAQUES, CARIES TEETH AND ALSO THE FEMALE GENITAL TRACT. IT HAS NOT BEEN ISOLATED FROM THE SOIL OR OTHER OBJECTS OUTSIDE THE BODY. [4] MOST OF THE SIMILAR INFECTIONS DOCUMENTED UNLIKE THIS CASE HERE, HAVE OCCURRED AFTER TRAUMATIC INJURY THAT CREATED AN ANAEROBIC CONDITION PREDISPOSING TO THIS BACTERIAL GROWTH. THE CULTURES ARE POSITIVE IN ONLY 24% OF CASES AND DIAGNOSIS IS OFTEN BASED ON HISTOPATHOLOGICAL FINDINGS. [5]

ACTINOMYCOSIS HAS BEEN CALLED "THE MOST MISDIAGNOSED DISEASE" AND IT REMAINS A DIAGNOSTIC CHALLENGE TO MOST CLINICIANS. ITS CHRONIC AND INDOLENT COURSE RESEMBLES THAT OF A FUNGAL INFECTION, TUBERCULOSIS OR MALIGNANCY, FOR WHICH THERE IS DELAY IN EARLY DIAGNOSIS. [6]

IN THE PRESENT DISCUSSED CASE, PATIENT HAD NO HISTORY OF INJURY TO HIS WRIST OR HAND. THE LESION STARTED AS A SWELLING ON THE WRIST WHICH GRADUALLY EXTENDED TO HIS HAND AND DISTAL FOREARM. HE WAS TREATED WITH DIFFERENT KINDS OF ANTIMICROBIAL AGENTS BEFORE APPROACHING US WHICH HAD LITTLE BENEFICIARY EFFECT. ALTHOUGH AN INITIAL SUSPICION WAS MADE FOR TUBERCULOSIS, IT WAS RULED OUT BY AFB STAINING AND CHEST X-RAYS. BASED ON THE CLINICAL PRESENTATION AND HISTOPATHOLOGICAL FINDINGS FROM THE BIOPSY TAKEN, THE PATIENT WAS DIAGNOSED AS A CASE OF PRIMARY ACTINOMYCOSIS AND SHOWED AN EXCELLENT RESPONSE

WITHOUT THE NEED FOR SURGICAL DEBRIDEMENT AND WITH JUST A LONG-TERM PENICILLIN THERAPY.

CONCLUSION

WE WANT TO HIGHLIGHT HERE THAT THE TREATING CLINICIAN SHOULD POSSESS KNOWLEDGE OF THIS DISEASE WHOSE PRESENTATION IS USUALLY INDOLENT AND CAN HAVE VARIED MANIFESTATIONS, INCLUDING NODULAR LESIONS, SUBCUTANEOUS ABSCESS, OR EVEN MASS LESION MIMICKING A TUMOR. ACTINOMYCETOMA SHOULD BE DISTINGUISHED FROM EUMYCETOMA AS THE TREATMENT VARIES. ACTINOMYCOSIS CAN BE CURED WITH MINIMAL SURGICAL DEBRIDEMENT AND APPROPRIATE ANTIBIOTIC THERAPY WHILE EUMYCETOMA IS ONLY PARTIALLY RESPONSIVE TO ANTIFUNGAL AGENTS [7], HAS HIGH RATE OF RECURRENCE AND MAY EVEN REQUIRE AMPUTATION. A HIGH DEGREE OF SUSPICION AND EARLY HISTOPATHOLOGICAL SAMPLING USING SPECIAL STAINING TECHNIQUES CAN HELP IN EARLY DIAGNOSIS AND TREATMENT WHICH WILL MINIMIZE UNNECESSARY SURGICAL INTERVENTIONS, MORBIDITY AND MORTALITY.

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