OCCURRENCE OF SUBCUTANEOUS ZYGOMYCOSIS (ENTOMOPHTHOROMYCOSIS BASIDIOBOLAE) CAUSED BY BASIDIOBOLUS HAPTOSPORUS WITH PULMONARY INVOLVEMENT

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Abstract

A case of a two-year-old boy with multiple subcutaneous lesions caused by *Basidiobolus haptosporus* is presented.

The child had also a non-toxic familial goiter and clinical and radiological features of a pulmonary illness. The pulmonary manifestations only disappeared with the treatment with potassium iodide. The authors think that the pulmonary lesions must have arisen by direct spread of the fungus from the subcutaneous lesions of the chest.

Introduction

Subcutaneous zygomycosis due to *Basidiobolus haptosporus* is an infection which occurs primarily in children and has been described in Asia and Africa (4). Two of the authors had the opportunity to describe the first three South American cases of this infection with mycological confirmation, all observed in the State of Bahia, Brazil (1, 3).

This paper reports a fourth case with features considerably different from those previously described.

Case history

A two-year old mulatto boy from Tucano, Bahia, was admitted on 9.20.78 in unstable condition. He was wasted, apathetic, and pale and had dyspnea, fever, intercostal retractions, productive cough and assymetric abdominal distention. There were indurated subcutaneous lesions with irregular surfaces as follows:

1 – Extensive plaque involving the anterior and posterior right chest and the entire right flank, measuring 17 cm × 12 cm. firmly adherent to deeper structures (Fig. 1).

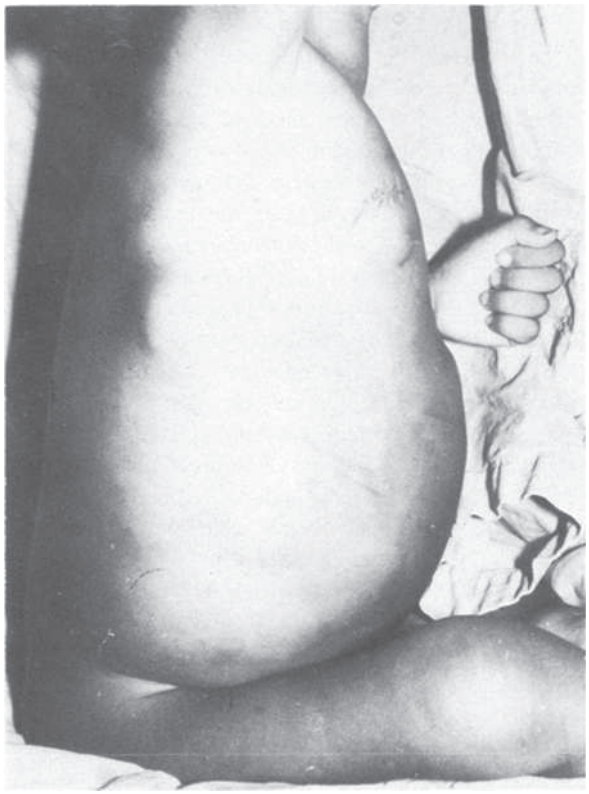

Fig. 1. See the lesions on the trunk and abdome.

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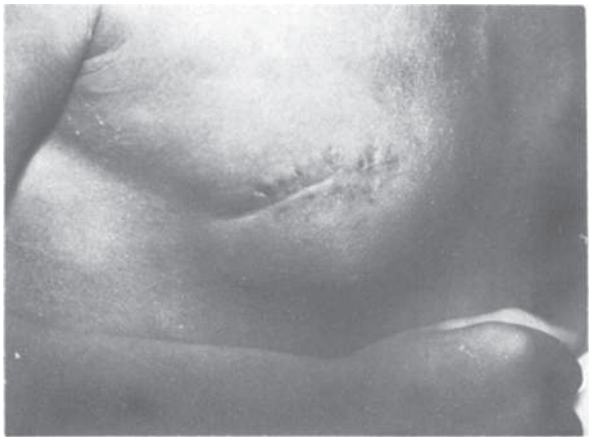

Fig. 2. A detail of the lesion of the right infraclavicular and mammary region.

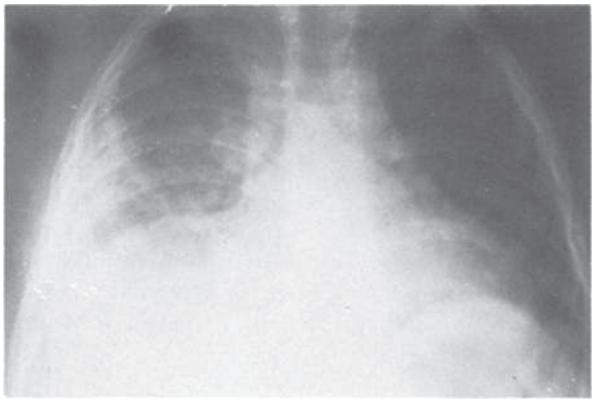

Fig. 3. Chest x-ray. See irregular densities in the right base oblitering the outline of the diaphragm.

2 – Single lesion in the left mammary region measuring 2 cm × 1,5 cm.

3 – Single lesion in the right infraclavicular and mammary region measuring 5,5 × 4 cm (Fig. 2).

4 – 4 cm × 4 cm lesion in the epigastric region

5 – Periumbilical lesion involving the umbilicus and measuring 3 cm × 3 cm.

Cervical, axillary and inguinal lymph nodes were enlarged. There was symmetrical enlargment of the thyroid. Examination of the chest revealed decreased expansion, dullness in the base, diminished breath sounds in the middle third, and absent breath sounds in the lower third of the right lung. The abdomen was markedly distended and assymetric due to the cutaneous infiltrative process. The liver and spleen were not palpable. There was a soft, depressible mass in the left iliac fossa. Laboratory: White blood cells were 13.100 with 22 % eosinophils. There was a marked hypochromic, microcytic anemia. SGOT, SGPT, alkaline phosphatase, glucose, cholesterol, calcium and phosphorus were normal. Total proteins were 10,1 gm %, with albumin of 4,5 gm% and globulins of 5,6 g %. IgC was 2, 250 mg %, IgM 326,3 mg %, IgA 242,1 mg %. T lymphocytes were 40 % by the rosette technique (normal value 56 % ± 6.2) and B lymphocytes were 13 % (normal value 26.5 % ± 6.2). Bone marrow morphology was normal. Complement fixation test for Chagas disease (Machado Guerreiro) was negative. Skin tests with PPD (2 T.U.), streptokinase-streptodornase, and candidin were negative. Contact sensitization with 2,4 dinitroChlorobenzene (DNCB) using a modified technique previously described (7) was positive at 2+. T3 (Triiodothyronine) and T4 (thyroxine) were within normal limits. TSH (thyroid stimulating hormone) was 48 mU (normal 2.1 to 915 mU). Anti-thyroid antibodies were absent. Chest x-rays showed irregular densities in the right base oblitering the outline of the diaphragm and an increase in soft tissues densities in the thorax and abdomen (Fig 3). Intravenous pyelogram and upper gastrointestinal series including small-bowel follow-through were normal. Barium enema revealed a dolichosigmoid. Biopsies were performed for histological and mycological identifications. There was a history of goiter in the family including the patient's mother. Tucano is not an endemic area of goiter. *Hospital course* – The clinical picture progressively worsened with an increase in dyspnea, cough, anemia and weight loss. Procain penicillin, 800.000 units per day, was given for seven days on account of the pulmonary signs, but without a change in the patient's condition. Follow-up radiographs showed opacification of the right base sugestive of pleural effusion. Following histologic diagnosis, treatment was begun with potassium iodide in increasing doses up to 40 mg/Kg/day. After two weeks there was marked improvement of the pulmonary picture and of the skin lesions, but the goiter showed a slight enlargement. The patient begun to receive Cynomel (L-Triiodotironine)* in an increasing doses up to 50 mcg/day. After one month the skin lesions disappeared and the patient was clinically normal, except for the goiter which had decreased in size. Chest x-ray showed only pleural thickening on the right. On follow-up examination two months later, the child remained well. *Pathology* Sections of skin showed fatty tissue replaced by an intense infiltrate of eosinophils, plasma cells, and histiocytes.

* Cynomel. Lab. Smith-Kline Enila Ltda.

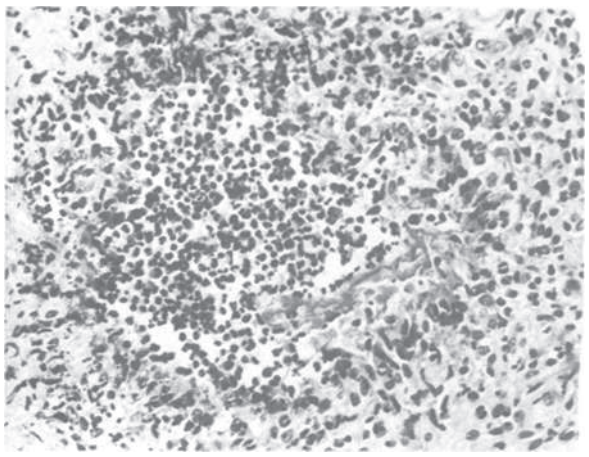

Fig. 4. An abscess surrounded by epithelioid and giant cells. HE (x400).

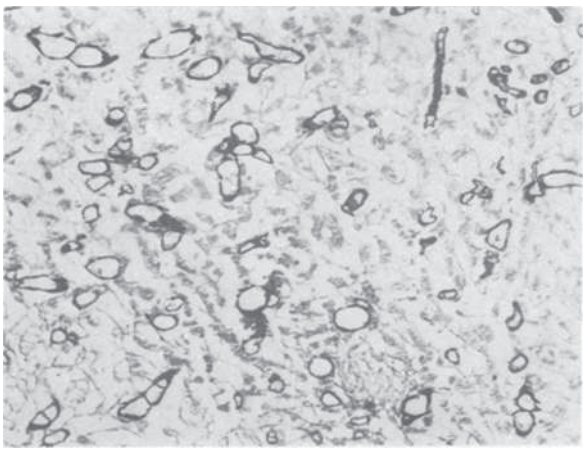

Fig. 5. Many hyphae coated with eosinophilic, PAS positive material (Splendore phenomenon). PAS (x400).

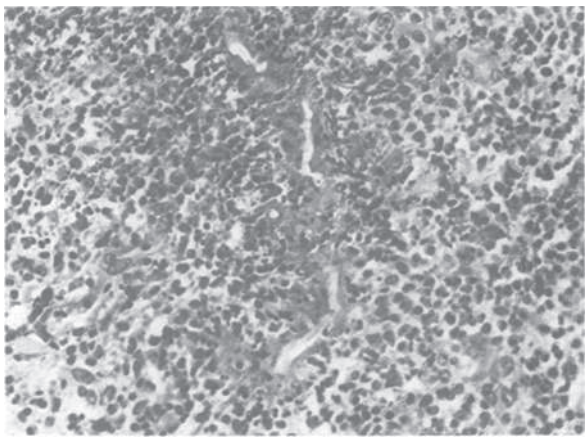

Fig. 6. Many hyphae impregnated by silver. Grocott's method (x450).

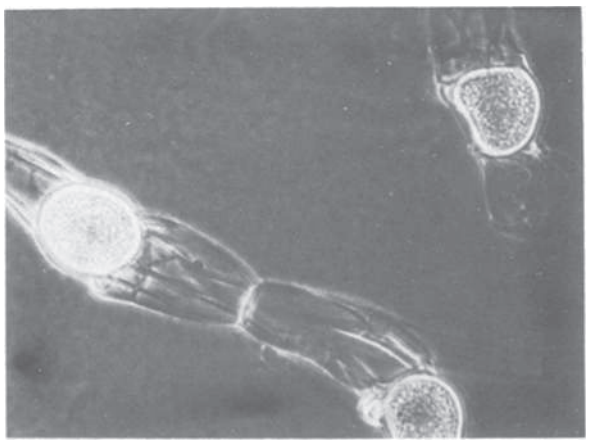

Fig. 7. See zygospores with smooth thick walls (x450).

Many abscesses consisting of eosinophils and neutrophils surrounded by epithelioid and giant cells were seen (Fig 4). In the midst of these abscesses large hyphae were noted coated with an eosinophilic PAS-positive material (Splendore phenomenon) (Fig 5). In fig. 6, we can see many hyphae impregnated with Grocott's silver. There were also zones of fibrosis and necrosis of fat. *Mycology* Cultures were obtained in Sabourand dextrose agar at room temperature. Rappidly growing grayish colonies were seen that were waxy in consistence and appeared flat, folded, and radially furrowed. Microscopical examination revealed large vegetative hyphae, conidia, meristopores and the characteristic zygospores with smooth and thick wall (Fig. 7).

Comments

This case appears to be the first case of visceral involvement in the chest by *B. haptosporus*. Deep thoracic involvement has been described for *Conidiobolus coronata*, a fungus of the same family as *Basidiobolus* and which also causes infection of subcutaneous tissue im man (5). Recently, one of us described two cases of involvement of abdominal viscera by entomophthorales in which there was no skin involvement. In these cases there was no mycological confirmation of the diagnosis (2). Our present patient presented clinical and radiological features of a pulmonary illness which only disappeared with specific treatment against the fungus. The pulmonary

lesions in this case must have arisen by direct spread from the subcutaneous lesions of the chest. He presented also multiple cutaneous lesions, an aspect rarely mentioned in the literature (4) and had association with a non-toxic familial goiter that increased with the isolate administration of iodide. Familial goiter results from defects in thyroxine biosynthesis and untreated patients may develop thyroid hormone deficiency (6). It is impossible to determine if the goiter favoured the development of the fungal infection in the present case.

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References

1. Bittencourt, A.L., C.R. Melo, O.M. Jalil & Z.A. Andrade. 1977. Basidiobolomicose. Apresentação de um caso. Rev. Inst. Med. trop. São Paulo 19: 208–212.
2. Bittencourt, A.L., M.A.R. Ayala, E.A.G. Ramos. 1979. A new form of abdominal zygomycosis different from mucormycosis. Am. J. Trop. Med. Hyg. 28: 564–569.
3. Bittencourt, A.L., A.T. Londrero, M.G.S. Araujo, N. Mendonça & J.A. Bastos. 1979. Occurrence of subcutaneous zygomycosis caused by Basidiobolus haptosporus in Brazil. Accepted for publication in Mycopathologia 68: 101–104.
4. Coremans-Pelsenner, J. 1974. Biologie des champignons du Genre Basidiobolus. Eidam 1886. Saprophytisme et pouvoir pathogene. Acta Zoologica e Pathologica 60: 7–143.
5. Eckert, H.L., G.H. Khoury, R.S. Pore, E.F. Gilbert, & J.R. Gaskell. 1972. Deep Entomophthora phycomycotic infection reported for the first time in the United States. Chest 61: 392–394.
6. Goldstein, J.L. & A.G. Motulsky. 1974. Genetics and endocrinology. pg 1014 in Williams, R.H. Textbook of Endocrinology. W.B. Saunders, Phyladelphia.
7. Morierty, P.L., A.L. Bittencourt, C. Pereira, R. Teixeira, & N. Guimarães. 1978. Borderline cutaneous leishmaniasis. Rev. Inst. Med. trop. São Paulo 20: 15–21.