Quiz

Correct answer to the quiz. Check your diagnosis

Chromoblastomycosis of the leg

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Case report: We herein report a case of chromoblastomycosis presenting as a verrucous lesion over the leg. A 56-year-old male patient was a known case of carcinoma larynx and was treated for the same. At presentation to our hospital, the patient, in addition to the recurrent local disease, was suspected to have second primary in the form of verrucous carcinoma of the leg. Histopathological examination of the skin biopsy revealed the presence of characteristic pigmented sclerotic bodies with pseudoepitheliomatous hyperplasia of the overlying epithelium. The case was reported as chromoblastomycosis and the patient responded well to anti-fungal chemotherapy in the form of itraconazole.

Key words: chromoblastomycosis, squamous carcinoma, medlar bodies.

Introduction

Chromoblastomycosis (CBM) is chronic fungal infection of skin and subcutaneous tissue caused by dematiaceous fungi [1]. The disease might present as a slowly developing verrucous cutaneous lesion, which clinically mimics squamous carcinoma [2]. We report a case of chromoblastomycosis in a 56-year-old male in a known case of squamous carcinoma of larynx.

Case report

A 56-year-old unemployed male patient was a known case of squamous carcinoma of the larynx, diagnosed and treated with radiotherapy in 2015 outside our institute. The patient was referred to our institute for further management. On follow-up examination this year, the patient was diagnosed with recurrence of the laryngeal disease. In addition, clinical examination revealed multiple verrucous ulcerative lesions over the right calf, the largest measuring 10 × 10 cm. The patient on probing said that these ulcers had been present for 10 years but had increased in size during the last year. The patient was suspected to have a second malignancy (verrucous carcinoma), and in view of the same a punch biopsy sample was sent for histopathological confirmation of clinical diagnosis of squamous carcinoma.

Section examined showed irregular acanthosis, pseudoepitheliomatous hyperplasia, hyperkeratosis, and parakeratosis of the overlying epidermis (Fig. 1). Upper dermis showed microabscesses, surrounded by granulomas, comprising Langhans giant cells, plasma cells, and eosinophils (Fig. 2-3). Characteristic pigmented sclerotic bodies (copper pennies) were noted within the giant cells and also within the centre of microabscesses (Fig. 3). These fungal elements were also highlighted on Grocott's methenamine silver (GMS) (Fig. 4) and periodic acid-Schiff (PAS) stains. Diagnosis of CBM was offered based on the characteristic histomorphological findings aided by ancillary techniques, and culture studies were advised.

Discussion

Chromoblastomycosis is a chronic cutaneous and subcutaneous fungal infection caused by dematiaceous fungus of order *Chaetothyriales* [3]. The char-

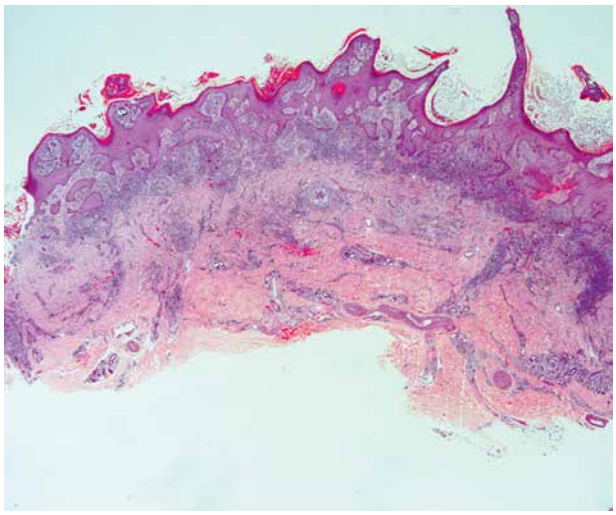

Fig. 1. Acanthosis, pseudoepitheliomatous hyperplasia, hyperkeratosis, and parakeratosis of the overlying epidermis (HE, objective magnification 1×)

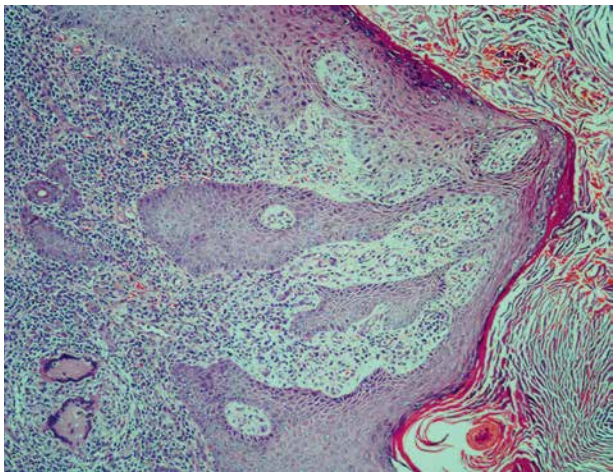

Fig. 2. Dermis showing microabscesses with granulomas comprising of Langerhans giant cells (HE, objective magnification 10×)

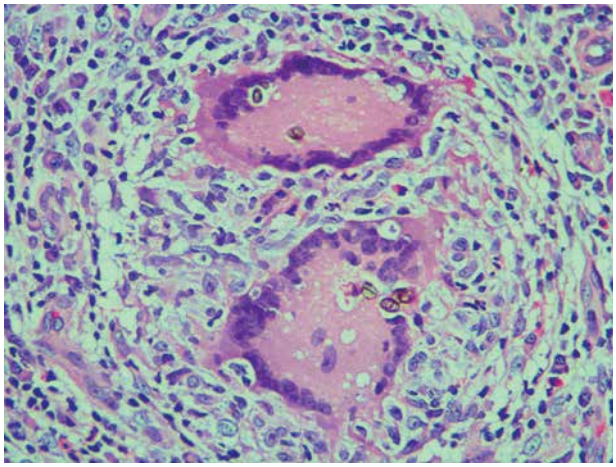

Fig. 3. Granulomas comprising of Langerhans giant cells, plasma cells, and eosinophils and characteristic sclerotic bodies (copper pennies) with transverse and longitudinal septations (HE, objective magnification 40×)

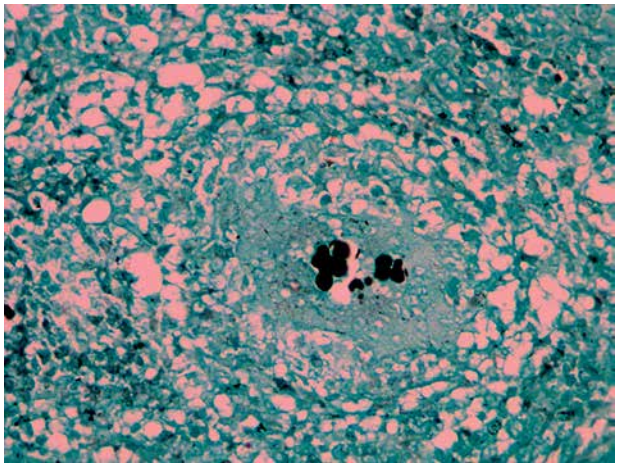

Fig. 4. Grocott's methenamine silver (GMS) stain highlighting fungal elements (objective magnification 40×)

acteristic histopathological hallmark for diagnosing chromoblastomycosis is the presence of pigmented, muriform, thick-walled cells called sclerotic cells/medlar bodies or copper pennies [3, 4].

The disease is most prevalent in humid tropical and subtropical regions of Africa, Asia, and America. It was first described by Alexandre Pedroso in 1911, and he consequently published his findings in 1920 [5]. The most frequent aetiological agents, namely *Fonsecaea pedrosoi*, *Phialophora verrucosa* and *Cladophialophora carrionii* belong to the *Chaetothyriales* order of fungi. The disease is frequently found in males (male-to-female ratio 4 : 1) engaged in agricultural activities [6]. The part of the plants affected by the fungi is responsible for the direct implantation of these agents at the site of the injury [7]. Consequently, the extremity is the most common site to be affected.

CBM presents clinically in one of five forms: nodular, tumoural, verrucous, plaque and cicatricial [3]. Furthermore, these lesions can be classified according to the severity [8]. Clinically, the tumoural and verrucous forms can be readily confused with cancer. There have been isolated case reports in the literature about chromoblastomycosis mimicking squamous carcinoma [2, 9]. The patient in our report was a known and treated case of carcinoma larynx. Although he had history of leg ulcers on lower limbs for 10 years, few of the ulcers along with the largest verrucous lesion had increased in size during the preceding year. Keeping in mind the history of cancer in the past, the second malignancy was the first differential diagnosis from the clinical side, and the patient was biopsied for the same.

Biopsy is often performed to confirm the diagnosis [8]. Histopathological examination shows epidermal

hyperkeratosis and pseudoepitheliomatous hyperplasia. Under the microscope pseudoepitheliomatous hyperplasia can be easily mistaken for verrucous carcinoma, especially when seen at low magnification power, especially when the patient is biopsied with clinical impression of squamous carcinoma, as in our case. This necessitates a high index of suspicion and careful microscopic evaluation at high magnification power to clinch the accurate diagnosis. Inflammatory response in most cases is in the form of granulomatous inflammation comprising of lymphocytes, histiocytes, epithelioid cells, and Langhans-type multinucleated giant cells. The characteristic pigmented sclerotic/medlar bodies can be found in the layers of the skin, granulomatous process, or even in the giant cells. The muriform cells or medlar bodies are formed from fungi that divide by septation. They are 5-12 μm chestnut-shaped, thick-walled, pigmented structures with both transverse and longitudinal septations. They are readily observed under haematoxylin-eosin-stained section, and hence special staining is not mandatory. Further, fresh tissue can be subjected to microbiological culture on Sabouraud-agar or alternatively for molecular studies using duplex PCR targeting the ribosomal DNA of *Fonsecaea* spp. [10]. Biopsy in our case had characteristic histomorphological findings. Ours being a cancer institute, the patient was referred to another tertiary care centre, where the biopsy was reviewed by dermatologists and the patient was advised to take itraconazole and local $KMnO_4$ soaks. The patient is responding well to the antifungal treatment. In addition, he was offered supportive care for the recurrent laryngeal disease. Unfortunately, the patient succumbed to death by virtue of his recurrent disease five months later.

Apart from antifungal chemotherapy, other treatment modalities include physical methods like surgical excision, cryosurgery, thermotherapy, and laser vaporisation [8]. The choice of treatment may vary according to the severity of disease, type of disease, and availability of treatment facilities. In many cases combination therapy consisting of physical treatment and chemotherapy is used. In spite of these varied treatment options, CBM remains a therapeutic challenge for treating physicians owing to its intractable nature.

To conclude, although rare, CBM should be considered in the differential diagnosis of verrucous skin ulcers, especially for long-standing lesions in tropical and subtropical areas. Our case report also highlights the importance of identifying this entity correctly when the differential includes squamous carcinoma because the treatment options for both are poles apart.

The authors declare no conflict of interest.

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