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Case Report

Pathology Section

Chromoblastomycosis in the Gluteal Region: A Case Report

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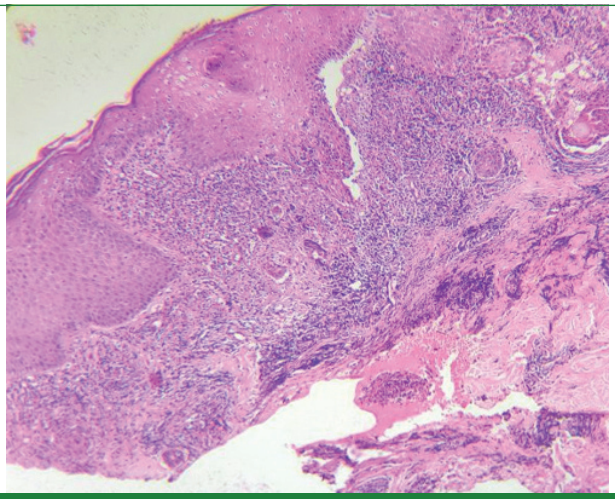
ABSTRACT

Chromoblastomycosis manifests as a persistent, gradual fungal infection affecting the skin and subcutaneous tissues, attributable to various dematiaceous fungal species. It predominantly afflicts rural labourers in tropical and subtropical regions, with its progression typically slow-paced. The lower limbs and hands are commonly affected. The condition manifests with lesions that are ulcerated, crusted, verrucoid, flat, or raised on the skin surface. Medlar first described the characteristic sclerotic bodies. The present case reported a 32-year-old male presenting with an indurated plaque on the left buttock. The diagnosis was confirmed by visualising pathognomonic sclerotic bodies on histopathological and microbiological examination. Fungal culture yields dematiaceous fungi with Fonsecaea type of sporulation suggestive of chromoblastomycosis in an unusual anatomical area. The patient was treated with itraconazole 200 mg once daily for three months and was relieved of the lesion.

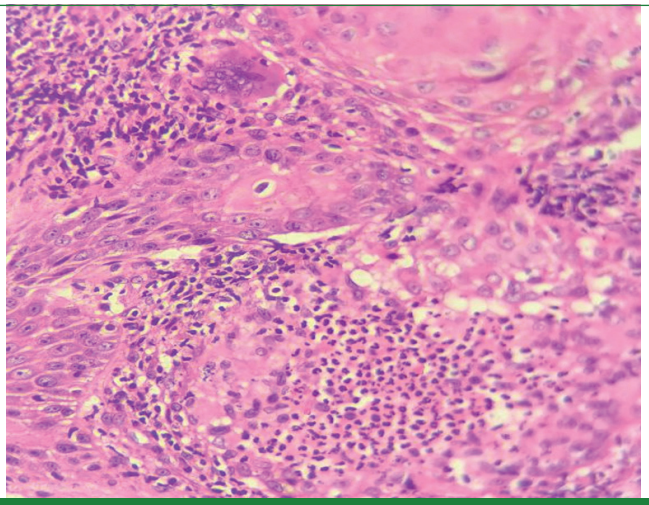
Keywords: Medlar bodies, Microabscess, Pseudoepitheliomatous hyperplasia

CASE REPORT

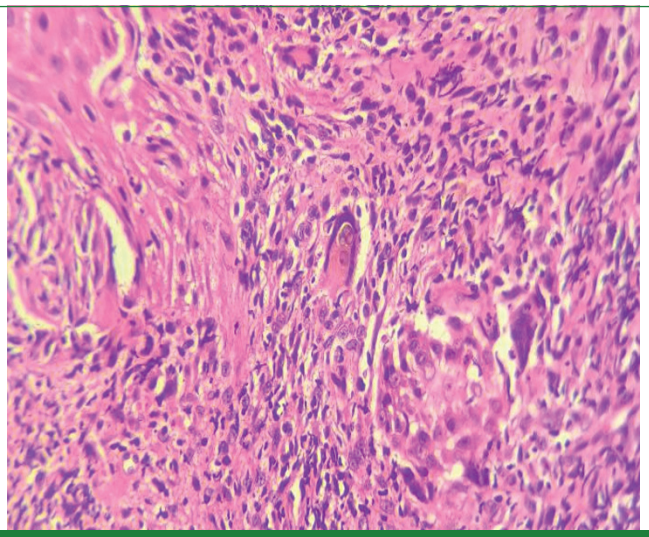
A 32-year-old male patient presented to the Dermatology Outpatient Department (OPD) with a single asymptomatic indurated plaque measuring 3×2 cm on the left buttock for six months. He developed it as a non tender nodule with a history of on-and-off crusting. Initially, he used medication such as mupirocin ointment for local application, but it did not heal. He works at a fish market, does not remember any trauma history but gave a history of sitting in the paddy fields while playing cricket. On clinical examination, there was a single non tender, indurated, and crusted 3×2 cm lesion on the medial aspect of the left buttock. During physical examination, there was no significant regional lymphadenopathy. His blood investigations, such as complete blood count, erythrocyte sedimentation rate, and liver function tests, were all within normal limits. The punch biopsy from the site of the lesion was taken and sent for histopathological examination, revealing pseudoepitheliomatous hyperplasia of the epidermis with an intraepidermal abscess [Table/Fig-1] and suppuration. The dermis showed inflammatory infiltrates composed of lymphocytes, neutrophils and multinucleated giant cells [Table/Fig-2]. The diagnostic round-to-oval golden-brown structures, suggestive of copper penny bodies, were observed singly, as well as, in clusters [Table/Fig-3]. As the pigmented copper penny bodies were easily appreciable, a diagnosis of chromoblastomycosis was made.



[Table/Fig-1]: Peudoepitheliomatous hyperplasia of epidermis (H&E, 10x).



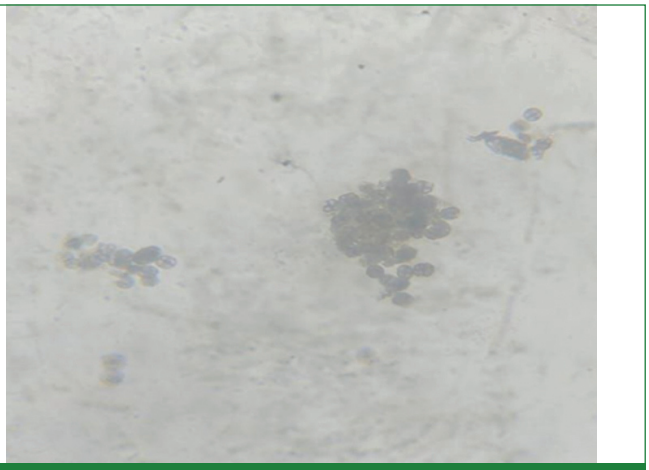
[Table/Fig-2]: Dermal microabcess with multinucleated giant cells (H&E, 40x).



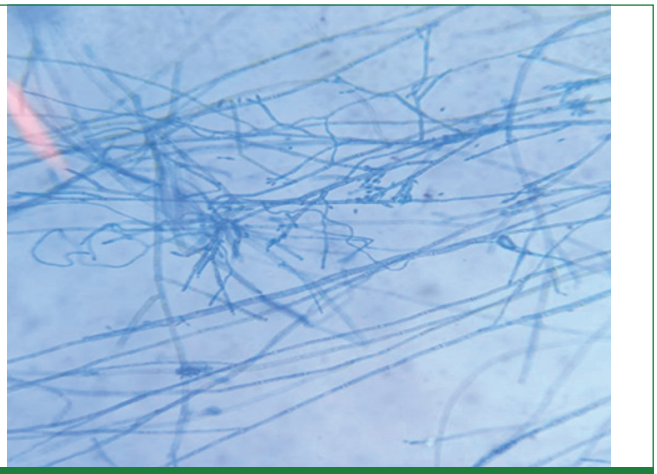
[Table/Fig-3]: Medlar bodies with copper penny appearance (H&E, 40x).

The microbiological examination by fungal scraping study using 10% potassium hydroxide showed sclerotic bodies [Table/Fig-4]. On fungal culture, the Lactophenol cotton blue mount of the colony yielded dematiaceous fungi with Fonsecaea type of sporulation,

suggestive of chromoblastomycosis [Table/Fig-5]. The patient was treated with itraconazole 200 mg once daily for three months and was relieved of the lesion.



[Table/Fig-4]: KOH mount with sclerotic bodies.



[Table/Fig-5]: Lactophenol cotton blue mount colony showing fonsecaea type of sporulation.

DISCUSSION

Chromoblastomycosis affects the skin and underlying tissues, stemming from certain types of fungi belonging to genera such as Cladophialophora, Rhinocladiella, Fonsecaea and Phialophora. *Fonsecaea pedrosoi* stands out as the most prevalent among these fungi, responsible for initiating the infection [1,2]. Cutaneous chromoblastomycosis was first described in 1914 in Brazil by Max Rudolph, a German physician. The fungi are usually found in soil, wood and rotting vegetables, and infection often results from trauma such as puncture from a splinter of wood [3].

The primary lesion develops as a result of traumatic implantation of the fungus into the skin. The lesion starts as a scaly papule, expands into a verrucous nodule or plaque, which has a predilection for the lower legs followed by the upper limbs [4]. The face, breast, toenail, axillae, genitals, pleural cavity, ileocaecal region, laryngotracheal area and tonsils are rarely affected. The illness extends to the surrounding skin, resulting in satellite lesions through autoinoculation, direct transmission, haematogenous dissemination, via lymphatic pathways, and seldom metastasises to distant organs [5]. The usual histopathological observations include pronounced pseudoepitheliomatous hyperplasia with occasional formation of microabscesses. The dermis shows granulomatous infiltrates featuring epithelioid cells, multinucleate giant cells, histiocytes and lymphocytes along with the identification of copper penny bodies [6].

Medlar, in 1915, described the characteristic histologic appearance of sclerotic bodies. They are brown round to polyhedral, thick-walled structures ranging from 4-12 microns in diameter. These forms replicate through septation, triggering a purulent and granulomatous inflammatory reaction in tissue [7]. Sclerotic bodies should be distinguished from haemosiderin and formalin pigment in tissue sections. The artificial pigments are irregular dense brown-coloured and do not show characteristic division, whereas sclerotic bodies have a definite morphology with intracellular division [8]. In the present case, histopathological and microbiological examination confirms the presence of sclerotic bodies, hence a diagnosis of chromoblastomycosis was made.

Torres E et al., explained a case of chromoblastomycosis in a 72-year-old agriculture worker affecting the lumbosacral region, groin, buttocks and perineum after an injury to the area. Appropriate treatment was delayed for a long period in this case and resulted in the development of squamous cell carcinoma [9]. Veerpandiyan A et al., reported another case of gluteal chromoblastomycosis in a 49-year-old male patient with a history of swelling in both buttocks for five months. He had a history of frequent gluteal intramuscular injections for Rheumatoid Arthritis (RA). Histopathological examination revealed fungal sclerotic bodies consistent with chromoblastomycosis [10]. Liu S et al., reported 11 cases of chromoblastomycosis from eastern China. Similar to the present case, most of the patients could not recall a history of trauma. There was a case of a severe verrucous lesion in the buttock of a patient who was on itraconazole 200 mg for 12 months [11].

The present case patient was treated with itraconazole 200 mg once daily for three months and was relieved of the lesion. From a clinical perspective, the plaque form of cutaneous chromoblastomycosis can bear similarities to cutaneous tuberculosis, sarcoidosis, phaeohyphomycosis and warts. Histopathological differentials include cutaneous leishmaniasis, tuberculosis, leprosy, sporotrichiosis, blastomycosis, tertiary syphilis, psoariasis and mycetoma is based on the histological identification, by KOH examination, and by fungal culture [2,8,12]. The approach for treating chromoblastomycosis involves chemotherapy, physical treatment and combination therapy. itraconazole and terbinafine have demonstrated the greatest efficacy since they act synergistically. Particularly, itraconazole tends to yield better outcomes when the responsible organism is *Cladosporium carrionii*. Flucytosine by itself or alongside amphotericin B could also prove effective. Additional alternatives encompass Terbinafine at a dosage of 250 mg per day, as well as Thiabendazole. Antifungal therapy should be continued until complete clinical resolution. Potassium iodide seems to be another effective drug. Surgery is recommended for tiny lesions, and even in these cases, it should be supplemented with chemotherapy. Alternative physical treatment methods such as cryotherapy, radiation, local thermotherapy, laser therapy and electrosurgery offer the benefit of shorter therapy duration and are also cost-effective. In susceptible individuals, efforts to prevent infection should focus on minimising traumatic transcutaneous inoculation in the environment. There are reports of failure of medical treatment, recurrences, and the potential for squamous cell carcinoma in affected regions [13,14].

CONCLUSION(S)

The authors reported the present case to highlight the rarity of localisation of chromoblastomycosis to an unusual anatomical area like the gluteal region. Chromoblastomycosis warrants consideration in the differential diagnosis when assessing chronic skin lesions, especially in individuals residing in tropical and subtropical areas. Early diagnosis and treatment can prevent permanent deformity. Chromoblastomycosis evolves slowly, seldom results in fatalities, generally has a good prognosis, but is a therapeutic challenge. The index case underscores the importance of raising awareness about this condition and fostering effective communication between clinicians and pathologists.

REFERENCES

[1] Chandran V, Sadanandan SM, Sobhanakumari K. Chromoblastomycosis in Kerala, India. Indian J Dermatol Venereol Leprol. 2012;78(6):728-33.

[2] Shanthala GB, Rudresh SM, Nagarathnamma T. Chromoblastomycosis: A case report. J Clin Diagn Res. 2011:5(4):865-66.
[3] Ramraje SN, Gokhale J, Gupta S. Cutaneous chromoblastomycosis. JCR. 2013;3(2):286-90.
[4] Abraham TS, Manjunatha P. Case report- A case of chromoblastomycosis effectively treated with oral Terbinafine. Int J Heal Sci Res. 2022;12(6):270-73.
[5] Verma GK, Verma S, Singh G, Shanker V, Tegta GR, Minhas S, et al. A case of extensive chromoblastomycosis from North India. Braz J Microbiol. 2014;45(1):275-77.
[6] Pradeepkumar NS, Joseph NM. Chromoblastomycosis caused by Cladophialophora carrionii in a child from India. J Infect Dev Ctries. 2011;5(7):556-60.
[7] Bhagwat PV, Tophakhane RS, Kudligi C, Noronha T. Multiple asymptomatic verrucous plaques over the legs. Indian J Dermatol Venereol Leprol. 2010;76(1):86.
[8] Baker JS, Miranpuri S. Perniosis: A case report with literature review. J Am Podiatr Med Assoc. 2016;106(2):138-40.
[9] Torres E, Beristain JG, Lievanos Z, Arenas R. Chromoblastomycosis associated with a lethal squamous cell carcinoma. An Bras Dermatol. 2010;85(2):267-70.
[10] Veerpandiyan A, Sekaran BS, Rahamathullah HM, Veerapandiyan A. A rare etiology for injection related gluteal abscess. J Infect Dis Epidemiol. 2020;6(6):01-02.
[11] Liu S, Zhi H, Shen H, Lv W, Sang B, Li Q, et al. Chromoblastomycosis: A case series from Eastern China. PLoS Negl Trop Dis [Internet]. 2022;16(9):01-11.
[12] Bandyopadhyay A, Majumdar K, Gangopadhyay M, Banerjee S. Cutaneous chromoblastomycosis mimicking tuberculosis verrucosa cutis: Look for copper pennies! Turk Patoloji Derg. 2015;31(3):223-25.
[13] Deb Roy A, Das D, Deka M. Chromoblastomycosis-A clinical mimic of squamous carcinoma. Australas Med J. 2013;6(9):458-60.
[14] De A, Gharami RC, Datta PK. Verrucous plaque on the face: What is your diagnosis? Dermatol Online J. 2010;16(1):6.

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