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Case Report

A giant mass on the left shin: report of a case

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Abstract: A woman presented to our department with an asymptomatic progressive peripherally expanding skin lesion on her left shin for 6 years. The doctor in a local clinic considered it was a deep fungal infection, however, treatment with itraconazole systemically was ineffective. A second biopsy showed papillomatous hyperplasia and a central keratin-filled crater with a buttress like extension of surrounding epidermis and a well-demarcated regular base. In the upper dermis, diffused infiltration of mixed inflammatory cells was observed with perivascular infiltrate. Deeper dermis, appendageal structures and subcutaneous tissue were unremarkable. Periodic acid-schiffic staining and diamine silver staining of the specimen were negative. A diagnosisof Keratoacanthoma centrifugum marginatum (KCM) was finally established.

Keywords: Mass, leg ulcer, cancer, dermatopathology, keratoacanthoma centrifugum marginatum, acitretin

Introduction

Keratoacanthoma (KA) is a rapidly growing tumor with a well-defined cycle of growth eventually ending in spontaneous involution [1, 2].

There are several types of KA described in the literature, but solitary KA is the most common type with its unique stage: proliferation, maturation and spontaneous [3]. Keratoacanthoma centrifugum marginatum (KCM) is a rare variant of KA. It is characterized by progressive peripheral expansion and concomitant central healing leaving regional atrophy. It is sometimes confused with squamous cell carcinoma (SCC) both clinically and histopathologically.

Case report

An 81-year-old female presented with an asymptomatic but progressive peripherally expanding skin lesion on her left shin for 6 years (**Figure 1**). She denied a history of prior trauma. The lesion had developed from a pea-sized keratotic nodule that gradually enlarged to 2/3 of her left shin. There were no similar family histories. Physical examination revealed the presence of a large mono-annular lump with marginal protusion and central atrophy. A part of the lesion was ulcerated. On palpation the lesion was firm with purulent secreted. No lymphadenectasis or any systemic problems were found. Serological test for HIV and TPHA were negative. PPD skin test, chest and tibiofibula X-ray results were normal. Bacterial culture of the lesion identified proteus mirabilis. Fungus culture of the lesion was negative. The first biopsy revealed hyperkeratosis, parakeratosis and irregular acanthosis in the epidermis. Meanwhile, there were some keratinous cysts, prickle cells in active proliferation. Periodic acid-schiffic staining and diamine silver staining were negative. According to the clinical and pathological features, the doctor in a local clinic considered the diagnosis as a deep fungal infection and administered with itraconazole (0.1 g qd), but there was no improvement after a month.

The key clinical feature was a circular or angular huge mass, extension with raise, rolled border and atrophy in the center, and a part of the lesion was ulcerated. There are some diseases needed to be differentiated, include Blastomycosis-like pyoderma, deep mycosis, tuberculosis infection, Squamous cell carcinoma, Keratoacanthoma centrifugum marginatum.

Blastomycosis-like pyoderma

It is a rare chronic tissue proliferative reactive pyoderma which is potentially caused by bacte-

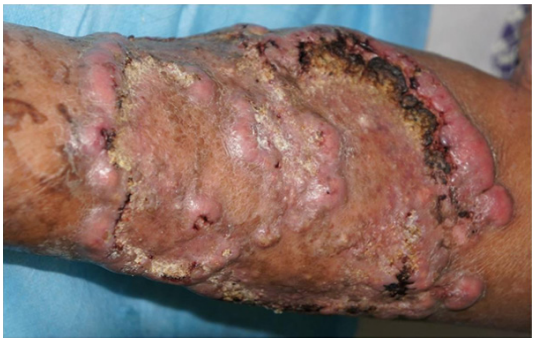

Figure 1. A mono-annular huge lump, with marginal rise and central atrophy on the left shin.

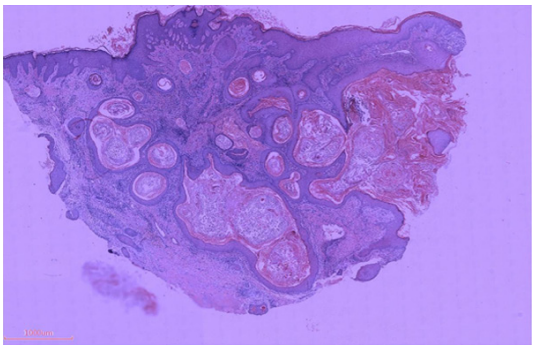

Figure 2. Papillomatous hyperplasia, the central keratin-filled crater with buttress like extension of surrounding epidermis, and well demarcated regular base (HE, original magnification × 100).

rial infection. Most patients have a compromised immune systemically either locally or system wide. Typical lesions are large, indurated and verrucous plaques with elevated borders and multiple pustules. The lesion features are in accordance with our case. Bacterial culture of lesion is proteus mirabilis. Antibiotic therapy is useless for our patient, but some cases who suffered from blastomycosis-like pyoderma response well to acitretin, rather than antibiotics. The diagnosis maybe blastomycosis-like pyoderma, but it is necessary to take another biopsy to confirm the diagnosid.

Deep mycosis and tuberculosis infection

The patient denied any history of trauma and contact with TB patient. The X-ray of chest is normal, PPD skin test is negative. The previous biopsy results of PAS and acid-fast staining were negative. Fungal culture of the lesion is negative with itraconazole was ineffective. So we can partly exclude these two diagnoses.

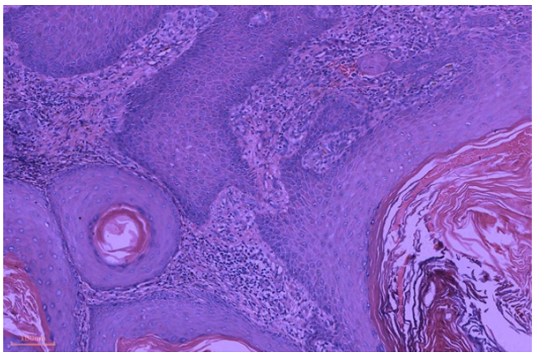

Figure 3. Diffused and mixture inflammatory cells perivascular infiltrated in the upper dermis (HE, original magnification × 400).

Squamous cell carcinoma

It is a malignant tumor and more prevalent among the people who over 50 years old. The lesion maybe superficial, rigid and may arise from an indurated, rounded, elevated base at the beginning. In a few months, the lesion may become larger, deeply nodular and ulcerated. Our patient is a 81 year-old female peasant and she has regular sun exposure. So there are two risks factors. The histopathology is characterized by irregular epidermal cell nests invading the dermis. The typical lesion of this disease is different from our patient. So we also can partly excluded this diagnosis.

Keratoacanthoma centrifugum marginatum

The typical lesion lesion is similar to this case. It has no tendency for spontaneous regression. This point can help distinguish KCM from keratoacanthoma. So the diagnosis should be considerate.

Histopathological examination revealed that papillomatous hyperplasia and a central keratin-filled crater with buttress like extension of surrounding epidermis and a well-demarcated regular base (**Figure 2**). In the upper dermis diffuse and mixed inflammatory cells was observed with perivascular infiltrate (**Figure 3**). Deeper dermis, appendegeal structures and subcutaneous tissue were unremrkable. Periodic acid-Schiff and diamine silver staining of the specimen was negative.

Based on the architecture and inflammation at the base of the lesion, the diagnosis of KCM was suggestive. After a four-day antibacterial

therapy and debridement, together with a one-month treatment with acitretin 20 mg/d, the lesion became smaller gradually. At last, the patient was lost to follow up.

Discussion

KCM is an extremely rare type of keratoacanthoma (KA). It was first described by Belisario as a separate entity [4]. Clinically, KCM is characterized by gradual peripheral expansion, a raised rolled-out margin and atrophy at the centre [5]. The lesion can be localized to any region of body but is more often found on chronic sun exposed areas like the dorsum of the hands and legs. The lesion may reach up to 20 cm in diameter with no tendency to spontaneous involution, as was seen in this case. The etiology is still uncertain. Some scholars consider it to be a form of regressing squamous cell carcinoma. Meanwhile, others believe sun exposure, in conjunction with chemical carcinogens, and trauma play important roles in promoting the disease.

KCM has no tendency for spontaneous regression. Treatments include systemically administered retinoids (acitretin, etretin or isotretinoin) and should be maintained until complete clearance of the lesion [6, 7]. In addtion, topical 5-fluorouracil or imiquimod cream, intralesional injection of interferon alpha, methotrexate, Er: YAG laser, and surgical excision are also of choice to treat the disease. Surgical excision for KCM is often desirable. If the lesion grows on the face, Mohs surgery allows for good margin control with minimal tissue removal [8].

In this patient, antibiotics or antifungal therapy was ineffective, while the continuous therapy may cause side effects and delayed resolution of the illness. Glucocorticoid treatment is inappropriate until an infectious etiology can be ruled out. Surgical excision is not recommended for such a large mass before the diagnosis established. Therefore, the first procedure recommended for this patient is skin biopsy. Once the diagnosis is confirmed, we can give the patient appropriate treatment.

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Disclosure of conflict of interest

None.

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