ORIGINAL RESEARCH PAPER

Medical Microbiology

AN UNUSUAL CASE OF NON-HEALING MULTIPLE ULCERS OF FOOT, CUTANEOUS BOTRYOMYCOSIS!

KEY WORDS: botryomycosis, ulcers, discharging sinus, immunosupression

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ABSTRACT

Botryomycosis is a chronic granulomatous bacterial disease, frequently involves skin and rarely internal viscera . It is an uncommon condition, with fewer cases reported worldwide, it is mostly reported in patients with dysfunctional immune system, but it can also occur in an otherwise healthy person. Gram positive agent *Staphylococcus aureus* is the major etiological agent however Gram-negative organisms like *Pseudomonas spp., Escherichia coli, Proteus spp.,* etc have also been reported to cause botryomycosis though less commonly.

A case report of cutaneous botryomycosis of foot in a 33-year-old male who presented with nonhealing ulcers and swelling on ventral aspect of right foot and ankle for a year. Microbiological and histopathological investigations helped in establishing the diagnosis. Culture from lesions revealed growth of *Methicillin sensitive S. aureus.* Slow resolution was achieved with surgical debridement and combination therapy with Piperacillin-Tazobactam and Gentamycin.

INTRODUCTION:

Botryomycosis or bacterial pseudomysosis/pyoderma vegetans is a chronic inflammatory response to a bacterial infection involving the skin and sometimes internal organs. It manifests clinically as tumours or plaques that are often ulcerated and have discharging sinuses draining small whitish granules; hence, they closely mimic mycetoma or other fungal infection [1]. The name is misleading; botryomycosis is a bacterial infection rather than a fungal infection. "Botryo" is a latin term for "bunch of grapes". Other authors have suggested that actinomycosis and botryomycosis should be classified under the single term 'granular bacteriosis'. The lesions are commonly present in the head/neck region and in the extremities [2]. The causative agents are usually pyogenic, gram-positive bacteria such as *S. aureus*. The gram negative organisms such as *P.aeruginosa, Proteus spp., E.coli ,Actinobacillus lignieresii* are lesser known causative agents[1]. Botryomycosis develops in the backdrop of immune suppressive states such as diabetes mellitus, HIV/AIDS, cystic fibrosis, and other immune deficiency conditions. Nevertheless, botryomycosis has been reported in an otherwise healthy individual. Hence there are other factors, besides improperly functioning immune system, behind the development of these lesions with a tomour/chronic fungal infection–like morphology [3,2].

Case Report:

A 33-years old-young male presented with more than 1 year history of swelling and multiple erythematous skin lesions on ventral aspect of right foot and ankle region, some of the lesions were discharging scanty foul-smelling purulent material while others were having dried crust on them. Patient gave a history of minor trauma without visible breach of skin integrity at affected site around a year back, however patient was not sure about history of trauma. The lesion started as a painless single papule, gradually developed into a nodular swelling, increased in size, and formed a discharging sinus. More nodules and discharging sinuses developed subsequently. (fig.1) patient has no significant co-morbidity, but he was an active smoker and alcoholic. Patient had taken multiple allopathic and homeopathic treatments (patients was not able to provide previous history of antibiotic usage). On examination, patient was afebrile affected site was swollen, hard and woody, with numerous elevated, well-demarcated, small and large nodular lesions and some discharging sinuses were present. Most of the sinus openings were inflamed and blocked with dried pus (some with oozing lesions). Tenderness was present however patient denied having pain. Discharge was yellowish, purulent, and foul smelling. sample was collected from lesion as per SOP. The differential diagnoses (DD) thought were actinomycosis, nocardiosis, eumycetoma, mycobacterial tuberculosis, non-tubercular mycobacteria, and botryomycosis. Hence the microbiological work up was carried out keeping all possible DD in consideration. A portion of sample was transferred into the tube and washed with normal saline; no granules were found. Four slides were prepared from the direct sample, one for gram staining, one of KOH wet mount, and other two for 20% and 1% acid fast. Sample was subsequently sub-cultured onto two sets of blood agar plate(cos-Biomerieux) and chromogenic agar (cps-Biomerieux), one set was incubated aerobically while another set was incubated anaerobically at 35□c±2o. A portion of sample was also cultured on Lowenstein Jensen medium (LJ) and Sabouraud Dextrose Agar (SDA) slant. Gram smear showed plenty of pus cells and gram-positive cocci in clusters and pairs. Smears were negative 1% and 20% acid fast staining. No fungal element observed in KOH mount.

Fig1: Showing Gross Lesion With Woody Texture, Multiple Sinus Tracts And Tumefaction Of Right Foot And Ankle.

Abundant growth was observed on both sets blood agar plate & chromogenic plate after 24 hrs of incubation, predominant colonies on blood agar plate were haemolytic and golden yellow. A small number of colonies on aerobic plate were mucoid. Gram smear of haemolytic colonies were showing gram positive cocci in clusters while mucoid colonies on gram staining showed gram positive bacilli (aerobic spore bearers). identification and antimicrobial sensitivity of

predominant haemolytic colonies was done by viket 2 compact. Organism was identified as *S.aureus* (MSSA), sensitive to gentamycin, amikacin, ceftriaxone and piperacillin-tazobactam but resistant to ciprofloxacin, cefixime and ampicillin.

Biopsy was done and specimen was submitted to microbiology and pathology department. The microbiological result of biopsy specimen was found consistent with pus discharge sample processed previously and further supported by histopathological findings. (fig.2 ,3) Complete blood picture, and inflammatory marker CRP, ESR were advised[table1]. Radiological study of right ankle and foot showed no bone involvement.

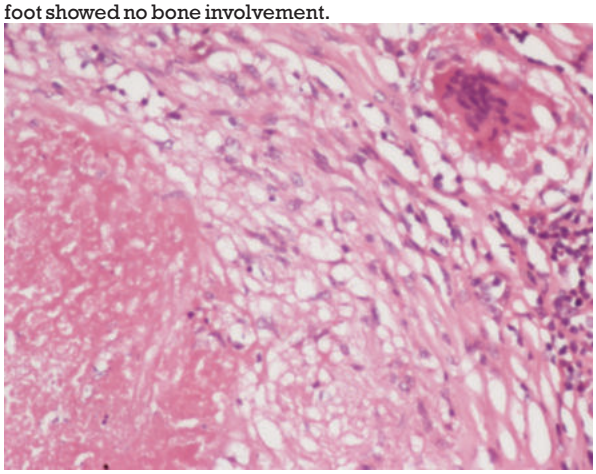

Fig 2: Multinucleated giant cells, granules surrounded by inflammatory response

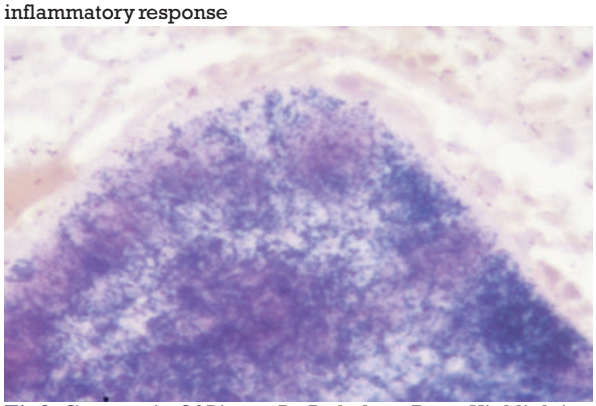

Fig3: Gram-stain Of Biopsy By Pathology Dept. Highlighting Gram Positive Cocci

Table 1: Laboratory parameters of the patient with cutaneous botryomycosis

Investigation	result
Haemoglobin (g/dl)	13.0
Total leukocyte count (/cu mm)	11,500
Platelet count (/cu mm)	150,000
HIV, HBV, HCV	Negative
Serum Creatinine (mg %)	1.5
ESR (mm/h)	55
CRP (mg/dl)	10
HbA1C (g/dL)	5

ESR: Erythrocyte sedimentation rate, CRP: C-Reactive Protein, HbA1C: Glycosylated haemoglobin, HCV: Hepatitis C virus, HBV: Hepatitis B virus, HIV: Human Immunodeficiency Virus

The patient responded very well to surgical debridement along with piperacillin-tazobactam and gentamicin. Significant reduction in swelling and discharge was observed in a month and patient was regularly followed up.

DISCUSSION:

Botryomycosis can be classified into cutaneous and visceral types, former being most common and generally involve head/neck and extremities while the visceral botryomycosis is seldom reported and mainly affecting the lungs.[3] cutaneous variant often presents as non-healing ulcers, sinuses, fistula as was seen in this patient. cutaneous infection can cause deeper tissue (muscles and bones) destruction and deformity. the visceral form is often misdiagnosed as as an abscess or a tumour. Weakened immune function is commonly seen in patients however cases of botryomycosis have been reported in healthy individuals also [3,4]. The clinical picture of cutaneous botryomycosis closely resembles other diseases causing fistula, such as scrofuloderma, actinomycosis, actinomycetoma, eumycetoma and atypical mycobacterial infections [5]. The gold standard diagnostic tool for botryomycosis is histopathology and culture. On eosin-haematoxylin preparation, botryomycosis is characterised by a central focus of necrosis surrounded by a chronic inflammatory reaction containing histiocytes, epithelioid cells, multinucleated giant cells and fibrosis. There may be granules called 'Bollinger granules' which consist of the bacteria surrounded by an eosinophilic matrix with club-like projections. This appearance is called the Splendore-Hoeppli phenomenon [6].

CONCLUSION:

Botryomycosis, though a rare condition but its possibility in a patient with long standing history of non-healing ulcers/sinuses/fistula should be thoroughly evaluated. Appropriate antibiotics therapy should be started after identifying the causative pathogen.

Patient Consent

The patient has given his consent for his images and other clinical information to be reported. Patient has been assured that his name will not be published, and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

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