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Original Research Paper

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CHROMOBLASTOMYCOSIS: A REPORT OF 2 CASES WITH LITERATURE REVIEW

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ABSTRACT Chromoblastomycosis is primarily a cutaneous mycosis caused by demetiaceous fungi. Microscopic examination with findings of muriform bodies are pathognomonic for the diagnosis of Chromoblastomycosis and if not specifically looked for, there is a chance of missing the diagnosis leading to improper treatment and complications. Here we report 2 cases, one of a middle aged woman, the other of an elderly man presenting with lower limb skin lesions histopathologically diagnosed as chromoblastomycosis.

KEYWORDS : Chromoblastomycosis, Fungal Skin Diseases, Dermatitis Verrucosa, Deep mycosis

INTRODUCTION

Chromoblastomycosis is a type of deep cutaneous mycosis caused by a variety of demetiaceous or melanised fungal agents. It occurs worldwide with tropical and subtropical regions considered to be endemic for the disease. Initially described in 1914 by Dr. Max Rudolph, a physician, it was a year later, in 1915 that Medlar and Lane discussed the histological aspects, now eponymous as 'Medlar bodies'.[1]

It is a gradually progressive disease, generally causing cutaneous and subcutaneous granulomas. Eventually the disease may lead to complications such as ulceration, involvement of lymphatic channels etc. Thus, the extent of involvement is in direct proportion to the swiftness in diagnosis and treatment.[1,2]

Here we describe two cases, one in a middle-aged female, the other in an elderly male presenting at a tertiary-care centre based in coastal Karnataka, India.

Case Reports

Case 1

A 45 year old lady presented with an itchy raised lesion over the dorsum of the left foot from 2 months, which gradually progressed to achieve the size at presentation. On examination, a well-circumscribed, reddish & scaly plaque measuring about 35 by 20 millimeters was visualised. Although the patient did regularly work bare-feet in her kitchen garden, she did not recollect any history of trauma. General physical examination, and routine laboratory evaluation revealed no significant abnormalities. A skin biopsy was done with differential diagnoses of Lupus vulgaris, Tuberculosis verrucosa cutis, deep mycosis, and sarcoidosis.

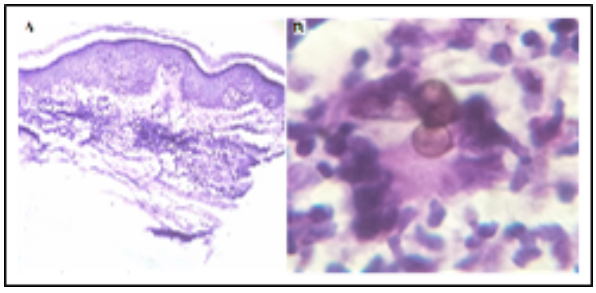

Figure 1: A. Scanner view showing acanthosis, dense inflammatory infiltrate in the mid, lower dermis, & subcutaneous tissue (H&E, x40). B. High power view showing muriform bodies adjacent to a Langhan's giant cell (H&E, x400)

Histopathology of the skin biopsy sent revealed epidermis showing hyperkeratosis, irregular acanthosis. Dermis showed intense lymphoplasmacytic inflammation which was most prominent in the deep dermis and subcutaneous tissue, with granulomas comprised of epithelioid histiocytic aggregates. A few characteristic melanised fungal elements, the muriform bodies were seen. Staining with Periodic Acid Schiff confirmed it and aided in clinching the diagnosis of Chromoblastomycosis. The microscopic findings are as seen in Figure 1.

The patient was put on oral Itraconazole for 6 months, underwent symptomatic remission of the disease.

Case 2

A 62 year old gentleman, a known case of type 2 Diabetes Mellitus, presented with an ulcerated lesion over his right leg from 1 and a 1/2 months, which gradually progressed to the present size. The patient mentioned of a traumatic event at the same site prior to the development of the nodule. On examination, a discrete, verrucous lesion of 45 by 35 millimeters was seen over distal anterior aspect of his right leg. Right inguinal lymph nodes were enlarged, rest of the physical examination was nil significant. With differentials of Lupus vulgaris and sarcoidosis, a skin biopsy was taken.

Histopathological examination of the lesion showed irregular epidermis with presence of muriform bodies in the stratum corneum. Dermis showed mixed inflammation, predominantly lymphoplasmacytic, along with microabscesses, and granulomas comprised of epithelioid histiocytic aggregates and Langhan's giant cells. Also seen in the dermis were muriform bodies. Periodic Acid Schiff stain further highlighted these melanised bodies, while special stain for Acid Fast Bacilli was found to be negative. The microscopic findings are as seen in Figure 2.

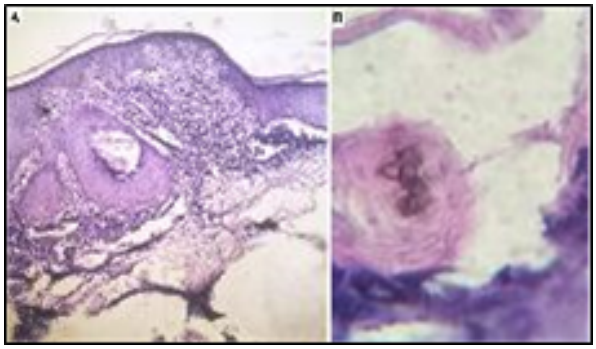

Figure 2: A. Scanner view showing irregular acanthosis,

dense inflammatory infiltrate of entire dermis, subcutaneous tissue (H&E, x40). B. High power view showing muriform bodies in the hyperkeratotic stratum corneum (H&E, x400)

With the diagnosis of Chromoblastomycosis, patient was put on oral Itraconazole for 6 months, but was lost to follow-up.

DISCUSSION & LITERATURE REVIEW

Chromoblastomycosis is a cutaneous and subcutaneous mycosis with a chronic course involving granulomatous inflammation. It was historically known by a myriad of names such as 'Chromomycosis', 'Cladosporiosis', 'Fonseca's disease', 'Pedroso's disease', 'Phaeosporotrichosis', 'Verrucous dermatitis' etc.[1,3]

The causative agents collectively are referred to as 'melanised molds'or 'demetiaceous fungi', with majority of the cases of Chromoblastomycosis being caused by Fonsecaea pedrosoi (commonest) and Cladophialophora carrionii. Other fungi include Phialophora verrucosa, Botryomyces caespitosus, Rhinocladiella aquaspersa, Aureobasidium pullulans, Exophiala jeanselmei, and Exophiala dermatitides.[2]

With a worldwide distribution, regions with highest prevalence include mostly tropical and subtropical countries. Brazil, Mexico, Madagascar, Malaysia, and Australia; in Asia, Japan and China, have reported highest prevalence. Prevalence and incidence in India cannot be exactly established as literature shows sporadic case reports in several regions such as Jammu & Kashmir, Bihar, Assam, Western & Eastern coast, and South India.[1,2]

In Brazil, the country with high prevalence, Fonseca pedrosoi fungi have been isolated from the thorns of Touch-me-not plant (also known as the Shame plant, scientifically as Mimosa pudica). In Venezuela, thorns of cacti were found to harbour Cladophialophora carrionii. These fungi can also be found in wood and soil. A history of a thorn-prick or splinters, thus, could be a useful pointer towards the diagnosis of Chromoblastomycosis.[2,4,5]

In the cases we reported, the second case i.e. the 62 year old male patient, did provide a clinical history of a traumatic event. However, the 45 year old female failed to provide any such detail. Thorn-pricks generally are trivial incidents which people tend to forget.

70 to 90% of the patients of Chromoblastomycosis are found to belong to the male gender, generally over the age of 30 years. Childhood presentation although rare, isn't unheard of. In Japan, however, the involvement was found to be same among both males and females.[2,5]

In the two cases we reported, no gender prediliction was noted, however, a large sample size is needed to note gender prediliction.

Risk factors for Chromoblastomycosis include working barefooted with lack of protective gear as boots, and immunosuppression. One of the studies in literature showed that HLA A29 allele was more prevalent among the patients of Chromoblastomycosis.[2]

One of the cases we reported was a known case of Diabetes Mellitus, which causes immunosuppresssion.

The lesions of Chromoblastomycosis typically begin at the site of trauma which commonly include feet and legs. Involvement of sites such as face, arms, genitalia although rare, have been reported in the literature.[4,6]

Initially a tiny, pink, smooth papular lesion develops, eventually increasing in size and forming scaly surface. Further clinical appearance may be polymorhic with one or more of the following appearances viz. nodular, cauliflower-like, verrucous, cicatricial, or plaque forms. A severity scoring system based on the morphology is given in Table 1.

Table 1. Queiroz-Telles et al Scoring system for severity of Chromoblastomycosis[2]

Severity	Morphology
Mild	Solitary plaque/ nodule with maximum diameter ≤ 5cm
Moderate	Solitary or multiple adjacent nodules/plaques/cauliflower-like lesions with maximum diameter ≤ 15cm
Severe	Non-adjacent/ extensive cutaneous lesions of any type

Both of the cases we reported fell under the Mild severity category.

Complications of Chromoblastomycosis include ulceration, lymphedema, secondary infection, and rarely evolves into a squamous cell carcinoma.[1,2]

Diagnosis ideally involves direct microscopy with KOH mount & culture of specimen such as skin scrapes, exudate, and also histopathology with special stains.[5,7]

Histopathological examination usually shows acanthosis or pseudoepitheliomatous hyperplasia of the epidermis. All the fungal species causing Chromoblastomycosis reveal a chronic granulomatous response, with the presence of golden-brownish muriform bodies arranged in singles or clumps. These muriform bodies are sometimes also referred to as 'Medlar bodies', 'copper-penny bodies', 'sclerotic bodies', or 'fumagoid bodies'. These muriform bodies are the fungal elements which display an isotropic pattern of growth i.e., growth in all the 3 dimensions.[1,2,5,8] The golden-brown colour is due to the presence of fungal melanin, which may be of diagnostic utility, as fungal-melanin can be demonstrated by Masson-Fontana stain.[9]

Chromoblastomycosis is closely related to Phaeohyphomycosis which is also caused by demetiaceous fungi, however, the latter does not show the presence of muriform bodies, and instead shows hyphal elements.[3] Haematoxylin & Eosin staining in histopathology usually suffices, but a Periodic Acid Schiff stain may be useful at times.[1,2,5] A study conducted by Sateesh S. Chavan et al compared the utilities of unstained & destained histpathology sections vs. special stains like Periodic Acid Schiff retrospectively in 6 cases of Chromoblastomycosis. They found that the unstained and destained slides could serve as an acceptable alternative to Periodic Acid Schiff.[10]

Culture of the specimen in Saborauds Dextrose Agar can be done, along with microscopy of the growth using Lactophenol cotton blue stain for typing the causative species.[5] However, this does not have much bearing on the treatment once the diagnosis of Chromoblastomycosis has already been made.

Treatment usually a combination of surgical excision combined with high doses monotherapy or combination of oral Itraconazole, Terbinafine for 6-12 month period. Good results have also been obtained using Potassium Iodide in Indian patients. A follow-up of 2 years is prudent to ensure complete remission of the disease.[2,5,7]

CONCLUSIONS

Identification of Chromoblastomycosis in a non-endemic area is a challenge. High index of suspicion should be exercised while dealing with chronic skin lesions refractory to treatment,

so that a histopathological examination can be done to specifically look for pathognomonic findings. Eary diagnosis is essential to adequately treat the patient and to avoid complications.

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