

Chromoblastomycosis with a sporotrichoid distribution

Sir,

An 82-year-old farmer presented with multiple asymptomatic verrucous growths on the left lower limb of 23 years duration. He could not recall any definite history of trauma, before the appearance of the lesions. On examination, there were three discrete verrucous plaques on the left leg [Figure 1a]. Multiple black spots were seen on the verrucous plaques extending from medial malleolus to lateral malleolus covering the entire anterior aspect of left ankle [Figure 1b]. Possibility of cutaneous chromoblastomycosis, tuberculosis verrucosa cutis and lichen planus hypertrophicus was considered. There was no regional or generalized lymphadenopathy. Complete blood count, liver function test and renal function test were within normal range. X-ray of the limb did not reveal any abnormality. Examination of the black dots in

10% potassium hydroxide (KOH) preparation did not show any spores. Histopathological examination of the lesion showed hyperkeratosis and papillomatosis with brownish thick-walled copper-penny bodies within mixed inflammatory infiltrates composed of neutrophils and lympho-histiocytes in the superficial dermis [Figure 2a and b]. Culture of the tissue biopsy specimen did not grow any organism. A diagnosis of chromoblastomycosis in sporotrichoid pattern was made based on morphology of the lesions and histopathology findings. The patient was treated with itraconazole 200mg twice daily. There was a marked response after 2 months of treatment [Figure 3]. He is on regular follow-up and continuing the treatment.

Chromoblastomycosis is a chronic cutaneous and subcutaneous fungal infection caused by dematiaceous fungi, following traumatic implantation of the organism.¹ It can spread by autoinoculation and rarely through lymphatics.^{2,3}

Cutaneous lesions begin as erythematous papules, cauliflower-like nodules or warty growths. They gradually enlarge to form large plaques with brown or black dots on the surface. The most common sites of involvement are the distal limbs. Rarely, it occurs on buttocks, trunk and face.¹ In advanced disease, it can spread to adjoining skin as satellite lesions. Such spread occurs from autoinoculation or transmission via the lymphatic system.⁴ In a review of literature, we found four cases of chromoblastomycosis with sporotrichoid distribution.^{1,4-6}

The sporotrichoid distribution of lesions is seen in sporotrichosis, cutaneous tuberculosis and atypical mycobacterial infection. Chromoblastomycosis can be differentiated from these conditions by identification of the copper-penny bodies, histopathology and culture.



Figure 1a: Multiple verrucous plaques on left leg in a linear distribution, forming a sporotrichoid pattern



Figure 1b: Black dots on the surface of verrucous plaques

Table 1: Comparison of cases of chromoblastomycosis with sporotrichoid pattern

Author	Age/sex/occupation	Geographic area of origin	Clinical morphology/site	Investigation findings	Treatment/duration/response
Muhammed <i>et al.</i>	40-year-old male agricultural worker	Kerala, India	Nonhealing ulcer on the right big toe and multiple nodules on the anterior aspect of the right foot and leg	Histopathology and culture showing <i>F. pedrosoi</i>	Oral itraconazole 100 mg daily. Skin lesions had regressed by 80% after 6 months of therapy
Nair <i>et al.</i>	21-year-old man working in Punjab	Punjab	Multiple discrete smooth soft nodules distributed in a linear pattern on the dorsum of the right hand and forearm	Biopsy with brownish thick-walled spherical bodies with a split-pea appearance	Concentrated solution of potassium iodide in a dose of three drops thrice daily with increasing dose for 4 weeks
Turkowski <i>et al.</i>	33-year-old male	History of travel to South Africa, Asia, Middle East	Erythematous nodules and subcutaneous papules in lymphatic distribution on anterior aspect of right lower leg	Histopathology and culture showing <i>C. carrionii</i>	Itraconazole 200 mg daily for 3-12 months
Kawtar <i>et al.</i>	70-year-old farmer	Morocco, North Africa	Vegetative nodules and plaques on the fingers and dorsum of right hand and right forearm in a sporotrichoid arrangement, associated with significant lymphedema	Culture positive followed by KOH positivity, <i>F. pedrosoi</i>	Oral terbinafine 500 mg daily for 3 months with cryotherapy
Our case	82-year-old farmer	India, eastern part	Verrucous plaques with linear distribution on left leg	Culture and KOH mount did not show any finding. Histopathology showed typical copper-penny bodies	Itraconazole 200 mg twice daily for 2 months

F. pedrosoi: *Fonsecaea pedrosoi*, *C. carrionii*: *Cladophialophora carrionii*, KOH: Potassium hydroxide

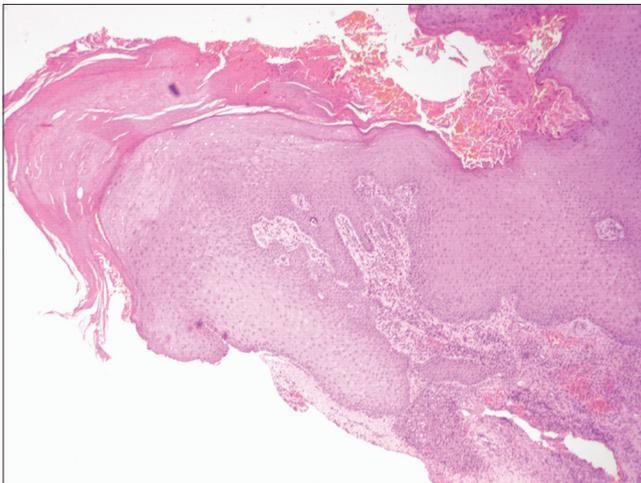


Figure 2a: Histopathological examination showing papillomatosis, acanthosis and collection of inflammatory infiltrates in upper dermis (hematoxylin and eosin, ×40)

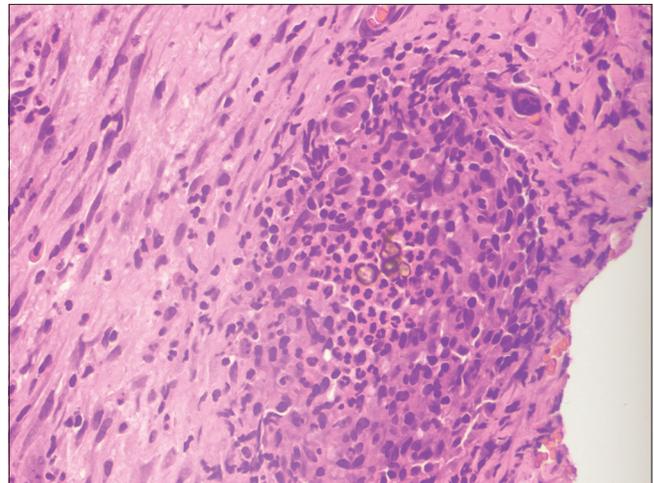


Figure 2b: Histopathology showing brownish circular copper-penny bodies surrounded by inflammatory infiltrates (hematoxylin and eosin, ×400)

Usually, histopathology reveals acanthosis and may demonstrate pseudo-carcinomatous hyperplasia. Neutrophils and giant cells may be seen infiltrating the epidermis with occasional formation of micro-abscesses. The dermis reveals a granulomatous tissue reaction with a mixed focal or diffuse inflammatory infiltrate consisting of lymphocytes, neutrophils, monocytes, plasma cells, eosinophils and giant cells of the foreign body and Langhans types. The

copper-penny bodies may be seen both intracellularly and extracellularly.

The diagnosis of chromoblastomycosis is from microscopic identification of Medlar bodies/fumagoid bodies/muriform bodies/copper-penny bodies/sclerotic bodies on scrapings from the lesion in 20% KOH and/or histological examination of a biopsy specimen and by culture of scrapings or biopsy material.¹ Our patient had multiple nodules in a linear fashion



Figure 3: Marked response after 2 months of oral itraconazole 200 mg twice daily

distant from the primary plaque and the diagnosis was confirmed by demonstration of copper-penny bodies in tissue section.

Treatment options include itraconazole, terbinafine and potassium iodide. Our patient was treated with oral itraconazole 200mg twice daily and after 2 months there was good response without any side effects of therapy.

In chromoblastomycosis with satellite lesions, presentation in a sporotrichoid fashion is rare.¹ In our case, based on the morphology, autoinoculation following scratching or minor trauma might have been the cause rather than lymphatic spread. We could not isolate the organism in culture, but the histopathology and response to treatment proved the diagnosis retrospectively. Though isolation of organism in culture is regarded as the gold standard for diagnosis, in resource-poor centers, classical morphology and histopathology can be the pointers to diagnosis. We have compiled and compared the four cases with our case in regards to clinical morphology, site of involvement, investigations and treatment outcomes [Table 1].^{1,4-6} We report this case because of its rarity and response to monotherapy with itraconazole.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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Access this article online	
Quick Response Code:	Website: www.ijdv.com
	DOI: 10.4103/ijdv.IJDVL_16_19

How to cite this article: Rout AN, Sahu K, Sirka CS. Chromoblastomycosis with a sporotrichoid distribution. *Indian J Dermatol Venereol Leprol* 2019;85:506-8.

Received: February, 2019. **Accepted:** April, 2019.

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