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OCCURRENCE OF SUBCUTANEOUS ZYGOMYCOSIS CAUSED BY BASIDIOBOLUS HAPTOSPORUS IN BRAZIL

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Abstract

There were described the first three South American cases of subcutaneous zygomycosis caused by *B. haptosporus*. The patients were children from nearby towns lying just north of 13 ° latitude S. The diagnosis was based on histopathological aspects plus cultural isolation of the fungus.

Introduction

Subcutaneous zygomycosis are chronic and indolent infections caused by *Basidiobolus haptosporus* or by *Conidiobolus coronata*. The mycosis occurs in apparently healthy individuals, appearing as a well circunscribed indurated lesion involving the subcutaneous tissue. Histologically, a granulomatous reaction with heavy eosinophilic infiltration and many abscesses are present. Fungal elements are easily visualized in H.E. stained sections, because they are surrounded by a large eosinophilic halo (Splendore phenomenon). But the localization of the lesions is characteristic for each agent: *C. coronata* lesions are situated on the dorsum of the nose and superior lip, rarely extending to other areas of the face; *B. haptosporus* lesions are located in the thigh, buttocks and trunk.

In the New World, Bras (5) described the first case of *C. coronata* infection. Since then, five additional cases of this infection were detected in the American continent (1, 2, 3, 8, 9). Only one American case of *B. haptosporus* infection was reported (4). This case and two additional ones, all of them occurring in Bahia, Brazil, will be described in this paper.

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Case reports

Case 1

A 6-year-old girl from Sta. Terezinha came to the hospital because of a red lesion of three months duration on the left thigh. On examination, there was an indurated plaque with well-defined raised margins involving the lateral and anterior surface of the left thigh, part of the vulva, and the left groin (Fig. 1). The lesion was erythematous and warm, but non-tender. There was painless left inguinal lymphadenopathy. The remainder of the physical examination was normal. Hemoglobin, hematocrit, leucocyte count, blood glucose, and radiographs of the chest, thigh and pelvis were all within normal limits. A diagnosis of subcutaneous zygomycosis by *B. haptosporus* was accomplished on the basis of histopathologic study and cultural findings. Humoral antibodies (IgG and IgM) against fungal elements were detected by immunofluorescence. Biopsy of an enlarged inguinal lymph node showed reactive hyperplasia. The patient was treated with potassium iodide, 30 mg/per Kg. body weight/per day. After two months, the lesion had regressed remarkably, and

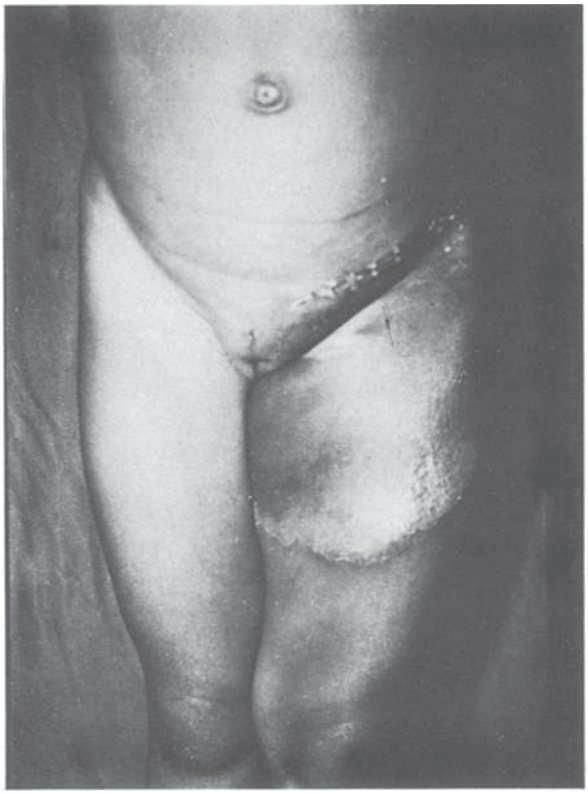


Fig. 1. Case 1. An indurated plaque involving the left thigh, groin, and part of the vulva.

the patient was discharged with instructions to continue the medication. At the time of re-examination, eight months later, the lesions had worsened and spread to the knee and buttock. Intradermal testing with PPD (2 TU), levedurin, histoplasmin, and phyto-themaglutinin and contact sensitization with dinitrochlorobenzene (DNCB) (7) gave negative results. Potassium iodide, at the same dose as earlier, produced no change in the lesions. However, after four months of treatment with 60mg of potassium iodide per Kg, daily, all lesions disappeared. At this time, the reaction to PPD was 2+ and to patch testing with DNCB 2+.

Case 2

A 4-year-old girl from Camaçari entered the hospital with a lesion of the thigh of 45 days' duration. She had recently undergone exploratory surgery of the thigh at an orthopedic hospital, because of a suspicion of osteomyelitis. On examination, there was generalized

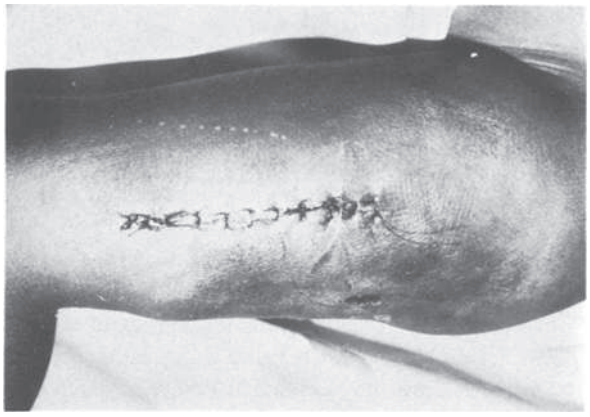


Fig. 2. Case 2. Enlargment of the left thigh with induration and raised well-defined margins. The wide suture and the underlying orifice were the result of an unnescessary surgery.

enlargement of the left thigh with induration, erythema, and warmth of the skin (Fig. 2). The remainder of the examination was unremarkable. Hemoglobin, hematocrit, and red cell count were normal, but there was a slight leucocytosis with neutrophilia, eosinophilia, and lymphopenia. Erythrocyte sedimentation rate was elevated. The blood sugar was normal, and serum immunoelectrophoresis showed a normal pattern. X-rays of the chest and thigh were normal. Intradermal tests with PPD (2 TU), coccidiomycin, and trichophitin were negative. Patch testing after contact sensitization with DNCB (7) was positive (2+). T cells were 36 per cent (nl 56% ± 6.2) and B cells 25 per cent (nl 26.5 % ± 6.2) by rosette technique. Tissue sections and culture revealed *B. haptosporus*. After one month of treatment with potassium iodide (30mg/Kg/day), the lesion decreased in size. She continued therapy for five months as an outpatient, and had no evidence of the lesion at examination seven months later.

Case 3

A 5-year-old boy from Castro Alves entered the hospital with a one-month history of a painless lesion on the trunk. Physical examination revealed an indurated erythematous and warm plaque with depressed center, on the right lateral aspect of the chest that measured 10 cm by 8 cm (Fig. 3). The remainder of the examination was negative. Hemoglobin, hematocrit, red cell count and blood sugar were within normal limits. There was a mild leukocytosis with eosinophilia. Intradermal tests with PPD (2 TU), coccidiomicin, streptokinase and DNCB (7) following senzitization were negative. The serum IgG

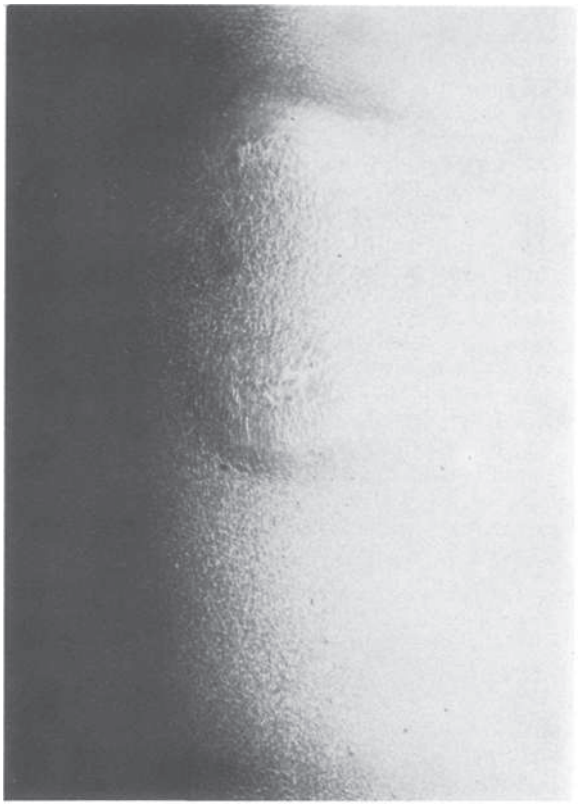


Fig. 3. Case 3. A firma plaque with central depression on the rigth lateral aspect of the chest.

level was 1,684mg %, IgM – 194mg %, IgA – 71,7mg %. T cells were 40 % and B cells 30 % as detected by the method of rosettes. Subcutaneous zygomycosis was diagnosed by histopathology and *B. haptosporus* was isolated in culture. The patient was treated foı 10 days with potassium iodide, 30mg/Kg/day, during which time the lesion decreased in size. Although he failed to continue his medication after this time, at follow-up, nine months later, the lesion had disappeared.

Histopathologic and mycologic studies

In all cases there was a heavy inflammatory reaction with fibrosis in the subcutaneous tissue. The inflammatory reaction consisted of a diffuse infiltration of eosinophils, plasma cells, histiocytes, neutrophils and few lymphocytes. There was a marked predominance of eosinophils in the infiltrate in two cases but in the first case they were rarely seen. Many abscesses containing hyphae and consisting of eosinophils or neutrophils could be seen throughout the

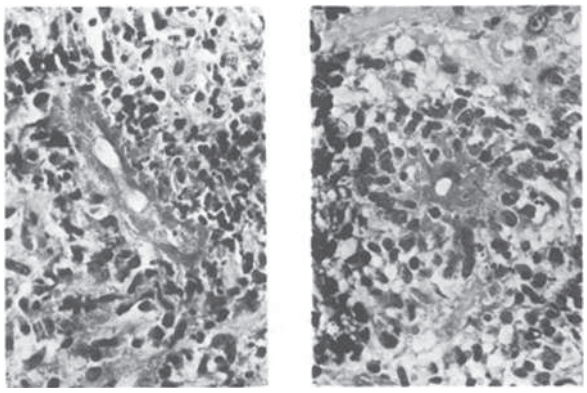
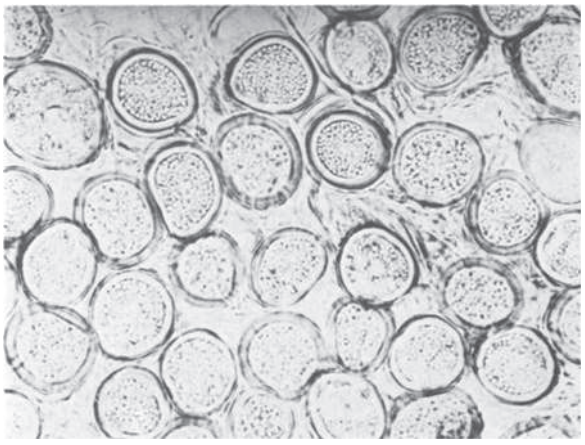


Fig. 4. Cross and oblique sections of hyphae with Splendore phenomenon. H & E (× 400).

inflammatory process. A palisade of giant cells and epithelioid cells surrounded the abscesses. Giant cells isolated or confluent and areas of lipophagia could also be seen. Fungal elements were found throughout the inflammatory process in longitudinal and cross-sections and were surrounded by a cuff of eosinophilic, PAS-positive material (Splendore phenomenon) (Fig. 4). The hyphae were PAS-positive and strongly argyrophilic by Grocott's method. The small arteries showed intimal thickening and inflammatory infiltration of the wall. The dermis contained only a mild, non-specific inflammatory reaction.

Cultures were obtained in Sabouraud dextrose agar at room temperature. Rapidly growing grayish colonies were obtained. They were waxy in consistence and appeared flat, folded and radially furrowed. On microscopic examination, large vegetative hyphae (10 to 20μ width)



were seen. As the spores production began, the hyphae became increasingly septate. Conidia, meristospores and the characteristic zygospores were also seen. Zygospores (20 to 50μ in diameter) were round and presented smooth thick wall (Fig. 5).

Discussion

It is interesting to note that subcutaneous zygomycosis by *C.coronata* was reported in Caribean Area, Colombia and in three Brazilian states, but the mycosis caused by *B. haptosporus* was diagnosed only in the Brazilian state of Bahia (4). All three patients come from nearby towns lying just north of 13 ° latitude S.

Our three patients were children presenting lesions on the thigh, buttocks, and trunk as it has been usually reported (6). All the lesions had the same characteristic aspect: an indurated erithematous and warm plaque with well defined raised margins.

The diagnosis of our cases was based on histopathologic aspects plus the cultural isolation of the fungus. It must be pointed out that a case was reported as a chronic type of zygomycosis of the abdominal wall extending to the abdominal viscera that presented an histopathological picture similar to that seen in subcutaneous infection (10). For that reason, the isolation of the fungus is important to the best knowlegde of this disease.

Humoral immune responses were normal in our patients but cellular immune responses showed some results that could lead to a more careful study of them, in order to verify their role in the pathogenesis of this micosis.

Potassium iodide is the drug of choice in the treatment of subcutaneous zygomycosis. It is worth noting the variable response to the drug presented by our patients.

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