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Two Cases of Chromomycosis

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More than 190 cases of chromomycosis have been reported in Japan since the first one in 1933. We encountered two cases of this fungal disease in the last four years. The diagnosis of chromomycosis is still difficult, especially in the early stage and brain metastasis is frequently seen in this country. We report here these two cases with a discussion on clinical manifestations of previous reports in Japan for early diagnosis.

(Key Words: Chromomycosis, Fonsecaea Pedrosoi, Amphotericine B)

INTRODUCTION

Chromomycosis occurs in tropical and subtropical regions and it has been said to be a relatively rare disease in Japan. However, case reports of this disease have recently increased in number in this country. From February 1975, when our hospital opened, to March 1979, we also encountered two cases of chromomycosis. The period between the development of the lesions and consultation was long because of erraneous diagnosis such as bacterial infection or hemangioma.

In this paper, we report these two cases and discuss the clinical manifestations compared with previous reports in Japan.

CASE REPORTS

Case 1 (9)

A 48-year-old male, company employee, living in Ninomiya, Kanagawa, consulted our clinic on September 26, 1975 because of a lesion on his back (Fig. 1). He had engaged in physical labor for a half year in 1967. After he left his work, the lesion on his back was pointed out by his family. The lesion was diagnosed as hemangioma by a local doctor. Eight years later, he visited our clinic because the lesion had become larger. His personal and family history were unremarkable.

Physical Examination

On the right side of the back, a well-defined, rough, somewhat elevated scaly erythematous plaque was present. The lesion was 10 × 10cm in size (Fig. 1). No enlargement of the neck, armpits and inguinal lymph nodes were palpable. He had no symptoms.

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KOH test

In direct microscopy of KOH mounts of lesion scale, sclerotic cells were observed.

Culture

A black, filamentous mold was revealed by the culture of biopsy specimens on Sabouraud's dextrose agar at room temperature and it was identified as *Fonsecaea pedrosoi* by conidia formation of the cladosporium type in corn-meal slide cultures.

Histologic Findings

Sclerotic cells were found in the upper dermis and in the giant cells of granulomatous tissue.

Treatment

The patient was said to have been given local injections of amphotericin B and a surgical operation by a local doctor.

Case 2 (12)

A 57-year-old male, company employee, living in Chigasaki, Kanagawa, consulted our clinic on February 7, 1978 because of a lesion on his right upperarm. He noticed it in 1974 and visited our clinic in 1978 because of the increasing size of this lesion. His personal and family history were unremarkable.

Physical Examination

On the flexural aspect of his right upperarm, a well-defined, verrucoid, somewhat elevated, scaly and crusted erythematous plaque was present and it was slightly infiltrated and surrounded by an area of brownish pigmentation. This lesion was 3.4 × 1.8cm in size (Fig. 2). No enlargements of the right armpit lymph nodes were palpable. The patient had no symptoms and appeared well in general.

KOH test

In direct microscopy of KOH mounts of lesion scale, sclerotic cells were observed (Fig. 3).

Culture

A black, filamentous mold (Fig. 4) was revealed by the culture of biopsy specimens on Sabouraud's dextrose agar at room temperature and it was identified as *Fonsecaea pedrosoi* by conidia formation of the cladosporium and acrotheca types in corn-meal slide cultures (Fig. 5).

Laboratory Findings

The blood sedimentation rate was 30mm in one hour and 57mm in two hours. Hemograms, urinalysis, X-ray examination of the chest, electrocardiogram and liver function tests were normal. The serological test for syphilis (STS) and rheumatoid arthritis test (RA-test) were negative. Antistreptolysin-O (ASLO) was 40U/L. C-reactive protein (CRP) was weakly positive.

Histologic Findings

Hyperkeratosis and acanthosis were shown in the epidermis. The dermis was infiltrated with lymphocytes, histiocytes and giant cells. By Periodic acid-schiff (PAS) and Grocotto staining, sclerotic cells were shown in the upper dermis (Fig. 6).

Treatment

Local infiltration with amphotericin B was performed once a week. On the second injection, urticaria and exsudative erythema also appeared eight hours later. The drug eruption due to amphotericin B was confirmed by a skin test.

A surgical excision was performed including the non-involved area 2cm outward from the involved area in the Department of Plastic Surgery in our school. The patient had no recurrence of the lesion in the following 10 months.

DISCUSSION

Chromomycosis is a type of deep mycosis caused by pathogenic dematiaceus fungi. The source of this fungus is soil and wood (1). It occurs mainly in tropical and subtropical zones (1). The lesion is on exposed sites, paticulary on the feet, legs, arms, face and neck (1) because the infection is usually followed minor trauma. Two types of clinical features of this disease are classified (3). One of them shows papillomatous hyperplasia which looks like a cauliflower. Another type develops a somewhat elevated and relatively smooth plaque. If the affected region is enlarged, central healing may be shown. Hematogenous spread occur in rare cases and brain abscesses,which are fatal, may develop. Fukushiro *et al* (3) noted that more than thirty cases of brain abscess of chromomycosis had been reported in the world and about a half of those cases had been seen in Japan. The brain abscess often affects young children.

Histological aspects include pseudo-epitheliomatous hyperplasia in the epidermis, and abscesses with neutrophils and granulomatous inflammation containing Langhans' giant cells in the dermis. Round, thick-walled fungus cells which are known as "sclerotic cells", may be found. The color of these cells is brown in hematoxylin eosin (HE) stained sections and deep ruby red in PAS stained sections. In direct microscopy of the KOH test of scales, brown, thick-walled cells may be seen.

The first case of chromomycosis in Japan was reported by Kano in 1933 (5). After that, 190 cases had been reported by 1976 (10) and 11 of these cases had been reported in Kanagawa prefecture up to 1972 (6).

We compared our cases with previous cases in Japan.

1) *Species of causative fungi*

The causative fungi isolated from 163 Japanese cases were *Fonsecaea pedrosoi* in 118 cases (72.4%), *Phialophora dermatitidis* in 16 (9.8%), *Phialophora gougerotii* in 10 (6.1%), *Phialophora verrucosa* in four (2.5%) and *Cladosporium trichoides* in one (0.6%) (3). Our two cases were identified as *Fonsecaea pedrosoi*.

2) *Factors for occurence*

Trauma, a decrease in immunoresponse and the influence of steroids have been reported as factors in this infection by Sasagawa *et al* (10). The patient reported as case 1 had engaged in physical labor for a half year and he used to lie down on the grass and the ground for a rest without any shirt on. The patient in case 2 had a kitchen garden. However, there was no apparent relation between trauma and infection of chromomycosis in either case. They had no past history of repeated viral infections

suggesting the presence of immunodeficiency. They had no history of oral or tropical steroid therapy.

3) *Age, Sex*

The condition has occurred equally in both sexs from 3 to 77-years of age in Japan (8). It was not suggested that agricultural workers are more often affected (4).

4) *Treatment*

Chromoycosis had been treated with local infiltration of amphotericin B (2), oral 5-fluorocytosine (5-FC) or surgical excision (11). Recently, it has been reported that systemic administration of 5-FC is effective and has no side effects (11). However, Maruta *et al* (7) reported that it is difficult to cure this disease with only systemic 5-FC because of the development of resistance to the drug in the early stage of treatment. Side reactions of local injections of amphotericin B are pain, swelling, fever and drug eruptions as seen in case 2. Therefore, surgical excision was often performed. Before excision, the involved area became smooth but infiltration remained and cultures of the involved area still showed growth of fungi. Therefore, we suggest that it is necessary to excise an area several centimeters larger than involved area even in relatively early stages of the infiltrated type.

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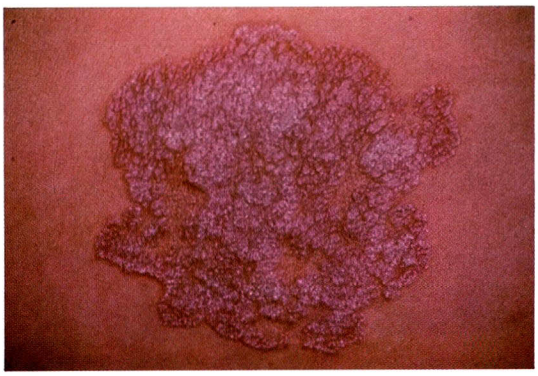

Fig. 1 Clinical features of case 1 (back).

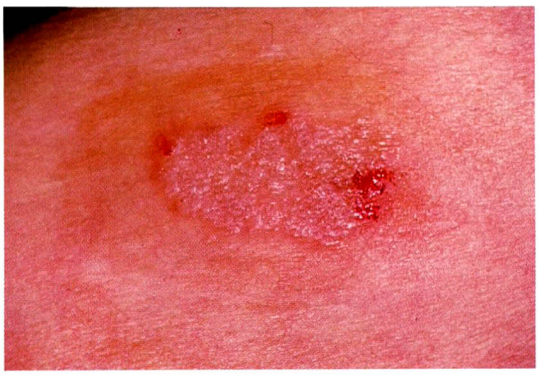

Fig. 2 Clinical features of case 2 (r-upper-arm).

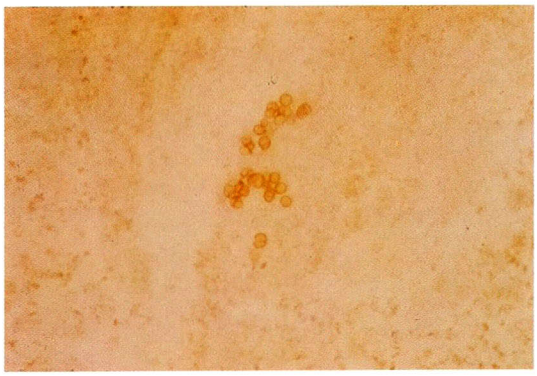

Fig. 3 Sclerotic cells in direct microscopy of KOH mount of scale (case 2).

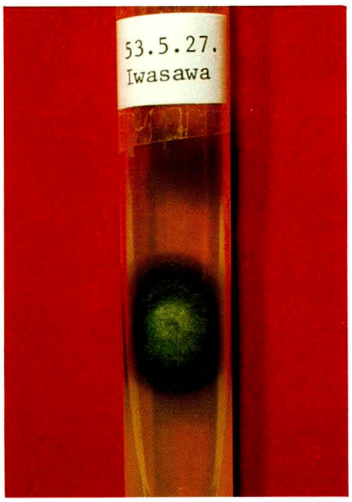

Fig. 4 Culture of scale (case 2).

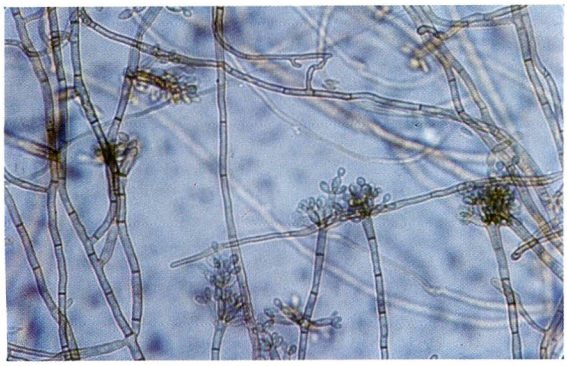

Fig. 5 Conidia formation in corm-meal slide culture (case 2).

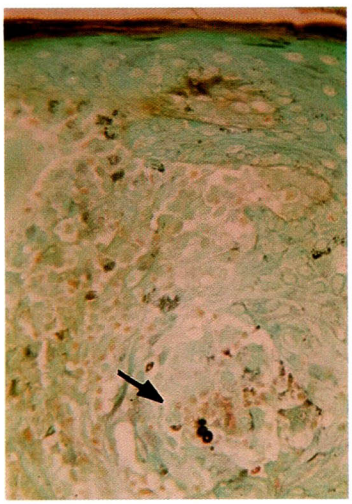

Fig. 6 Sclerotic cells in the upper dermis (Grocotto stain) (case 2).

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