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Rare case report of localized cutaneous protothecosis in an immunocompetent male

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Abstract

Human protothecosis is a rare infection caused by members of genus Prototheca. The occurrence of protothecosis can be local or disseminated; the latter being more common in immunosuppressed patients. The skin is most commonly involved, resulting from primary inoculation through a wound or abrasion. It is a rare disease, with only 117 cases reported worldwide till date. Here we report the case of a 14 year old boy with localized cutaneous protothecosis presenting with eczematous lesions over arms and legs.

Introduction

Human protothecosis is a rare infection caused by members of genus Prototheca. Prototheca species are achlorophyllic algae and ubiquitous in nature. The occurrence of protothecosis can be local or disseminated the latter being more common in immunosuppressed patients. The skin is most commonly involved, resulting from primary inoculation through a wound or abrasion. The definitive diagnosis of infection depends upon finding characteristic structures observed on histopathological examination of tissue. [1]

Here we report the case of a 14 year old boy with localized cutaneous protothecosis presenting with eczematous lesions over arms and legs.

Case Report

A 14 year old male child presented in the Dermatology outpatient department of Government Medical College, Amritsar with skin lesions distributed asymmetrically over exposed areas of arms and legs in the form of multiple itchy papules with keratotic surface, coalescing at places to form plaques (**Fig 1,2**). There was a history of contact with soil and contaminated water as the child used to play outdoor cricket with his friends and at times he had to take out the ball from drains. In an attempt to get rid of these lesions, patient used to scrub the lesions with pumice stones.



Fig 1: Lesions over the extensor aspect of left arm are in the form of multiple skin coloured papules arranged in a circular pattern coalescing to form plaques on an erythematous base. Secondary eczematization is seen. Few satellite papules are present.



Fig 2: Lesions over the right infrapatellar region shows multiple hyperpigmented follicular papules arranged in a circular pattern.

Atypical mycobacterial diseases, chromoblastomycosis, foreign body reaction, sarcoidosis, keratotic disorders were kept in the differentials.

Hematological profile (Hb12.5g/dl, total leukocytic count 9800/cu mm, differential leukocytic count: neutrophils 66/ lymphocytes 42/ eosinophils 02/ monocytes 02, ESR14mm, PBF showed normocytic normochromic anemia) and urine examination were normal. Antibodies against HIV were not detected and Mantoux test was normal. Radiographs of chest & hands were normal.

Histopathological examination of the lesions obtained from arms and legs showed acanthosis, ortho-hyperkeratosis, focal parakeratosis and plasma exudation. In the dermis, dense perivascular inflammatory infiltrate composed of lymphocytes, plasma cells, giant cells and occasional eosinophils was noticed. Eosinophilic and basophilic endosporulating spherules of prototheca species were seen in the dermal infiltrate and in the giant cells. Transepidermal elimination of prototheca was also noticed. (Fig 3,4,5,6)

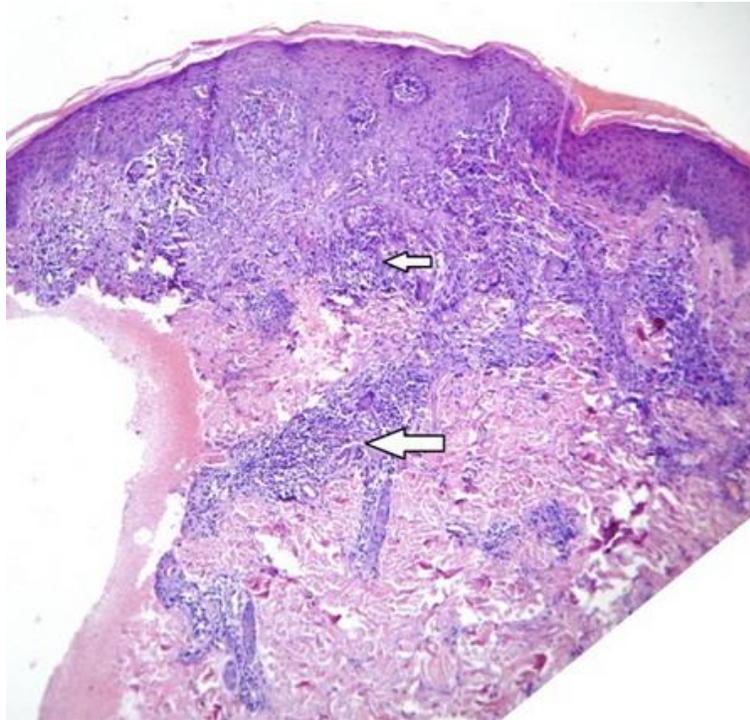


Fig 3: Acanthosis, orthohyperkeratosis plasma exudation, superficial and deep perivascular infiltrate mainly composed of lymphocytes, plasma cells and giant cells. In giant cells prototheca species were seen under low magnification.

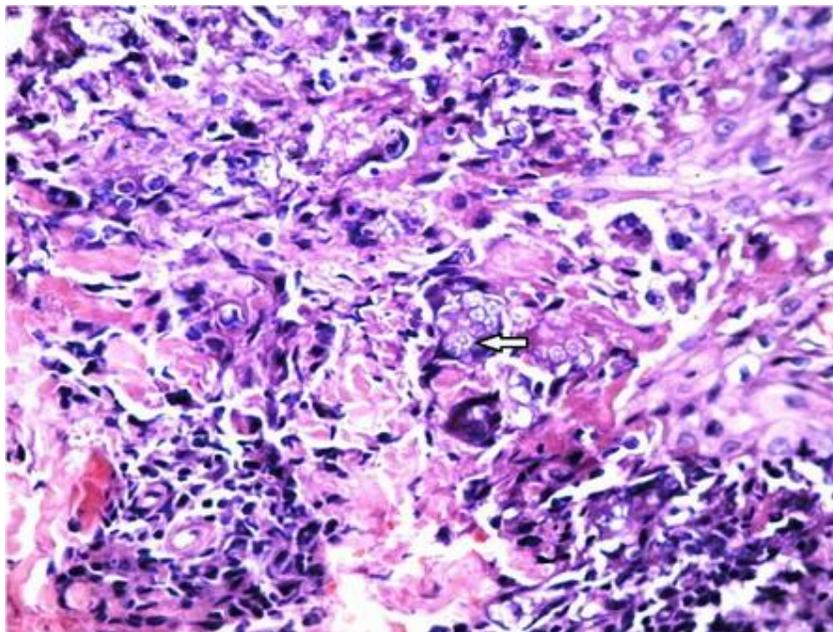


Fig 4: Basophilic spherules of prototheca are seen (40X).

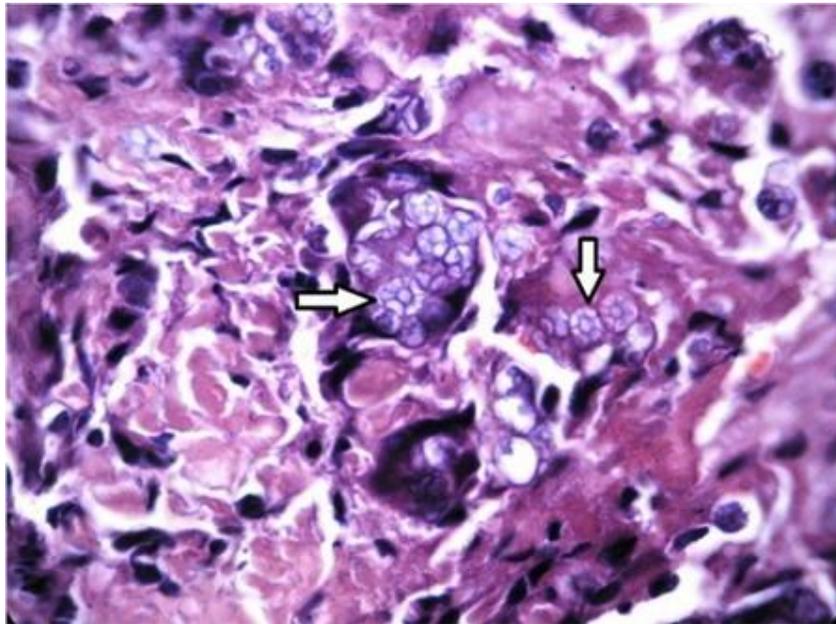


Fig 5: Light basophilic spherules of prototheca seen under oil immersion.

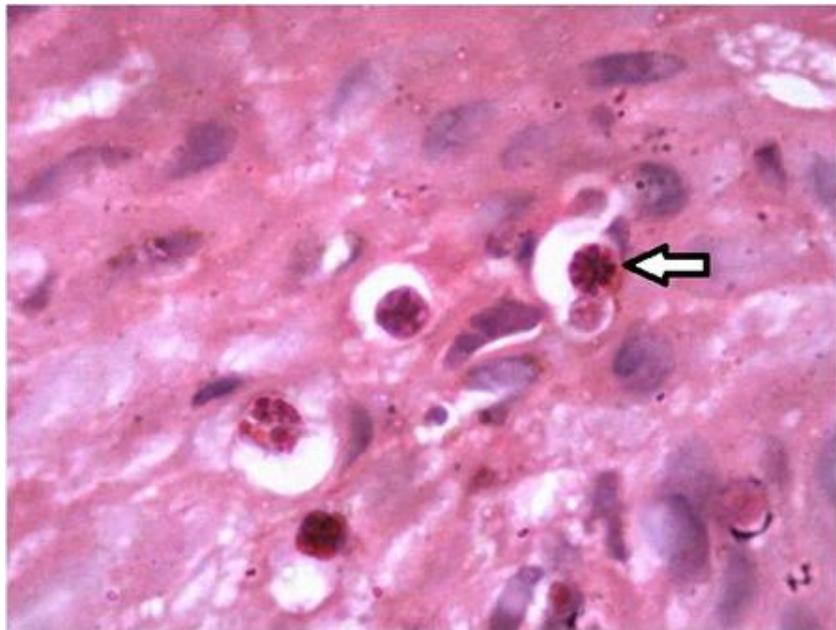


Fig 6: Transepidermal elimination of eosinophilic spherules of prototheca is seen (oil immersion).

Culture: After incubation for 48 hours on sabouraud's dextrose agar at 28°C and 37°C, smooth creamy white moist colonies were obtained. Further subcultures and biochemical tests could not be performed due to bacterial contamination of the culture.

Patient was put on Itraconazole 200mg/ day for 20 days and he showed significant improvement. After this, the patient became irregular and was subsequently lost to follow up.

Discussion

The first description of human infection attributed to prototheca species was made by Davies and colleagues in 1964 on the foot of a barefoot rice farmer from Sierra Leone. [2] Prototheca is a rare cause of opportunistic infection in humans. Most of these infections are possibly due to the traumatic inoculation into the subcutaneous tissue. Localized cutaneous infections are seen in immunocompetent patients whereas dissemination is seen in patients with compromised immunity especially cell mediated immunity. [3]

Protothecosis can present as: [1]

- Cutaneous lesions
- Olecranon bursitis
- Disseminated or systemic manifestations

Infection may occur by penetration of agent when injured skin comes in contact with contaminated water. Till now 117 cases of prototheca infection have been described in the literature, out of which 66% were associated with cutaneous infection, 19% with systemic infection and 15% with olecranon bursitis. Prototheca species have been found to colonize the human skin, finger nails, respiratory and digestive systems. [1]

Rare case of disseminated protothecosis in a child with combined immunodeficiency and failure to eradicate the infection with amphotericin B has been reported from India. [4]

Incubation period is not well documented, [5] it varies from 10 days to 4 months. [6] Potential sources of infection are contaminated soil, water, insect bite and surgical and orthopedic procedures. [7] There is no racial or sexual predilection. It typically affects elders although pediatric cases have been reported. [6]

Prototheca species are globally ubiquitous. [8] They can be isolated from various environmental reservoirs, animals and dirty water. [9] Even chlorination of water is not effective in eliminating the organism. Prototheca are opportunist organisms with minimal pathogenicity,

requiring an alteration in host resistance before they can act as pathogens. [10]

The various skin presentations can be erythematous plaques, pustules, papules, nodules or verrucous lesions, pyodermic, herpetiform, vesicles, ulcers, hypopigmented or atrophic lesions. [11]

Various treatment modalities like Amphotericin B, ketoconazole, itraconazole, fluconazole, tetracyclines, gentamycin are indicated in cases where disease is diffuse, while surgery is indicated in localized lesions. The duration of treatment varies from days to weeks. [12]

To the best of our knowledge, the disease is extremely rare in humans as only few hundred cases have been reported throughout the world and only one case has been reported from India (Pubmed, Medline, Cochrane library, Medscape). Even the case reported in India was in a child with lowered immunity and having disseminated disease with no response to Amphotericin B. [4] Our case is being reported due to its presentation in a healthy immunocompetent child. Through our case report, we want to emphasize the need to diagnose this infection in early stage in order to prevent dissemination. Also, high index of suspicion should be kept in recurrent and nonresponsive cases with similar presentation.

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