Case Report

Subcutaneous Phycomycosis—Fungal Infection Mimicking a Soft Tissue Tumor: A Case Report and Review of Literature

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Summary

Subcutaneous phycomycosis is a rare fungal infection of the deeper layers of skin. We describe a representative case. A 10-year-old boy presented with a large swelling on the back of 3 months duration. Biopsy of the lesion confirmed the diagnosis of subcutaneous phycomycosis. There was complete resolution of the lesion after treatment with oral potassium iodide for 3 months.

Key words: phycomycosis, Basidiobolus, potassium iodide.

Introduction

Subcutaneous phycomycosis is a fungal infection of subcutaneous tissues [1, 2]. As it mimics a soft tissue tumor, biopsy is essential to prove the diagnosis. It resolves completely on treatment with oral potassium iodide [1-7].

Case Report

A 10-year-old boy presented with swelling of the upper back of 3 months duration. The swelling was painless and patient was afebrile throughout. Examination revealed a large swelling $(20 \times 18 \text{ cm})$ over the upper back extending down over both scapulae and anteriorly to the neck and both axillae (Fig. 1). The swelling was indurated and nontender with distinct rounded edges. Skin was involved over the entire swelling. Underlying bone fixity was doubtful. There were a few discrete, nontender 1×2 cm lymph nodes in the posterior triangle on the right side. Systemic examination was unremarkable. Clinically, soft tissue sarcoma or plexiform neurofibromatosis was considered. Fine needle aspiration cytology showed a predominantly eosinophilic aspirate and histiocytic tumor was suggested. Biopsies were taken from the lesion and the cervical lymph node. The lesion showed broad infrequently septate thin-walled hyphae bordered by a brightly

Correspondence: Sundeep Payyanur Thotan, Assistant Professor, Dept of Paediatric Surgery, Kasturba Medical College, Manipal, India. Tel.: 91 820 2922116. E-mail: <sunip2@yahoo.com>. eosinophilic zone of granular Splendor Hoeppli material giving the diagnosis of subcutaneous phycomycosis. The fungal hyphae were weakly periodic



FIG. 1. Lesion on presentation.

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FIG. 2. Lesion 2 months after treatment.

acid Shiff (PAS) positive. X-rays did not show any bony involvement. The patient was treated with Lugol's iodine for 3 months and was monitored for side effects. Treatment was begun with 5 drops per day and increased gradually to 15 drops per day. The swelling showed complete resolution (Fig. 2). At 2 years follow up, the child has no lesions.

Discussion

The causative organism of subcutaneous phycomycosis is *Basidiobolus haptosporus/Basidiobolus ranarum* belonging to the Zygomatic species [2], Entomophthoraceae family [1].

The organism is found in the tropical forests of Africa and Asia, cases being reported from Indonesia [8], Africa [1] and India [2, 6, 7]. The fungus resides in decaying vegetable matter and as a saprophyte in the gut of insectivorous reptiles (lizards), amphibians and fish [1, 4]. Entry into skin is usually through an

innocuous skin trauma [1, 2, 4] or an infected insect bite [3], which often goes unnoticed. Following inoculation it produces a spreading painless swelling [1, 2, 4]. Boys are more commonly affected [1, 4]. Lesions are mainly seen in extremities followed by trunk [1, 2, 4]. A woody indurated feel, ability to insinuate fingers under the swelling virtually lifting it up and absence of systemic signs are characteristic clinical features [1, 2, 4]. Lymph nodes are usually not involved. Our case had few palpable nodes, biopsy however reported as nonspecific lymphadenitis. Biopsy is diagnostic showing broad thin-walled hyphae surrounded by brightly eosinophilic material (Splendor hoeppli protein), with the background showing a dense eosinophilic infiltrate [1, 4]. The hyphae stain positive with Gomori's stain, methenamine silver and Masson's trichrome and faintly with PAS [3]. The organism responds best to potassium iodide [8]. Treatment is with saturated solution of potassium iodide (SSKI) at a dose of $30 \text{ mg}^{-1} \text{kg}^{-1} \text{day}^{-1}$ [1]. Treatment is begun at five drops per day and gradually increasing the dose with monitoring for side effects [1, 6]. Complete resolution takes about 2-3 months [1, 2, 4, 6]. Lugol's iodine that contains potassium iodide was used in our case as SSKI was not available. As the patient showed response, the same was continued till complete resolution. This is the first case in literature treated with Lugol's iodine and could be a good alternative to potassium iodide as it is widely available.

References

- 1. Burkitt DP, Wilson AMM, Jelliffe DB. Subcutaneous phycomycosis; a review of 31 cases seen in Uganda. Br Med J 1969;1:1669–72.
- 2. Prasad PV, Paul EK, George RV, *et al.* Subcutaneous phycomycosis in a child. Indian J Dermatol Venereol Leprol 2002;68:303–4.
- Chandrasekhar HR, Shashikala P, Haravi R, Kadam RS. Subcutaneous phycomycosis. Indian J Dermatol Venereol Leprol 1998;64:89–90.
- 4. Gugnani HC. A review of zygomycosis due to Basidiobolus ranarum. Eur J Epidemiol 1999;15:923–9.
- 5. Joe LK, Eng NIT, Pohan A, *et al.* Basidiobolus Ranarum as a cause of subcutaneous mycosis in Indonesia. Arch Dermatol 1956;74:378–83.
- Lal S, Baruah MC, Padiyar NV. Clinicopathological study of subcutaneous phycomycosis. Indian J Dermatol Venereal Leprol 1984;50:245–8.
- Sivaraman, Thappa DM, Karthikeyan, Hemanthkumar. Subcutaneous phycomycosis mimicking synovial sarcoma. Int J Dermatol 1999;38:920–3.
- 8. Yangco BG, Okafor JI, TeStrake D. In vitro susceptibilities of human and wild-type isolates of Basidiobolus and Conidiobolus species. Antimicrob Agents Chemother 1984;25:413–6.