

Nasal Subcutaneous Phycomycosis: An uncommon occurrence

Gustave Buname¹✉, Richard Byaruhanga¹, Kakande Emily¹, Justine Namwagala¹, David Alele², Michael Odida², Christopher Ndoleriire¹

1. Department of Ear, Nose and Throat Makerere University, Kampala, Uganda.

2. Department of Pathology, Makerere University, Kampala, Uganda.

✉ Corresponding author email: bishop4jesus@gmail.com

International Journal of Clinical Case Reports, 2014, Vol.4, No.7 doi: 10.5376/ijccr.2014.04.0007

Received: 25 Jul., 2014

Accepted: 27 Aug., 2014

Published: 28 Sep., 2014

Copyright © 2014 Buname et al., This is an open access article published under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.

Preferred citation for this article:

Buname et al., 2014, Nasal Subcutaneous Phycomycosis: An uncommon occurrence, International Journal of Clinical Case Reports, Vol.4, No.7 1-3 (doi: [10.5376/ijccr.2014.04.0007](https://doi.org/10.5376/ijccr.2014.04.0007))

Abstract Subcutaneous phycomycosis is a very rare fungal infection of the subcutaneous fats caused by Zygomatous group of fungi. We report an uncommon case of an adult male with nasal subcutaneous phycomycosis which presented as painless, non tender, itchy swelling on his nose.

Histopathological analysis of the biopsied tissues revealed chronic granulomatous inflammation containing fungal hyphae with surrounding langhans giant cells and eosinophilia. Periodic acid Schiff outlined the fungal hyphae and spores. The swelling resolved completely in the second month of treatment after the patient was started on oral potassium iodide and fluconazole.

Keywords Phycomycosis; Zygomatous fungi; potassium iodide; Azoles

Introduction

Subcutaneous phycomycosis is a fungal infection of the subcutaneous fats caused by Zygomatous group of fungi in the family Entomophthoraceae, mainly in the genus *Basidiobolus* (common in children) and *Conidiobolus* in adults (Prasad PV, 2002). It was firstly reported in Indonesia in 1956 (Joe LK, 1960). In Africa the first case of phycomycosis was reported from Uganda (1961) where the fungus was identified in tissues removed from a child in Mulago Hospital (Burkitt DP, 1964). Being a saprophytic fungus it can present with subcutaneous, gastrointestinal or acute illness symptoms. Subcutaneous phycomycosis is the commonest which normally presents with a painless subcutaneous induration that progressively increases in size (Sujatha S, 2003). Definitive diagnosis is made by histological findings and treatment is mainly by potassium iodide solution (Manjir RN, 2009). We present a rare case of an adult with nasal subcutaneous phycomycosis.

Case Report

A 32 years old man presented in our outpatient clinic with a painless nasal swelling for 3 months which was associated with nasal itching and mild nasal obstruction. He had no other nasal, otologic or ocular

symptoms. No headache or loss of consciousness with no feeding problems. He reported no fever, cough or night sweats. No history of trauma. Our patient is a peasant farmer from central Uganda. He keeps animals like goats, cows and sheep Figure 1.

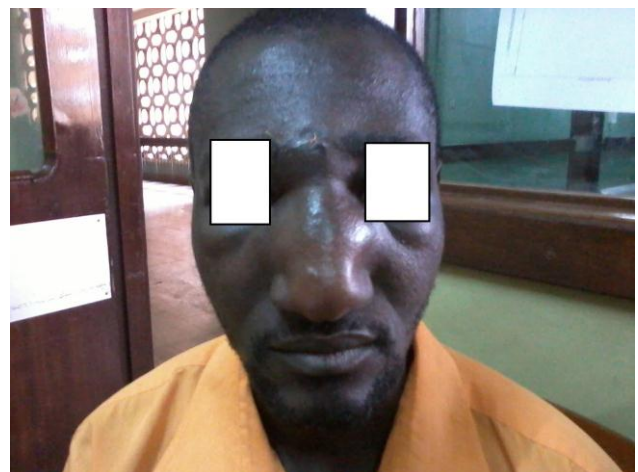


Figure 1 Nasal swelling

On examination the swelling was non-tender with stony hard consistency, smooth rounded edges, from the nasal tip extending to the nasal bridge, and about 5cm in width (laterally) and another small separate swelling above the nasal bridge with similar

consistency. It was extending inside pushing the roof of the nose down decreasing patency of the nasal cavity. Both the nasal septum and floor were unaffected. All other systems were essentially normal. Routine laboratory investigations like complete blood count, liver and renal function tests were all normal. X-ray of the nose revealed a soft tissue swelling without any bone involvement. An impression of subcutaneous phycomycosis was made on the basis of these clinical features.

A biopsy was taken and sent for histopathological analysis. Under Haematoxylin and Eosin stain chronic granulomatous inflammation containing fungal hyphae with surrounding langhans giant cells and eosinophilia was seen. Periodic acid Schiff outlined the fungal hyphae and spores Figure 2~4.

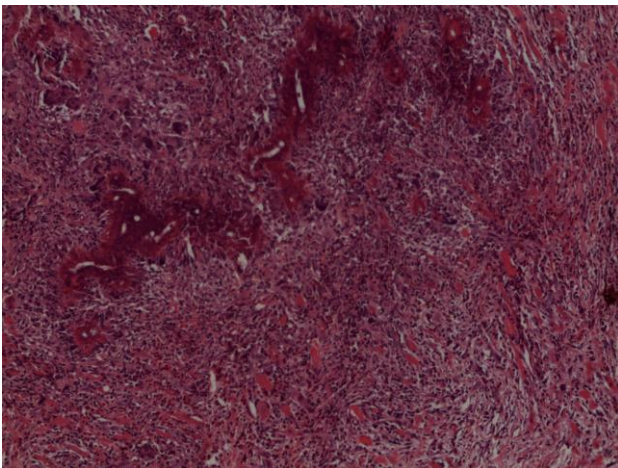


Figure 2 Granulomatous inflammation with numerous eosinophils

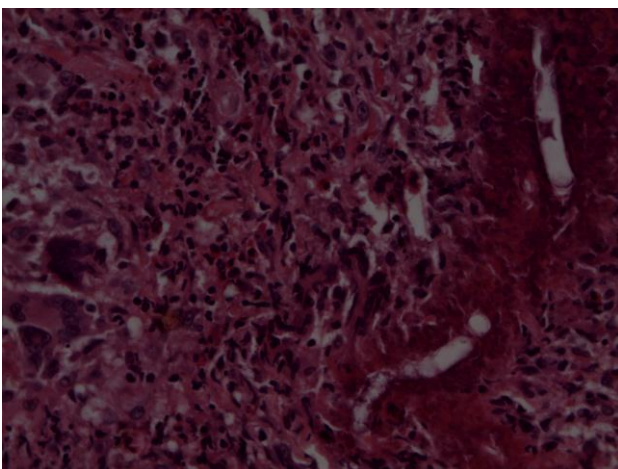


Figure 3 Under H and E section shows chronic granulomatous inflammation containing fungal hyphae with surrounding langhans giant cells and eosinophilia

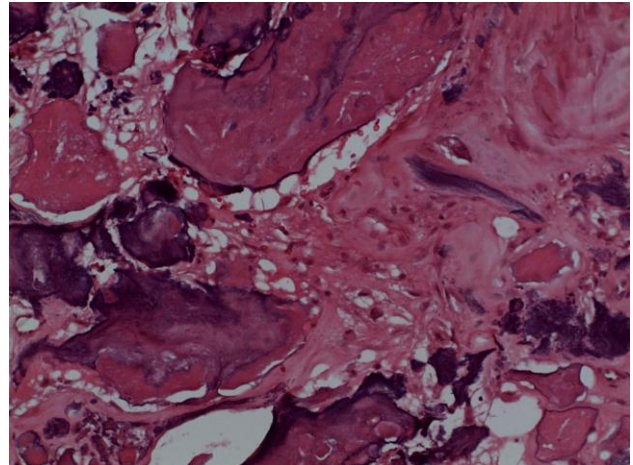


Figure 4 Periodic acid Schiff outlines the fungal hyphae and occasional spores

The patient was immediately started on potassium Iodide solution three drops thrice daily and increased daily up to 8 drops thrice in a day where the patient was maintained on it for a month. The swelling reduced significantly after one month, but the patient came back with severe nasal itching. Tabs Fluconazole 200mg once daily and cetirizine 10mg once daily were added in the next month of treatment. The patient improved, itching stopped and the swelling completely resolved after another month of treatment.

Discussion

Subcutaneous phycomycosis is a fungal infection of the subcutaneous fats caused by Zygomatous group of fungi in the family Entomophthoraceae, mainly in the genus *Basidiobolus* (common in children) and *Conidiobolus* in adults (Prasad PV, 2002). The causative organisms are *Basidiobolus ranarum* and *Conidiobolus coronatus* respectively. It can cause a variety of clinical manifestations including subcutaneous zygomycosis, gastrointestinal zygomycosis and occasionally an acute systemic illness.

Documentations as early as 1886 describe this fungus as a saprophyte in the gastro-intestinal tract of reptiles and amphibians. Soil and vegetation contaminated by faeces from these animals seems a likely source of the infection in man (Vismer HF, 1980). Reports have shown fungal spores to be found on the bristles of mites other insects. When these are eaten by reptiles and amphibians, the saprophytic cycle of the fungus is complete. Such mites and insects may comprise a further possible source of infection in man (HC, 1999).

In many instances, as it is evident in our patient, there is no history of trauma; hence insect bites could be the mode of infection. Our patient is a peasant farmer, keeps animals and lives in an area with a lot of Tsetse flies.

Granulomatous lesions due to *Basidiobolus ranarum* have been found in the nostrils and on the legs of horses, but reports on animal infection are scanty (Vismer HF, 1980). Little success has been achieved by inoculating laboratory animals with pathogenic strains. In humans a chronic infection of the subcutaneous tissue is usually produced characterized by the formation of firm and non-tender swellings, generally on the extremities, trunk and rarely other parts of the body. Our patient is among the rare cases as he presented with a nasal swelling. The disease appears to be a primary rather than a secondary infection, no predisposing factors are known (HC, 1999). The disease usually occurs in children, less often in adolescents and rarely in adults. Males are much more frequently affected than females.

Definitive diagnosis is made by histopathological analysis of the tissues from the swellings. Features are of extensive dermal and subcutaneous fibrosis and large zygomorphic hyphae. The hyphae appear as short sections of broad hyphae without septae. Granulomatous inflammatory features may also be present (Manjir RN, 2009).

Most patients with phycomycosis respond very well to oral potassium iodide therapy as also to azoles (Sujatha S, 2003). Our patient also responded well to potassium iodide and the swelling reduced dramatically after one month of treatment. Relapse chances are high in patient who undergo excisional biopsy (Manjir RN, 2009). Hence surgery should be avoided as it may hasten the spread of infection.

Conclusion

Subcutaneous phycomycosis of the nose is a rare fungal infection. Its diagnosis needs high index of

suspicion and it is confirmed by histopathological findings which are characteristic. Patients respond well on Potassium iodide solution and azoles. Surgical excision should be avoided as it may increase the chances of infection spread. Predisposing factors and its exact route of transmission is not clear, further studies are needed in this area.

Consent

Consent was obtained from the patient for publication of this case.

Competing interests

We declare that we have no competing interests

Authors' contributions

GB did the initial work-up and took biopsy, did literature review and wrote the initial manuscript. AD and MO helped in the preparation of the histopathological slides and did special staining. RB, EK, JN and CN helped in the literature review and writing of the manuscript.

Acknowledgement

We would like to thank all the Doctors and Nurses in our department who participated in the treatment of this patient. Many thanks to the Head of research in our department Dr A. Bugembe for his support.

References

- Burkitt DP, W. A., Jelliffe DB. (1964). Subcutaneous Phycomycosis: A review of 31 cases seen in Uganda. *British Medical Journal*, 1, 1669-1672
<http://dx.doi.org/10.1136/bmj.1.5399.1669>
- HC, G. (1999). A review of Zygomycosis due to *Basidiobolus ranarum*. *European Journal of Epidemiology*, 15(10), 923-929S
<http://dx.doi.org/10.1023/A:1007656818038>
- Joe LK, T. N. I., Pohan A. (1960). Subcutaneous Phycomycosis: A new disease found in Indonesia. *Archives of Dermatology*, 74, 378
<http://dx.doi.org/10.1001/archderm.1956.01550100046008>
- Manjir RN, S. V., Balkrishna PN et al. (2009). Subcutaneous phycomycosis in a child. *Online Journal of Health and Allied Sciences*, 8(3), 14
- Prasad PV, P. E., George RV, Ambujan S, Viswanthan P. (2002). subcutaneous phycomycosis in a child. *Indian Journal of Dermatology, Venereology, leprology*, 68, 303-304
- Sujatha S, S. C., Khyriem AB, Parija SC, THappa DM. (2003). Subcutaneous Zygomycosis caused by *Basidiomycosis ranarum*. *Journal of Medical microbiology*, 21(3), 205-206
- Vismer HF, D. B. H., Dreyer L. (1980). Subcutaneous phycomycosis caused by *Basidiobolus haptosporus*. *South African Medical Journal*, 58, 644