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Case Report

Post Marsupialisation Vulval Botryomycosis in Northeast Nigeria: A Rare Case Report

Ishiyaku UM[1, 2], Ibrahim M[1, 2], Hassan UM[1,2], Farouk HU[3], Lawan AI [4], Manga MM[1, 2]

1.Department of Medical Microbiology and Immunology, College of Medical Sciences Gombe State University, Gombe. 2. Department of Medical Microbiology and Immunology, Federal Teaching Hospital Gombe. 3. Department Obstetrics and Gynecology Gombe State University, Gombe/ Federal Teaching Hospital Gombe 4. Department of Histopathology College of Medical Sciences Gombe State University, Gombe/ Federal Teaching Hospital Gombe

Correspondence: U.M Ishiyaku, Department of Medical Microbiology and Immunology, College of Medical Sciences Gombe State University, Gombe. Email: uimaigado@gmail.com

ABSTRACT

Botryomycosis is an uncommon chronic suppurative granulomatous infection that affects humans, usually involves the skin, and rarely viscera; it mimics other conditions like actinomycosis, mycetoma, and subcutaneous mycoses. We here report a unique case of a 42-year-old woman who is a known Human Immunodeficiency Virus (HIV) patient who had failed first-line anti-retroviral therapy, she presented with a 3-month history of post-marsupialization left labia multinodular swelling with discharging sinuses. Tissue biopsies for Microbiological and histologic diagnosis yielded *Pseudomonas aeruginosa, Acinetobacter baumannii,* and micro abscesses respectively. She was treated with Intravenous Amikacin for 10 days and later switched to oral levofloxacin for another 10 days based on the susceptibility result. This is the first reported case of Botryomycosis in Nigeria and the first case of Pelvic botryomycosis in the world.

KEYWORDS**:** Botryomycosis, Marsupialization, Nigeria

INTRODUCTION

Botryomycosis (bacterial pseudomycosis or pyoderma vegetans) is a chronic suppurative granulomatous infection that occurs due to the reaction of the affected tissue or organ to the presence of an organism. A lot of organisms are implicated, however, *Staphylococcus aureus, Pseudomonas aeruginosa,* Escherichia *coli,* and *coagulase-negative Staphylococci* are the most prevalent agents responsible[1]. Botryomycosis is classified into two broad types based on the area of affectation: visceral and cutaneous botryomycosis[2]

The first case was identified in Horses following a surgical procedure in 1870 while the first human case was reported in 1913, and *Staphylococcus aureus* was the first etiologic agent implicated in 1919 [3]

Clinical presentations of Botryomycosis are similar to that of Actinomycosis and Mycetoma, typical Presentation begins initially as a papule which progressively increases in size and number with wider tissue involvement to Nodules subsequently ruptures leading to sinuses draining purulent effluent of varied colours and some lesions ulcerate[4,5]. Factors associated with the risk of developing Botryomycosis include but are not limited to diabetes mellitus, Human Immunodeficiency viral infections (HIV), trauma, surgery, alcoholism, and cystic fibrosis.[2,6]

Management of botryomycosis requires a multidisciplinary approach involving Surgeons preferably plastic surgeons, Clinical Microbiologists, Dermatologists, and Histopathologists. Prolonged anti-biotherapy with susceptible antibiotics in conjunction with surgical excision, debridement, and regular dressings is key in the management[4]

CASE PRESENTATION

She is a 42 years old known HIV patient on Highly Active Antiretroviral Therapy (HAART) who was referred to Federal Teaching Hospital Gombe with a Three (3) months history of left groin swelling that initially began as a single peanut size swelling, which later progressively increased in size and number and subsequently discharging brownish effluent. The swelling was painless initially; however, it became severely painful at the time the discharge started. There was associated foul-smelling white to brown vaginal discharge, body weakness, inability to walk well and low-grade fever. No other swelling in other parts of her body.

She had a procedure (marsupialization) done on account of a Bartholin cyst in a peripheral hospital in Akwa-Ibom state Nigeria at the same site about 4 weeks before this index presentation. She has been a known HIV patient for 13 years and has failed first-line anti-retroviral therapy and is currently on second-line Tenofovir/Lamivudine plus Atazanavir/ritonavir. She is not a known diabetic, hypertensive, or sickle cell disease patient and has no other known chronic disease or malignancies, she neither smokes nor takes alcohol. She had several antibiotics before her referral, however, she could only remember Doxycycline and Erythromycin

On initial examination, we noted a chronically ill-looking middle-aged woman that was pale, acyanosed, febrile with a temperature of 38.8^{o}C, and bilateral pitting pedal eodema. Pulse was 108 beats per minute, Blood pressure: systolic 90 and diastolic 60 millimeters of mercury with and second heart sounds heard—respiratory rate 22 circles per minute. Pelvic examination showed huge multi-nodular tendered swelling measuring about 10cm× 8cm on the left labia, numerous discharging sinuses like-areas with foul smelling brownish to white vaginal discharge.

She was referred to Federal Teaching Hospital Gombe northeast Nigeria with a working diagnosis of Lymphogranuloma venereum (LGV), however, after clinical evaluation and examination in our hospital, a diagnosis of pelvic actinomycosis to rule out mycetoma was made initially. Complete blood count, CD4 cell count, HIV viral load, blood culture, high vaginal swab, urine Microscopy culture and sensitivity, and two Tissue biopsies for Culture and histology were taken and processed according to standard microbiological and histopathological techniques.

The results of the investigations were complete blood count: Packed cell volume (PCV) 26% white blood cell 12.1× 10^{9}/L (neutrophilia 90% lymphocytes 8% and monocytes 2%), Platelets 802 ×10^{9}/L CD4 521 cells/ul, viral load 770 copies per ml. Blood culture, high vaginal swab, urine microscopy culture and sensitivity, and Ziehl-Neelsen stains were all unremarkable. Multiple tissue biopsies for culture and sensitivity showed: Numerous white blood cells, mixed Gram-negative bacilli and coccobacilli, culture yielded two organisms: *Pseudomonas aeruginosa* susceptible to amikacin, cefepime, ciprofloxacin, levofloxacin, gentamycin and meropenem and resistant to ceftazidime, while *Acinetobacter baumannii* was only susceptible to amikacin and meropenem and resistant to cefepime,

Figure 1: Multiple nodular lesions with discharging sinuses on the vulva covering mainly the labia

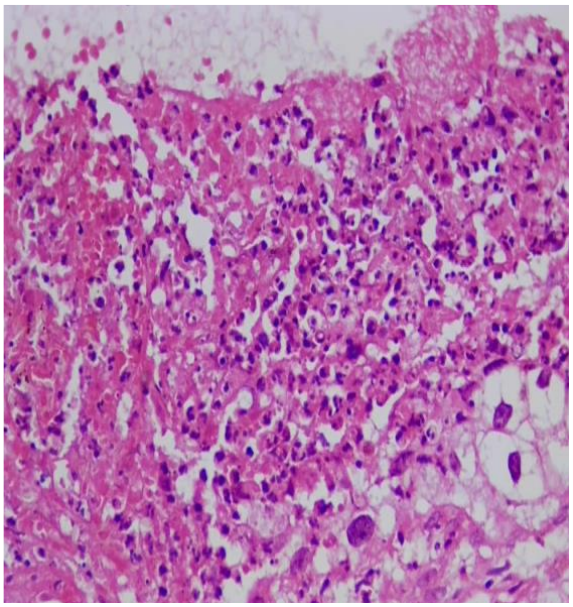

Figure 1: Multiple nodular lesions with discharging sinuses on the vulva covering mainly the labia

ceftazidime, gentamycin and tetracycline. Histology showed foci of micro abscesses with bacteria colonies.

She was managed by a multidisciplinary team consisting of a gynecologist, clinical microbiologist, general surgeon, plastic surgeon, and histopathologist. She had daily dressings, and sitz baths with warm saline two times daily. Targeted antibiotherapy was imitated with intravenous amikacin 250mg 12 hourly for 10 days after which an Intravenous to oral switch to levofloxacin 750mg daily for additional 10 days. A significant improvement was noted and she was discharged. Altogether she spent 5 weeks in the ward.

DISCUSSION

Botryomycosis is an uncommon suppurative granulomatous infection that mimics other diseases like Subcutaneous mycosis generally and Actinomycosis, However, it is caused by an array of bacteria agents[1]. Cutaneous botryomycosis is more prevalent than the visceral type in a ratio of 3:1[2] .

Pathogenesis of botryomycosis is still equivocal; But several risk factors have been postulated to be associated, some of which include: Immunosuppressive conditions and disorders, Diabetes mellitus, HIV, Trauma, Surgery, and lots more[3]. Our reported case is immunocompromised with HIV infection and had marsupialization done at her left vulva four weeks before her presentation with these symptoms. These two factors were instrumental in making the diagnosis of botryomycosis.

Our patient presented with a month's history of left vulval painless swelling, which progressively increased in size and number, this later ruptured and became painful, discharging brownish effluent. This is not in conformity with that of the case reported by Elas et al, in 2012, here an 85-year-old postmenopausal woman presented with a vulva ulcer with associated bleeding and itching, this was initially thought to be a vulva neoplasm. [4]

Diagnosis of botryomycosis is very challenging as the clinical presentation together with the histopathologic diagnosis will relatively not differentiate botryomycosis from actinomycosis, mycetoma, and other subcutaneous granulomatous infections[3] therefore, a combination of symptoms, signs, microbiological and histopathological investigations are crucial in making the diagnosis[5]. We collected multiple tissue biopsies for Microscopy culture and sensitivity and Histology respectively, *Pseudomonas aeruginosa* and *Acinetobacter baumannii* were isolated from the culture, This is partly similar to that reported by Eyer-Silva WA, *et al*. but differ in that, the second organism isolated was *Staphylococcus aureus* rather than *Pseudomonas aeruginosa* [5]. While on Hematoxylin and eosin stain (H and E) foci of microabscesses with bacteria colonies were noted, this is similar to that reported by G. H. Findlay *et al.*, among three patients of African origin[6].

Treatment of botryomycosis depends on the type of botryomycosis, the location of the lesion, the surface area affected, the immune status of the individual, and the susceptibility result of the organism isolated. Surgical excision of the lesion, aggressive debridement, and dressing of the wound with prolonged administration of single susceptible antibiotics is recommended[3]. Though she had different antibiotics administered empirically before confirmation of the diagnosis, eventually they were changed to intravenous amikacin since both organisms isolated were susceptible to it. However, on discharge, an intravenous to-oral switch (levofloxacin) was done.

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