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ORIGINAL ARTICLE

# Cutaneous basidiobolomycosis: Seven cases in southern Benin

## *Basidiobolomycose cutanée : sept cas observés dans le sud du Bénin*

suppl. Informations

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**KEYWORDS**
Cutaneous basidiobolomycosis;
*Basidiobolus ranarum*;
Child;
Benin

**Summary**
*Background.* — Cutaneous basidiobolomycosis is the most common form of entomophthoramycosis. Herein we report seven cases of cutaneous basidiobolomycosis.
*Patients and methods.* — A retrospective observational study was conducted at the Buruli ulcer treatment centre in Pobè and at the national teaching hospital in Cotonou from 2010 to 2015.
*Results.* — Seven cases of cutaneous basidiobolomycosis were diagnosed. The mean patient age was 9.53 years. There were 4 female and 3 male patients, all from southeast Benin. Clinically, the disease presented in all cases as a hard, well-defined, subcutaneous plaque with little inflammation, and which could easily be lifted from the deep structures but remained attached to the surface structures. The overlying skin was hyperpigmented. Plaques were localized to the buttocks or thighs. All patients had inflammatory anaemia with an accelerated

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erythrocyte sedimentation rate (30 to 70 mm over the first hour), and a low haemoglobin count (8.7 to 11.4 g/dL). Blood hypereosinophilia (650 to 3784 elements/mm$^3$) was present in six of the seven subjects. Histopathology (performed for 5 of the 7 subjects) showed granulomatous lesions with foreign-body giant cells, and inflammatory cells, with occasional eosinophils surrounding fungal hyphae (Splendore-Hoeppli phenomenon). Mycological analysis revealed *Basidiobolus ranarum* in three cases. The patients were treated with ketoconazole (5/7) and itraconazole (2/7), with good outcomes after 10 to 24 weeks of therapy.
*Discussion.* — Cutaneous basidiobolomycosis is uncommon in southern Benin, with only seven cases being diagnosed over 6 years. The diagnosis of cutaneous basidiobolomycosis is a challenge in the field in Benin due to the non-specific clinical presentation, the lack of technical resources, and the existence of numerous differential diagnoses.
*Conclusion.* — Cutaneous basidiobolomycosis is an uncommon fungal infection in southern Benin chiefly affecting children.


**MOTS CLÉS**
Basidiobolomycose cutanée ;
*Basidiobolus ranarum* ;
Enfants ;
Bénin

**Résumé**
*Introduction.* — La basidiobolomycose cutanée est la plus fréquente des entomophtoromycoses. Nous en rapportons sept cas observés dans le sud du Bénin et discutons leurs particularités cliniques et épidémiologiques.
*Patients et méthodes.* — Une étude transversale rétrospective a porté sur les cas de basidiobolomycose cutanée observés en 6 ans dans le sud du Bénin et dont le diagnostic a été confirmé par l'examen histopathologique et/ou l'examen mycologique.
*Résultats.* — L'âge moyen des patients était de 9 ans et demi. Quatre sur sept étaient de sexe féminin. L'aspect clinique était un placard dermo-hypodermique peu inflammatoire, ferme, mobile par rapport au plan profond et fixé par rapport au plan superficiel dans tous les cas. Il siégeait aux membres inférieurs chez tous les patients. Dans tous les cas il existait un syndrome inflammatoire avec vitesse de sédimentation augmentée (30 à 70 mm à la première heure), taux d'hémoglobine bas (8,7 à 11,4 g/dL) et hyperéosinophilie sanguine (650 à 3784 éléments/mm$^3$). L'examen histopathologique, réalisé dans 5 cas sur 7, montrait un granulome épithélioïde et giganto-cellulaire avec parfois un phénomène de Splendore et Hoeppli (manchon de polynucléaires éosinophiles autour des filaments mycéliens, sans envahissement vasculaire). Les patients étaient traités par kétoconazole (5 cas sur 7) ou itraconazole (2 cas sur 7), avec amélioration clinique des lésions en 18 à 24 semaines.
*Discussion.* — Sept cas de basidiobolomycose cutanée ont été diagnostiqués en 6 ans, témoignant de la rareté de cette affection. Le diagnostic de basidiobolomycose cutanée n'est pas aisé dans nos conditions d'exercice car la présentation clinique n'est pas spécifique, le plateau technique est peu équipé et les diagnostics différentiels sont nombreux.
*Conclusion.* — Les basidiobolomycoses cutanées sont des affections rares au sud du Bénin, touchant principalement les enfants.


Cutaneous basidiobolomycosis is a form of entomophthoramycosis endemic in tropical and subtropical regions of Africa, Latin America and Asia [1]. It generally affects children and adolescents living in rural environments [2]. Herein, we report seven cases of the disease collated in Benin.

## Patients and methods

A retrospective observational study was conducted in cases of cutaneous basidiobolomycosis diagnosed between 2010 and 2015 at the Buruli ulcer screening and treatment centre in Pobè and in the Dermatology-Venerology Department of the Hubert Koutoukou Maga National University Hospital Centre in Cotonou in southern Benin. The diagnosis of cutaneous basidiobolomycosis was suspected on clinical examination and was confirmed by histopathology or mycological examination. The other laboratory tests included bone radiography, ultrasound, complete blood count, blood biochemistry, HIV serology and syphilis serology. Patients were treated with either ketoconazole (7 mg/kg/day) or itraconazole (5 to 10 mg/kg/day), with monthly monitoring. The sociodemographic variables (age, gender, profession,

geographical origin), clinical data (duration of the disease prior to consultation, type and localisation of lesions) and laboratory data, as well as therapeutic and disease progression data were collected. Free and informed consent to take part in this study was given by the patients or their legal guardians.

## Results

Seven cases of cutaneous basidiobolomycosis were observed over a 6-year period. The age of patients ranged from 17 months to 30 years; six patients were aged 8 years or less. Four of the seven patients were female and all patients were originally from the Plateau region situated in southeastern Benin ($n = 6$) or from Niger ($n = 1$). The patients comprised schoolchildren ($n = 3$), pre-school children ($n = 3$) and one farmer ($n = 1$). The duration of the disease prior to consultation ranged from 2 weeks to 12 months. One patient had allergic asthma and a 7-year-old girl was presenting functional deficit; no disease history was reported in the other patients.

In all seven cases, the lesions were single, consisting in all cases of a hard, painless subcutaneous plaque with little inflammation, and which could easily be lifted from the deep structures but remained attached to the surface structures. These plaques were well defined and curled fingers could easily be inserted under the plaque before lifting. The skin above was either hyperpigmented ($n = 5$) or normally pigmented ($n = 2$). The length of the plaques ranged from 5 cm to 28 cm. None of the lesions were ulcerated. They were located on the left thigh ($n = 3$), the right thigh ($n = 2$), the right buttock ($n = 2$) or the left buttock ($n = 1$). In one patient, the lesion was situated both on the right buttock and thigh (Figs. 1 and 2). Homolateral inguinal adenopathies ranging in diameter from 1.5 to 2 cm were present in 5 cases and absent in 2 others.

The laboratory abnormalities noted concerned and increased erythrocyte sedimentation rate, between 30 mm and 70 mm in the first hour, inflammatory anaemia with haemoglobin counts of 8.7 to 11.4 g/dL, and hypereosinophilia in 6 of the 7 patients, ranging from 650 to 3784 elements/mm$^3$. All patients were negative for HIV. Hepatic and renal function were normal. Mycological examination was performed in 3 of the 7 patients and *Basidiobolus ranarum* was isolated (Fig. 3). Skin biopsy was performed in 5 of the 7 patients and exhibited histopathological features characteristic of inflammatory epithelioid and giant-cell granuloma, in some cases with Splendore and Hoeppli phenomenon, i.e. eosinophils surrounding non-septal fungal hyphae without vascular invasion (Fig. 4). The inflammatory granuloma was either dermal ($n = 2$), or dermal and hypodermal ($n = 3$).

The patients were treated with ketoconazole ($n = 5$) at a dosage of 7 mg/kg/day for the children and of 400 mg/day for the adult, or with itraconazole ($n = 2$) at a dosage of 5 mg/kg/day for the children. The duration of treatment ranged from 10 to 24 weeks. The outcome was favourable, with a notable decrease in oedema and in other clinical signs. This clinical improvement in the lesions was noted after the first 8 weeks of treatment. Treatment

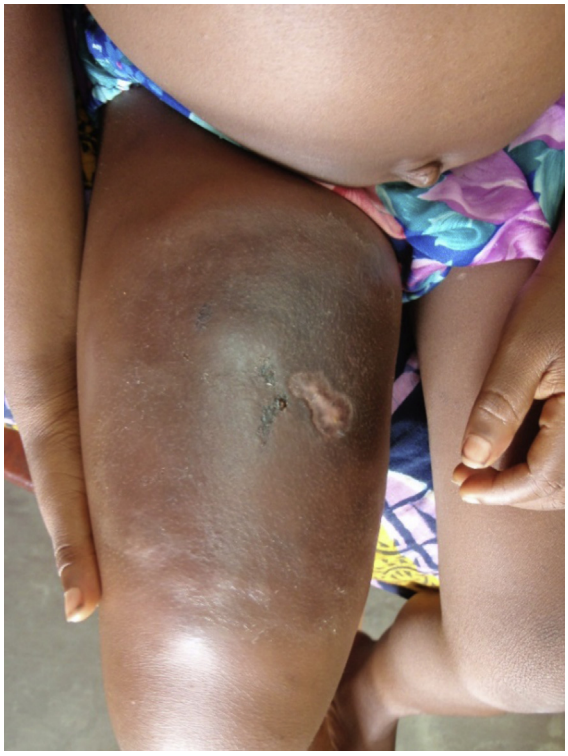

**Figure 1.** Cutaneous basidiobolomycosis of the right thigh in an 8-year-old girl : hard, hyperpigmented, painless plaque, which could be lifted from the deep structures but remained attached to the surface structures.

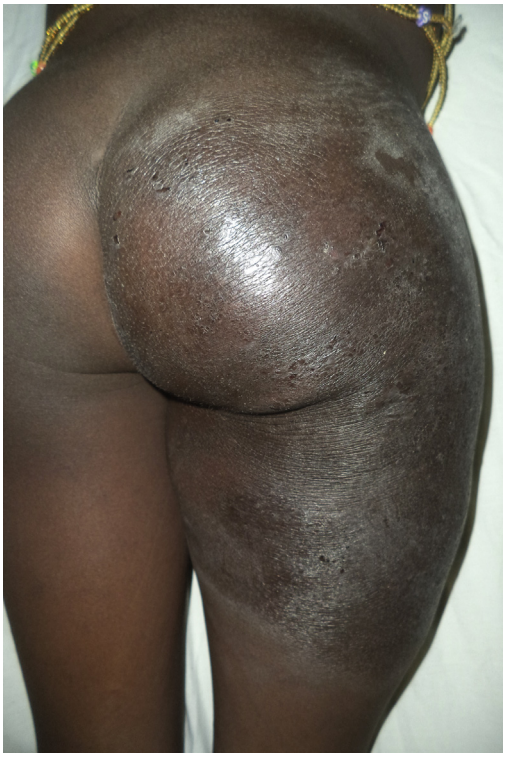

**Figure 2.** Basidiobolomycosis of the right thigh in a 6-year-old girl : the loss of substance corresponds to the area biopsied.

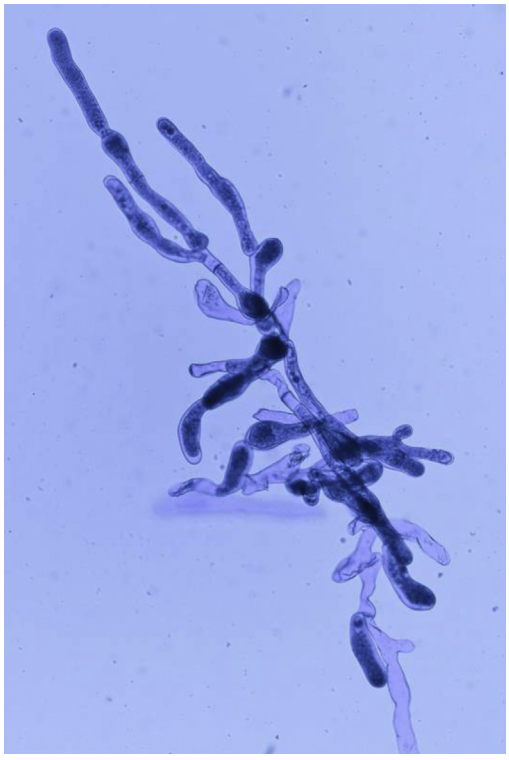

**Figure 3.** Appearance of *Basidiobolus ranarum* in culture, showing relatively non-septal hyphae and the characteristic beak-like appendages of zygospores.

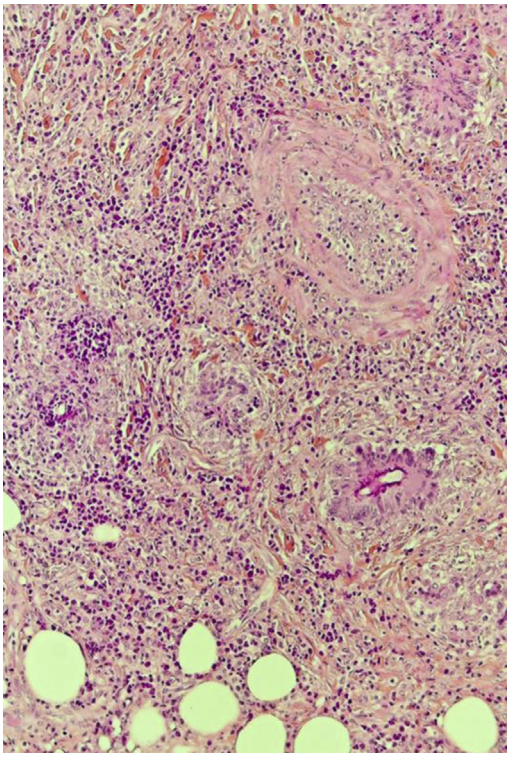

**Figure 4.** Histopathological appearance of cutaneous basidiobolomycosis showing a giant-cell inflammatory granuloma with Splendore-Hoepli phenomenon.

tolerability was good. None of the patients underwent surgery.

## Discussion

We noted 7 cases of cutaneous basidiobolomycosis diagnosed on the basis of histopathology and/or mycology. Our observations were noteworthy in terms of the geographical origin of the patients, the presence of satellite adenopathies, the existence of hypereosinophilia, and the efficacy of the drug therapy with azole antifungals.

Geographical origin was one of the factors studied in our series. The majority of the reported cases of cutaneous basidiobolomycosis concerned patients in tropical regions of Africa [1—5], Asia [6—11], Latin America and the United States, particularly Arizona [12]. An arid climate effectively appears to be propitious to the development of the fungus [12]. All of our patients were from the Plateau region in southeast Benin, where climate change has resulted in decreased rainfall and in the emergence of increasingly arid zones.

The clinical presentation of our cases was typical of the cases of cutaneous basidiobolomycosis published in the literature [1,2,5,10]. It nevertheless differed in terms of the presence, in 5 of the 7 patients, of unilateral satellite adenopathies on the side of the lesion. These adenopathies were hard, and either painless or sensitive to palpation. Adenopathies are reported by only a few authors, with Burkitt [5] and Krishnan [10] noticing this feature in some of their patients. Their significance is unclear. Only biopsy with mycological and histopathological examination could have confirmed whether or not there was specific involvement of *Basidiobolus ranarum*. However, the unilateral and homolateral nature of the lesions, as well as their decreased volume or complete regression under therapy, are potential arguments militating in favour of specific involvement.

The clinical presentation of cutaneous basidiobolomycosis is not very specific, making diagnosis somewhat difficult, and several other diseases may in fact mimic the clinical signs. Within the context of our study, the clinical presentation was evocative of the non-ulcerative form of Buruli ulcer, as a result of which 5 of our 7 cases were diagnosed at a screening and treatment centre for Buruli ulcer. Laboratory examinations were necessary for confirmation of the diagnosis.

The existence of hypereosinophilia in blood was also a noteworthy aspect of our cases. Although it was seen in 6 of the 7 patients in our study, Burkitt et al. [5] noted no cases of hypereosinophilia in their series of 31 patients. A corollary of this hypereosinophilia in blood is tissue hypereosinophilia. According to Sujatha et al. [13], eosinophilic infiltration of tissue is a result of type TH2 immune response with production of cytokines such as IL-4 and IL-10, enabling recruitment of eosinophils. However, interpretation of blood hypereosinophilia in tropical settings is not straightforward. Other forms of parasitic infestation, particularly intestinal, may be associated with such blood hypereosinophilia and systematic screening for the latter was not performed.

Treatment of cutaneous basidiobolomycosis is pharmaceutical and based on the use of azole antifungals or potassium iodide. The latter agent is very widely used by

Indian authors [14]; although the cost is very reasonable, but use of the latter drug exposes patients to dysthyroidism, and monitoring of thyroid function is thus necessary. Further, gastrointestinal and cutaneous tolerability are poor. Use of azole antifungals is consequently preferable, with the most commonly used agents being ketoconazole (7 mg/kg/day) and itraconazole (5 mg/kg/day). Both treatments proved efficacious in our patients. These drugs are available in Africa but their use requires monitoring of liver function.

## Conclusion

Cutaneous basidiobolomycosis is a rare form of mycosis in the south of Benin, and primarily affects children in rural settings without any clear predilection for gender. Treatment is drug-based and involves the use of azole antifungals.

## Disclosure of interest

The authors declare that they have no competing interest.

## Appendix A. Supplementary data

Supplementary data associated with this article can be found, in the online version, at http://dx.doi.org/10.1016/j.annder.2016.10.017.

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