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SUBCUTANEOUS SCEDOSPORIOSIS. REPORT OF TWO CASES AND REVIEW OF THE LITERATURE

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SUMMARY

Two cases of subcutaneous scedosporiosis, caused by *Scedosporium apiospermum*, are reported. Both patients had lesions localized in the forearm: a solitary ulceration in one and a sporotrichoid-like lesion in the other. The literature is reviewed.

KEYWORDS: *Scedosporium apiospermum*; Subcutaneous scedosporiosis; Subcutaneous mycosis.

INTRODUCTION

Scedosporium apiospermum (teleomorph, *Pseudallescheria boydii*) is a largely widespread water and soil inhabiting fungus[9] that causes a wide spectrum of diseases[5]. It may be classified among the pathogenic as well as among the opportunistic agents of mycoses[21]. It may also be a colonizer of the respiratory tract[25] and, in addition, has been implicated in allergic bronchopulmonary conditions[15].

Usually *S. apiospermum* proceeds as a pathogenic fungus when introduced traumatically into the skin or subcutaneous tissue of a normal host, causing mycetoma. But it may also elicit other types of subcutaneous lesion. However, in the immunosuppressed host, polymorphic cutaneous or subcutaneous lesions may result from the inoculation of the fungus.

Two cases of subcutaneous lesions caused by *S. apiospermum* will be reported and a review on the subject will be presented.

CASE REPORTS

Case 1. A 73 year-old white woman was seen by an ulceration in his left forearm, in December 1986. She complained of dyspnea, weight loss (25 kg in 10 months), anorexia and weakness. Her medical history included a carcinoma of the breast (February 1986) treated with chemotherapy and prednisone (60 mg/day) and a pleural effusion at the right lung (March 1986), which was shown to be neoplastic by a thoracocentesis.

The ulceration on the flexor surface of the left forearm measured 1 × 2 cm, was limited by a smooth erythematous border and was covered by a yellowish crust (Fig. 1). A biopsy was performed. A potassium hydroxide preparation of a portion of biopsied tissue demonstrated hyaline, septate, dichotomously branched hyphae. Another part of the biopsy was planted onto Sabouraud's dextrose agar incubated at room temperature. White floccose colonies were obtained. Microscopically solitary one-celled, pale brown ovoid conidia (4 to 9 × 6 to 12 μm) with truncate base on the apex of conidiophores (*Scedosporium* stage) were observed in slide culture.

Patient's pulmonary function continued to deteriorate and she died before receiving antifungal therapy.

Case 2. A 66 year-old white woman presented with three painful subcutaneous nodules on the flexor surface of the left forearm, three months ago. The lesions became ulcerated and were covered by dark necrotic crusts. A biopsy was performed and a diagnosis of pyoderma gangrenosum was established, in December, 1994. In spite of antibioticotherapy and topical use of mupirocin, the lesions progressed to necrotic plaques (Fig. 2).

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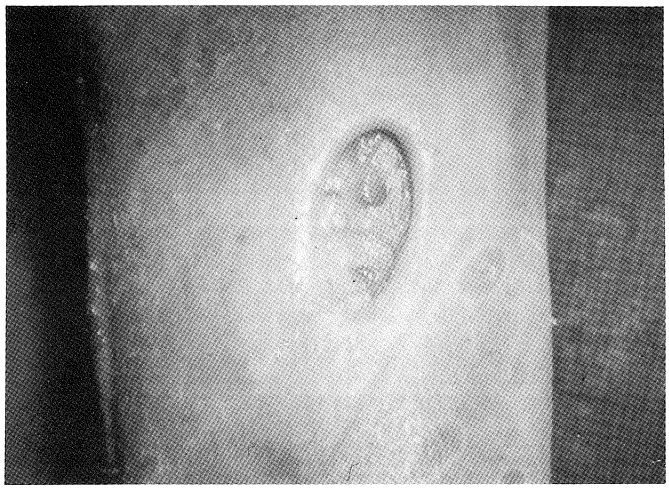

Fig. 1 – Patient 1. Forearm lesion showing ulceration with seropurulent discharge.

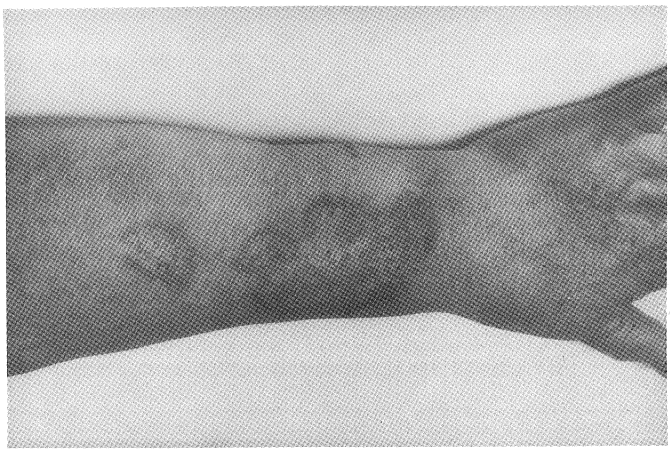

Fig. 2 – Hyperpigmented plaque lesion in forearm of patient 2.

In March, 1995 another biopsy was performed. A chronic inflammatory reaction was observed. A potassium hydroxide preparation of the exudate revealed hyaline, branched, septate hyphae and conidia. These fungal elements were well stained by Grocott (Fig. 3). Biopsied tissue was also planted onto Sabouraud medium with chloramphenicol. White floccose colonies was obtained; microscopically the conidial state of *Scedosporium* were observed.

The patient was treated with itraconazole (100 mg/day); in six months the lesions healed, and treatment was continued for one year.

DISCUSSION

Eleven cases of subcutaneous lesions caused by *S. apiospermum* could be gathered in the literature, two from Brazil[8, 11]. Patients data, localization and type of lesions, underlying conditions, diagnosis and treatment, including our cases, were shown in table 1.

Interesting findings on these 11 cases were as follows. Trauma at the site of the lesion and isolation of the fungus from the soil of the patient's garden was quoted by MORIN et al. (1987). Introduction of the fungus by surgical wounds was referred in three cases[10,25]. Corticotherapy was the underlying conditions in five patients[4,10,11,17,26], hyperfunction of adrenal glands and diabetes in one patient[3], and diabetes alone in another[25]. In nine patients the disease seems to be limited to the soft tissue; in one patient localized signs of arthritis and osteitis was disclosed by 99 mTc scan[26]; in the remaining patient, after being limited during 3 months, hematogenic disseminated lesions appeared in the opposite member and death occurred by respiratory failure in spite of no evidence of lung lesions[17]. Two patients presented lesions resembling those of lymphangitic sporothricosis[4,26]. It is interesting to refer that mycetoma can also, exceptionally, manifests as a sporotrichoid-like lesion[18]. Like our case nº 2, the patient reported by VAN HECKE et al. had a histopathological diagnosis of pyoderma grangrenosum in the examination of the first biopsy.

Two other reported cases have been reported in the literature as subcutaneous *S. apiospermum* infection. However, in one of them[27] the authors called into question if the cutaneous microabscess was not really the result of hematogenic dissemination of a non-elucidated pneumopathy. The other report[29], based only on medico-legal protocols, deals with a patient with a two year (1917-1919) non-elucidated suppurative lesion on a finger; two years later (1921) a furuncular-like lesions appeared on the hand and forearm, associated with subcutaneous

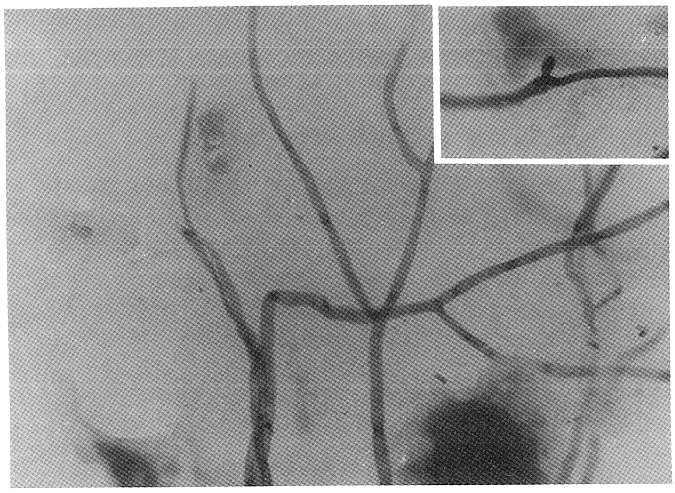

Fig. 3 – Branching, septate hyphae and conidium (insert) seen in biopsied tissue smear (Case 2). GMS (× 1000).

TABLE 1
Clinical features of thirteen cases of subcutaneous lesions caused by *Scedosporium apiospermum*.

Case (ref.)	Age,sex	Subcutaneous lesions	Underline condition	Diagnosis	Treatment;outcome
1 (12)	76, M	Soft tissue painful, erythematous swelling of the 4th and 5th finger; in one year, involved the hand, wrist and forearm. X-ray did not reveal bone lesions.	None	Culture of synovia and synovium	Amphotericin B and miconazole; relapse
2 (26)	76, M	Nodules (1 mm to 4 cm) from the dorsum of the 5th finger along the ulnar margin of the right forearm; arthritis and osteitis of the wrist, disclose by X-ray and 99mTc bone scan.	Prednisone, bronchitis, hypertension, temporal arteritis	Pus (direct, smears PAS); biopsy (Grocott) Culture of the pus	Ketoconazole; death: heart failure
3 (3)	59, F	Non tender, swollen, erythematous induration, covered by many pustular lesions on the righ leg 2 months duration.	Diabetes Cushing's disease	Biopsy (PAS). Culture of the pus and biopsied fragments	Miconazole. Control of immunosuppression
4 (25)	46, M	Infected surgical wound resulting from partial hemilaminectomy.	None	Culture of infected tissue	Surgical debridement
5 (25)	64, M	Infected axillo femoral graft.	Diabetes	Culture of the graft	Graft resection
6 (10)	24, M	Purulent material draining from a craniotomy wound.	Dexamethazone	Culture of the pus	Miconazole
7 (16)	45, F	Ulceration (2 × 5 cm) covered by adherent slough on the dorsum of right ankle.	None	Direct and culture of slough biopsy	Surgical debridement and skin graft
8 (17)	64, M	Painless tumefaction and ulceration (1 cm) with erythematous border, covered by a necrotic scar and draining a yellowish pus, 3 month duration.	Waldenstrom's disease Corticotherapy Cytotoxic drugs	Pus (direct), biopsy (Grocott), culture	Ketoconazole
9 (8)	38, M	Tumefaction of the right malleolar region with a fistula draining purulent material.	None	Culture of the pus	Surgical extraction of a rubber fragment
10 (11)	45, M	One painless, tender, flutuant nodule (2 cm) in the left knee 4 month duration.	Diabetes, renal transplantation	Biopsy (Grocott and culture) and corticotherapy	Surgery Itraconazole
11 (4)	73, M	Subcutaneous nodules distributed on the foot, tibial crest and tigh (0.5 to 3 cm), 4 month duration	Horton's disease Corticotherapy	Biopsy (Grocott) Culture of the pus	Itraconazole
This report					
Case 1	73, F	Ulceration (1 × 2 cm) on the flexor surface of the forearm.	Carcinoma of the breast Cytotoxic drugs corticotherapy	Direct and culture	None; death: pulmonary failure
Case 2	66, F	Three painfull nodules on the flexor surface of the left forearm.	None	Direct, smears (Grocott) and culture	Itraconazole; cured.

and muscular gumma-like lesions, arthritis, periostitis of many bones and orchiepidimitis; *S. apiospermum* was then isolated from an abscess; treated with vaccine obtained from the culture of the fungus the patient's lesions healed; death occurred two years later (1923), after a septicemic-like disease.

On the other hand, cutaneous or subcutaneous lesions resulting from hematogenic dissemination *S. apiospermum* infections have also been reported. They manifest as erytemathous papular rash[7], an eritemathous papular lesion with a necrotic center[6], a solitary pustule[24], or nodules with dusky, light green center[1]. Tumefaction[24] or induration of the skin[1], at the site of a catheter, has also been implicated as portal of entry of the fungus. Biopsy, direct examinations of pus and culture of these lesions led to the diagnosis of the mycosis.

Conidia of *Scedosporium* type are produced in the host tissue as well as in culture[9]. For that reason diagnosis of *S. apiospermum* or *S. inflatum* can be presumed on the basis of microscopic examination of clinical specimens in which hyphae and conidia are seen, as in our case n° 2[2,7,14,19,20,22,23,28]. However, species identification of the agent requires the isolation of the fungus.

RESUMO

Scedosporiose subcutânea. Relato de dois casos e revisão da literatura

São relatados dois casos de scedosporiose subcutânea por *S. apiospermum*. Em ambos os pacientes as lesões localizavam-se

no antebraço: em um deles, uma ulceração, no outro, uma lesão "esporotricóide". Foi feita revisão comentada da literatura pertinente.

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