EUMYCOTIC MYCETOMA IN A YOUNG GIRL FROM SOKOTO, NIGERIA: A RARE AND UNUSUAL PRESENTATION.

[1]Ibitoye Paul Kehinde, [1]Jiya Fatima Bello, [2,3]Mohammed Yahaya, [4]Mohammed Umar, [3]Sabitu Muhammad Zainu, [5]Adayi Susan Opkodo, [5]Yusuf Abduganiy, [3]Jimoh Ahmed Kolawole, [5] Inoh Ikemesit Imeh, [6]Oladele Rita.

[1]Department of Paediatrics, Faculty of Clinical Sciences, College of Health Sciences, Usmanu Danfodiyo University Sokoto, Nigeria.
[2]Department of Medical Microbiology and Parasitology, Faculty of Basic Clinical Sciences, College of Health Sciences, Usmanu Danfodiyo University, Sokoto, Nigeria.
[3]Department of Microbiology, Usmanu Danfodiyo University Teaching Hospital, Sokoto, Nigeria.
[4]Department of Morbid Anatomy and Forensic Medicine, Faculty of Basic Clinical Sciences, College of Health Sciences, Usmanu Danfodiyo University, Sokoto, Nigeria.
[5]Department of Paediatrics, Usmanu Danfodiyo University Teaching Hospital, Sokoto, Nigeria.
[6]Department of Medical Microbiology and Parasitology, Lagos University Teaching Hospital.

ABSTRACT

INTRODUCTION: A typical presentation of Mycetoma is not uncommon although clinical manifestations might be misleading leading to delay in diagnosis, treatment and consequently leading to poor prognosis. Mycetoma can have a fungal or bacterial etiology and manifestation is usually that of a disfiguring subcutaneous infection that can affect any part of the exposed body. We are reporting a case of Mycetoma in an eleven years old girl that occurred in parts of the lower abdomen, perineum and gluteal region that was initially thought to be a soft tissue sarcoma or disseminated tuberculosis.

CASE PRESENTATION: An eleven years old girl presented to Usmanu Danfodiyo University Teaching Hospital, Sokoto with lower abdominal mass and multiple nodular masses with discharging sinuses on the upper part of the right thigh, perineum and gluteal region of six months duration. Swellings started as multiple small boils that subsequently started discharging from various points. Patient usually fetches firewood in the forest for her parents as her routine house chores and she remembered an incident where she had pricks from thorns in the bush around her lower thigh and perineum.
On examination she was chronically ill looking, in painful distress, with bilateral inguinal lymphadenopathy. She had nodular lesions of varying sizes ranging from 1x1cm to 4x4cm, tender, involving the upper part of the thigh bilaterally, but more on the right, lower abdomen, labia and gluteal region. Some of the lesions had hyper-pigmented sinuses discharging mucopurulent fluid, with areas of soft tissue swelling around the lower abdomen and upper right thigh extending to the leg. Patient was observed to walk with a limp gait.

MANAGEMENT AND OUTCOME: An initial diagnosis of soft tissue infection to rule out soft tissue sarcoma and disseminated tuberculosis (abdomen and lymph nodes) and deep tissue mycosis was made. However, with further investigations and reviews by the medical microbiologist and anatomic pathologist, along with bacteriologic, and mycologic studies of swab samples and aspirate and tissue biopsy for Histology revealed an eumycotic mycetoma. She received Ketoconazole and Trimetoprim-sulphametoxazole. She responded significantly as lesions reduced in sizes, abdominal swelling and leg swelling reduced with closure of discharging sinuses. Patient could walk with some resolution of the limp. Repeat abdominal ultrasound scan showed resolution of initial findings. She spent four weeks in the hospital and was discharged. On subsequent follow-up; she was walking without any limp and lesions were healed with some scar and few areas left to dry up. Further follow-up visits after one month and three month showed progressive healing and complete resolution of lesion respectively. However. Patient was however lost to further follow-up which would have enabled monitoring as to any re-occurrence or not.

CONCLUSION: We presented a case of a young girl with an abnormal presentation of eumycotic mycetoma. Patient achieved near cure on medications without the need of surgery due to an excellent multidisciplinary approach between pediatricians, clinicians, clinical microbiologists and anatomic pathologists.

KEYWORDS: Mycetoma, Eumycetoma, Paediatrics

NigerJmed2019: 337 -340

INTRODUCTION

Mycetoma is a chronic infection of the subcutaneous tissues that is caused by either a bacterial (actinomycotic) or fungal (eumycotic) pathogens. It presents with indurations and granules from discharging sinuses.[1] Mycetoma is restricted to the mycetoma belt that lie between 30°N and 15°S of the equator and further characterized by dry, hot climate with a short but heavy rainfall. Even though imported mycetoma can occur even outside the typical mycetoma belt in any parts of the world.[2]

Mycetoma has been recently classified among the neglected tropical diseases by the World Health

Correspondence to: Mohammed Yahaya
[2]Department of Medical Microbiology and Parasitology, Faculty of Basic Clinical Sciences, College of Health Sciences, Usmanu Danfodiyo University, Sokoto, Nigeria
E-mail: yahyakt@yahoo.com
Tel: +234 803 6867 478

Organization with an aim of improving reportage, management and control measures.[3] Mycetoma has no vector or animal reservoir, infection is usually transmitted to susceptible host via traumatic inoculation of any of the agents into the subcutaneous tissues.[4] There are several genera of agents implicated in either the actinomycotic or the eumycotic form of mycetoma. The most common agents for the actinomycotic mycetoma are *Nocardia asteroids, Nocardia brasiliensis, Streptomyces somaliensis,* and *Actinomadura madurae* among others. However, the most common eumycotic mycetoma are *Madurella mycetomatis, Madurella grisea* and *Pseudoallescheria boydii*.[5]

Case Presentation

An eleven years old girl presented to Usmanu Danfodiyo University Teaching Hospital, Sokoto in February 2017 with complaints of lower abdominal mass and multiple nodular masses with discharging sinuses on the upper part of the right thigh, perineum and gluteal region of six months duration. The swellings started as small multiple boils initially, pruritic, more on the upper part of the right thigh but progressed over months to become nodular masses involving the thigh, perineum, gluteal region and lower abdomen and subsequently started discharging from various points. There was associated unilateral right lower limb swelling with limping from pain. There is history of weight loss but no history of street hawking or sexual contact. Patient usually fetches firewood in the forest for her parents as a routine house chore and she remembered an incident (almost three months prior to presentation) where she had severe pricks from thorns in the bush around her lower thigh and perineum while fetching firewood. Her parents are rural farmers located in a remote village of Illela local government area of Sokoto State bordering Nigeria and Niger Republic.

On examination she was ill looking, in painful distress, not pale, afebrile (36.8^0C - axillary), anicteric, acyanosed, not dehydrated, with bilateral inguinal lymphadenopathy, smallest one measuring 1x1cm to 2cmx2cm, firm, non-tender with associated unilateral right lower limb swelling. Her weight was 25kg (70% of expected body weight). She had nodular lesions of varying sizes ranging from 1x1cm to 4x4cm, tender, involving the upper part of the thigh bilaterally, but more on the right, lower abdomen, labia and gluteal region. Some of the lesions had hyper-pigmented sinuses discharging mucopurulent fluid, with areas of soft tissue swelling around the lower abdomen and upper right thigh extending to the leg. She had a mass in the supra pubic region extending below the umbilicus measuring about 8x8cm. The upper surface can be reached but the lower border could not be reached, firm, smooth, non-mobile and slightly tender. Abdominal organs were not palpably enlarged.

Her pulse rate was 92beats per minute and respiratory rate was 28 cycles per minute, blood pressure was 100/60mmHg. She had vesicular breath sounds and normal heart sounds. Abdominal organs were not enlarged, hymen was intact and neurologic examination was normal.

Management and Outcome

Swab samples of the lesion and discharges showed Gram –negative bacilli, while culture yielded *Escherichia coli* sensitive to ceftriaxone, nitrofurantoin and Trimetoprim-sulphamethoxazole (TMP/SMX). Abdominal ultrasound scan showed features in keeping with bladder mass and bilateral hydronephrosis and mesenteric lymph nodes. Retroviral screening was not reactive, Mantoux and genexpert were negative. Her packed cell volume was 32%, total white cell count 17.2 $X10^3$/uL (reference range 4-11 x 10^3/uL), with 57.7% neutrophils and 29.4%, Platelets were 559 $x10^3$/uL. The erythrocyte sedimentation rate was 120mm fall/hour. Diagnosis of soft tissue infection was made to rule out soft tissue sarcoma, disseminated tuberculosis (abdomen and lymph nodes) and deep tissue mycosis.

Further review by the medical microbiologist and pathologist, along with bacteriologic and mycologic studies of swab samples and aspirate and tissue biopsy for histology were done. A diagnosis of eumycotic mycetoma was made. Histology further confirmed mycetoma. She received intravenous antibiotics (ceftriaxone), had wound dressing daily, she had minimal improvement and lesions did not reduce in size. Following the final diagnosis of mycetoma, patient received ketoconazole and initial Intravenous antibiotics were changed to oral ciprofloxacin. She responded significantly as lesions reduced in sizes, abdominal swelling and leg swelling reduced, patient could walk with less limp and repeat abdominal ultrasound scan showed resolution of

initial findings. She spent four weeks in the hospital and was discharged.

On subsequent follow-up visit one weeks later, she was doing well on ketoconazole, oral TMP/SMX was added to her treatment. She was walking without any limp and lesions were healed with some scar and few areas left to dry up. Further follow-up visits after one month and three month showed patient has done very well and all lesions were healed. Unfortunately, patient was lost to follow up after three months and efforts to trace her further assessment were unsuccessful.

Figure 1: Patients condition at presentation with nodular lesions (circle blue) and discharging sinuses (red circle)

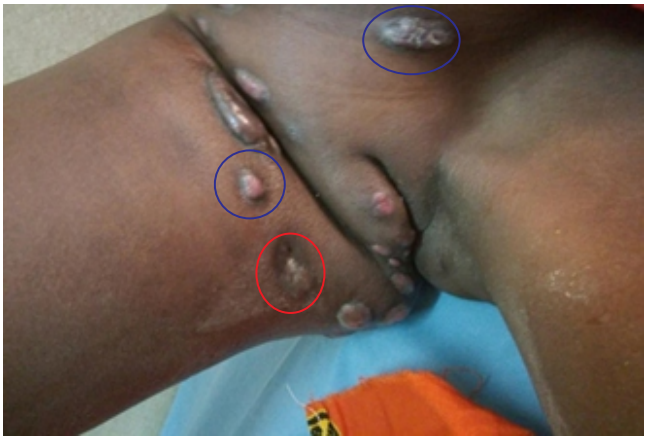


Figure 2: Unilateral leg swelling and discharging sinuses at presentation

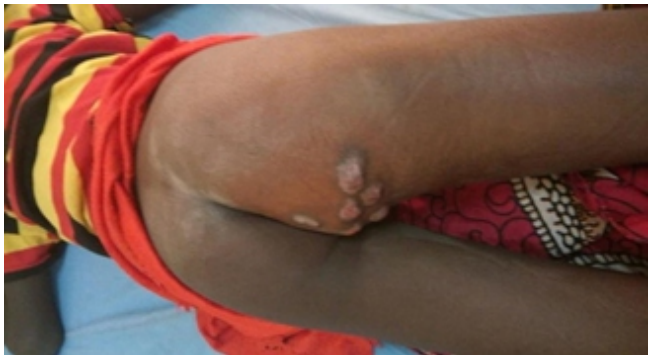
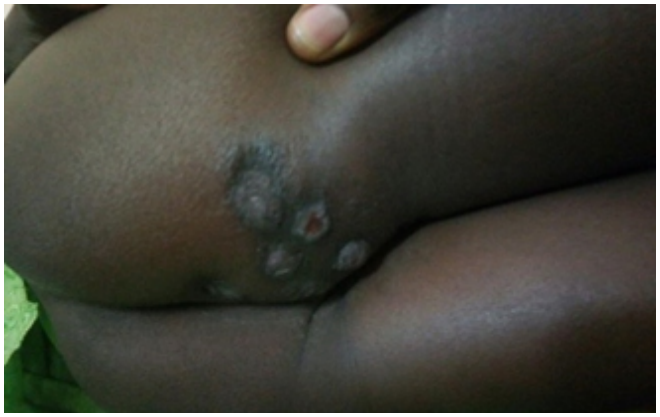


Figure 3: One week after treatment, lesions started resolving and swelling subsided



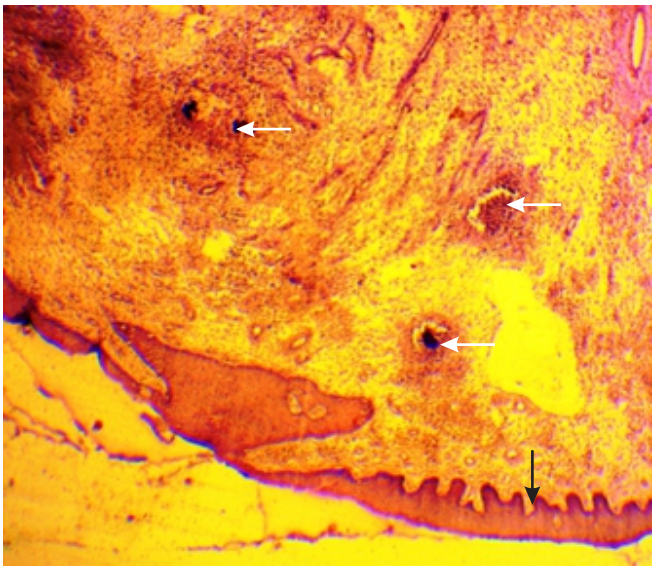


Figure 4: Section show acanthotic skin (black arrow), with lympho-plasma cell including eosinophils infiltrate. Amorphous to filamentous eosinophilic to basophilic materials that is consistent with colony of bacteria/fungi are noted (white arrow). H and E X 100

DISCUSSION

Despite the chronic disfiguring ailments with the attendant social stigma caused by mycetoma it was not until recently that the disease was included in the list of neglected tropical diseases. Even at that, the WHO is yet to issue guideline for treatment, reporting or prevention of this condition.[3]

Epidemiologic studies have elucidated that mycetoma is commonly a disease affecting young adults, males from an agrarian background resident of endemic areas.[3] Our index patient is a young female from a family of farmers. Her sex and age offer her no social protection from being exposed to the agents due to mostly demands of her family. Even though, the lower incidence of mycetoma among female was initially attributed to the possible protective effect of progesterone[3] but this was debunked later since mycetoma was found to be more aggressive during pregnancy.[4] Greater emphasis is now placed on higher physical activities rather than hormonal factors.[3]

Our patient could remember a day that she had a thorn prick around her abdomen and perineum while fetching firewood at least three months prior to presentation. This is in line with the known transmission pathway for mycetoma by available literature where the infective agent from soil or animal dung gain access to the subcutaneous tissue following traumatic inoculation after piercing the skin by thorn usually.[6]

One of the bad prognostic factors that worsens

patient's outcome is delay in seeking treatment due to the slow painless progression of the lesion caused by mycetoma. This was not the case for the index patient as she reported early enough (six months) after infection.[3]This is a good timing when compared with a case of a patient from Senegal who presented 15 years after primary infection with complicated mycetoma.[7]

Normally, mycetoma presents on body parts exposed to the outside environment. Commonest sites are the foot followed by hand while the head, neck, abdominal walls, perineum are hardly affected.[8, 9] Our patient presented with an atypical presentation of mycetoma in the lower abdomen, perineum and gluteal region outside the normal sites and could have easily been misdiagnosed if not for the excellent multi-disciplinary practice and collaboration amongst physicians in our institution.

Eumycotic mycetoma was formerly treated by amputation or multiple excisions alone. However, medical management with antifungal agents is now strongly advocated especially for cases that present early. [10, 11] Our index patient was managed successfully with ketoconazole without the need of surgery despite the case being an eumycotic mycetoma probably because of the patient's early presentation. Zein *et al* attributed patients' drop out of follow-up visits to either treatment failure or complete resolution of lesion.[12] Unfortunately, our patient was lost to follow up after six months and efforts to trace her were unsuccessful. Likely reason for her being lost to follow up is due to good clinical outcome as her previous visits showed that patient has done very well, and all lesions were healed.

CONCLUSION

We bring to fore an abnormal presentation of eumycotic mycetoma. Patient presented early and had diagnosis made on time with commencement of appropriate medication due to the excellent multidisciplinary collaborations among clinicians and pathologists. Patient achieved near cure on medications without the need of surgery.

REFERENCES

1. Mohamed HT, Fahal A, van de Sande WJ. Mycetoma: epidemiology, treatment challenges, and progress. *Research and Reports in Tropical Medicine* 2015; 6: 31–36.
2. Efared B, Tahiri L, Boubacar MS, Atsam-Ebang G, Hammas N, Hinde EF, *et al*. Mycetoma in a non-endemic area: a diagnostic challenge. *BMC Clin Pathol 2017*;17:1. https ://doi.org/10.1186/s1290 7-017-0040-5.
3. Emmanuel P, Dumre SP, John S, Karbwang J, Hirayama K. Mycetoma: a clinical dilemma in resource limited settings. *Ann Clin Microbiol Antimicrob* 2018; 17:35.
4. van de Sande WW. Global burden of human mycetoma: a systematic review and meta-analysis. *PLoS Negl Trop Dis* 2013;7(11):e2550.
5. Verma P, Jha A. Mycetoma: reviewing a neglected disease. *Clinical and experimental dermatology* 2018. https ://doi.org/10.1111/ced.13642.
6. Al Gannass A. Chronic Madura foot: mycetoma and/or Actinomyces species or actinomycosis. *BMJ Case Rep* 2018. doi:10.1136/bcr-2018-224859.
7. Veraldi S, Grancini A, Venegoni L, Merlo V, Guanziroli E, Menicanit C, *et al.* Mycetoma caused by *Aspergillus nidulans*. *Acta Dermato Venereol* 2016;96(1):118–9. https ://doi.org/10.2340/00015 555-2144.
8. Ameen M. Managing mycetomas. *Trop Doc* 2009;39(2):66–8. https:// doi.org/ 10.1258/td.2008. 080151.
9. Fahal AH. Mycetoma: a thorn in the flesh. *Trans R Soc Trop Med Hyg* 2004;98(1):3–11.
10. Welsh O, Al-Abdely HM, Salinas-Carmona MC, Fahal AH. Mycetoma medical therapy. *PLoS Negl Trop Dis* 2014;8(10):e3218.
11. Drugs for Neglected Diseases Initiatives (DNDi). About Mycetoma, Available from https:// www.dndi.org/diseases-projects/ mycetoma/ Accessed on 23/02/2019.
12. Zein HA, Fahal AH, Mahgoub el S, El Hassan TA, Abdel-Rahman ME. Predictors of cure, amputation and follow-up dropout among patients with mycetoma seen at the Mycetoma Research Centre, University of Khartoum, Sudan. *Trans R Soc Trop Med Hyg*. 2012;106(11): 639–644.