CASE REPORT

Cutaneous protothecosis: a case report

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Introduction: Cutaneous protothecosis usually presents as pyoderma-like lesions or infiltrating papules and plaques on the extensor side of the extremities. It can be misdiagnosed as eczema, pyoderma, or a fungal infection. Although it has been isolated from a swimming pool, sewers and rivers in the Philippines, there has been no reported case of cutaneous protothecosis in the country.

Case summary: A 78-year-old Taiwanese male farmer visited the dermatology clinic due to a six-month history of a large, pruritic erythematous plaque studded with papulopustules on his left forearm. A potassium hydroxide (KOH) examination showed negative for hyphae or spores. And a skin biopsy showed morula-like bodies, which were highlighted by the Periodic acid-Schiff stain.

Conclusion: We report a case of cutaneous protothecosis from Taiwan so Filipino dermatologists will be aware of the clinical and histopathologic manifestations and management of cutaneous protothecosis.

Keywords: Protothecosis, dermatopathology, tropical dermatology

Introduction

Human protothecosis was first described on a rice farmer's foot in 1964 at Sierra Leone by Davies et al.[1] The etiologic agent is attributed to an achlorophyllic alga belonging to the *Prototheca* genus, which is found in sewage and soil worldwide.[1] This organism can colonize the human digestive system, respiratory tract, skin, and nails.[1,2] The cutaneous manifestations range from localized to disseminated papules, plaques, pustules or nodules. [1] Protothecosis has been reported in every continent except in Antarctica.[2] Although *Prototheca* sp. has been isolated in Philippine water sources, it is surprising that there are no published cases of cutaneous protothecosis in the Philippines nor reported cases by the Philippine Dermatological Society's Health Information System (PDS-HIS).[1,2]

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Case report

A 78-year-old Taiwanese male farmer visited the dermatology clinic of National Cheng Kung University Hospital due to a large, pruritic erythematous plaque studded with papulopustules on his left forearm **(Figure 1A)**. The lesion had been present for more than six months. He could not recall any history of trauma before the onset of the lesion. He had been on maintenance medications for congestive heart failure and diabetes mellitus for several years. There had been no prior treatment for the skin lesions, nor had herbal concoction been taken or applied. Potassium hydroxide (KOH) examination was negative for hyphae or spores. A biopsy specimen of the skin lesion revealed suppurative and granulomatous inflammation in the dermis with morula-like bodies, which were highlighted by the Periodic acid-

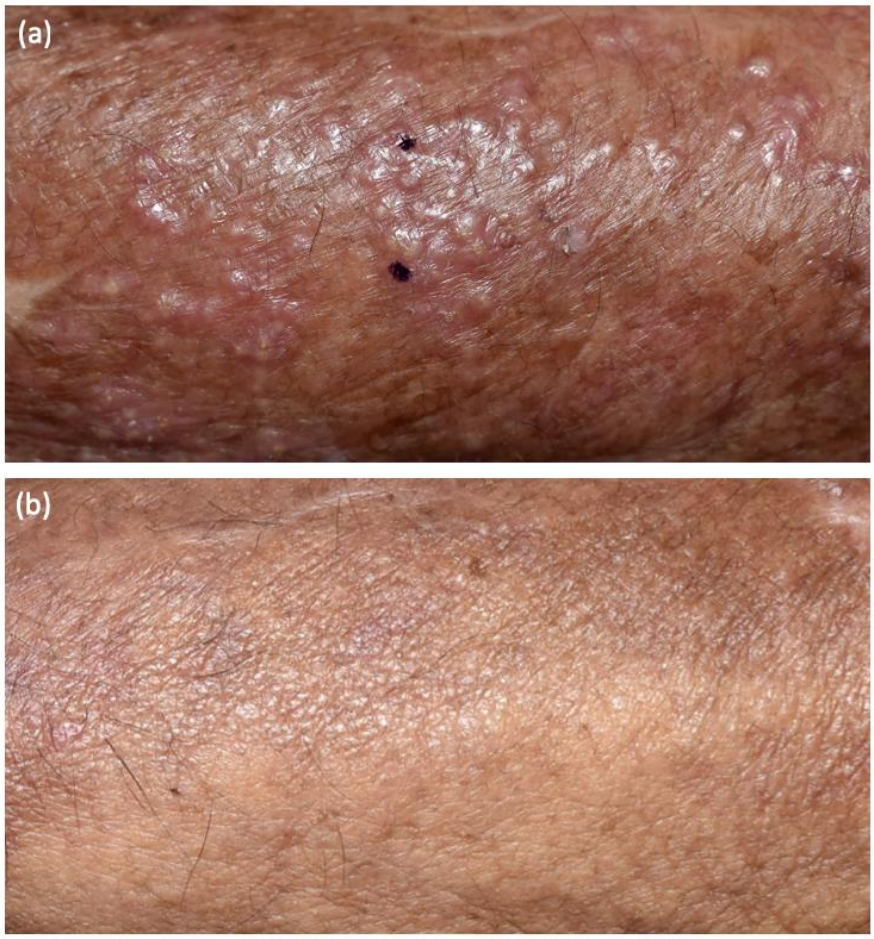

Figure 1. Clinical findings of cutaneous protothecosis: (a) a large erythematous plaque on the extensor of the forearm studded with papulopustules; and (b) complete resolution of the skin lesions after 3 months of itraconazole therapy.

Schiff stain **(Figure 2A-D)**. He was treated with itraconazole 100mg twice a day for three months, which resulted in complete resolution of lesions **(Figure 1B)**.

Discussion

Protothecosis has been reported to affect humans and animals, mostly through traumatic inoculation into the skin.[1] Most human infection is caused by *P. wickerhamii*. Protothecal infections commonly occur in patients with underlying immunosuppression or underlying diseases such as hematologic malignancy or cancer, diabetes mellitus, acquired immunodeficiency, and prolonged local or systemic steroid use. [9] A study of cutaneous protothecosis by Tseng et al. shows that most patients in Taiwan were industrial workers or farmers living in rural tropical areas.[5] Furthermore, their findings show that Iatrogenic Cushing Syndrome (75%) was the most significant risk factor for protothecosis. It has been noted that these patients were taking systemic corticosteroids or black pills (Chinese traditional medicine containing corticosteroids) for pain control such as arthritis. In that case series, chronic arthralgia and diabetes mellitus were the significant associations at 50% and 30%, respectively.[5]

Cutaneous protothecosis usually presents as pyoderma-like lesions or infiltrating papules and plaques on the extensor side of the extremities,[1] as seen in our case. Clinically, the skin lesions may be diagnosed as eczema, pyoderma, tinea corporis, Majocchi's granuloma, deep fungal infection or lupus vulgaris. Hence, KOH smear, culture, and skin biopsy are important for the diagnosis. The *Prototheca* sp. grows on Sabouraud dextrose agar with a spoked wheel appearance but differs from fungi by lacking glucosamine in their cell wall.[9] However, the yield

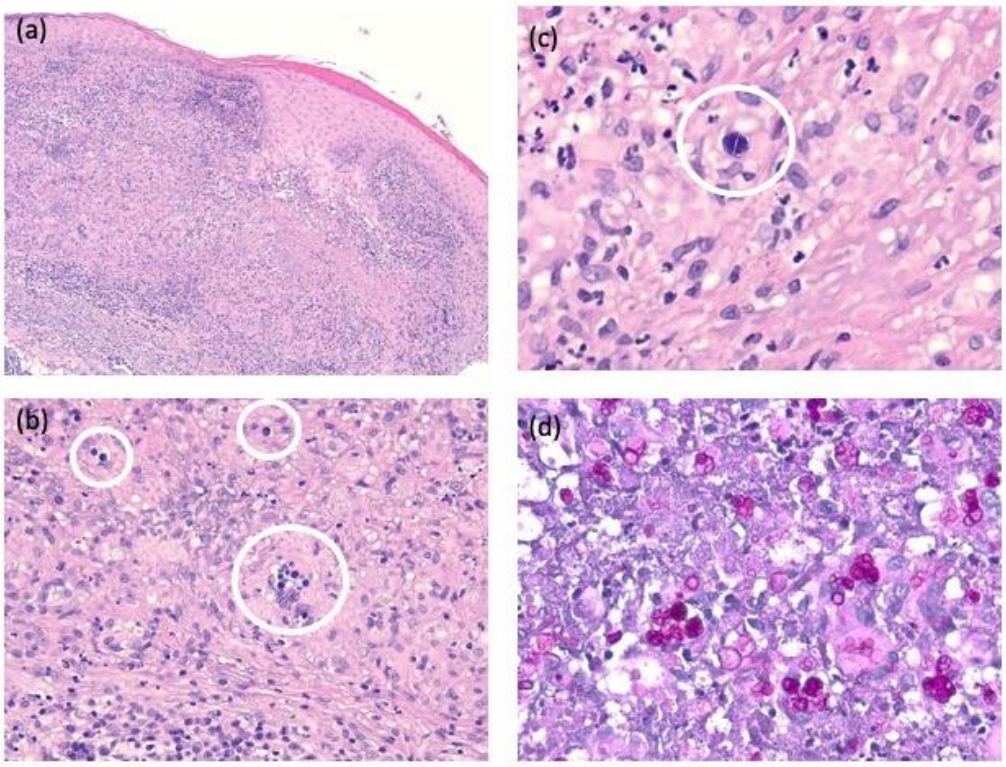

Figure 2. A suppurative and granulomatous infiltrate in the dermis with morula-like bodies (white circles) (Hematoxylin and eosin stain, x40 [a], x100 [b], x400 [c]); *Prototheca* sporangia highlighted by the Periodic acid-Schiff stain (PAS stain, x400 [d])

rate for culture is usually less than 50%.[5] Hence a skin biopsy is a better alternative for diagnosis.

The histopathologic features of cutaneous protothecosis are epidermal hyperplasia and dermal granuloma with suppurative changes, and the pathognomonic sporangia with morula-like structures can be highlighted by Gomori methenamine silver and Periodic acid-Schiff stains.[5,10] The large nonbudding cell (3–30 µm) is readily seen in the superficial dermis, which has a spheroid, ovoid, or elliptical shape with a prominent wall containing several thick-walled autospores **(Figure 3)**.[9] The unique morphology of *Prototheca* sp. endospores can be distinguished from Coccidioidomycosis, Blastomycosis, Paracoccidioidomycosis, Cryptococcus, among others **(Figure 3)**.

Prototheca sp. has been isolated from Philippine water sources such as the Pasig River (Manila), a swimming pool (Rizal Province), and sewer canals (Manila & Angeles City).[7] It is surprising however, that there has been no published case of cutaneous protothecosis in the Philippines nor reported cases through the National dermatology registry.[8] It is possible that there could be cases of cutaneous protothecosis in the Philippines, but the cases might have been diagnosed and treated as a fungal infection without pathological or microbiological confirmation since the infection is responsive to systemic antifungal therapy. Although there have been no standard therapies due to the rarity of the disease, reported cases have shown a good response to itraconazole, fluconazole, voriconazole, or amphotericin B with a range of 2 weeks to 6 months of treatment duration.[5, 10]

Conclusion

Cutaneous protothecosis has yet to be reported in the Philippines. While unreported in the PDS-HIS, cases of cutaneous protothecosis might have been treated as deep fungal infections. Dermatologists in the Philippines should be aware of cutaneous protothecosis and include this infection in the differential diagnosis of pyoderma, cellulitis, and deep fungal infection, particularly if a

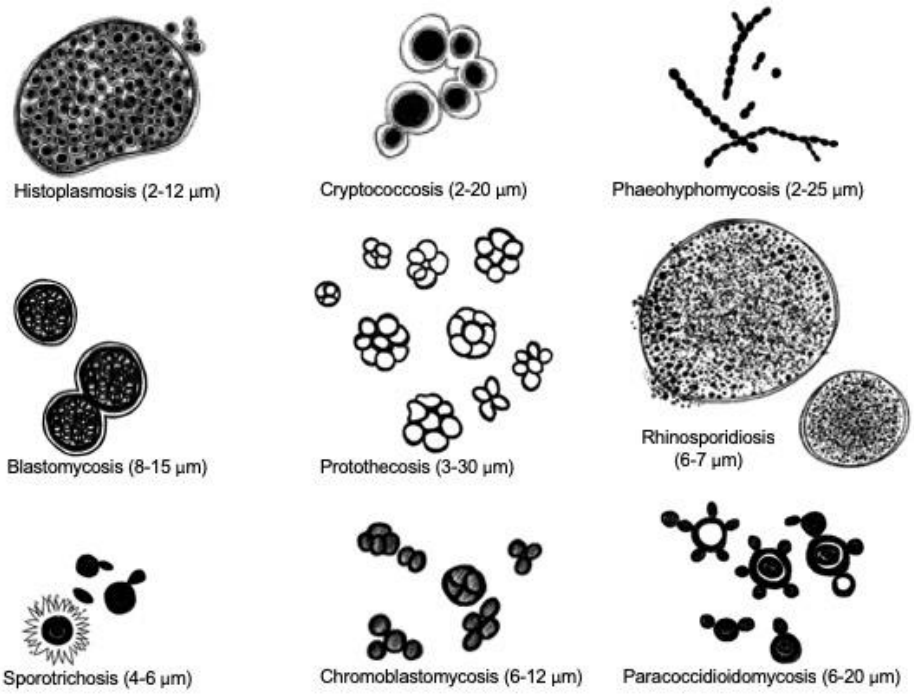

Figure 3. Morphology of *Prototheca* sp. sporangia compared to other spores.

patient presents with chronic plaques on the extremities with a history of exposure to contaminated water and chronic use of steroids. A skin biopsy that highlights the morula-like sporangia in a granulomatous infiltrate remains the cornerstone for the diagnosis.

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