

A CASE REPORT OF CHRONIC MUCOCUTANEOUS CANDIDIASIS IN IRAN

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ABSTRACT

A case of chronic mucocutaneous candidiasis due to *Candida albicans* in a 13 yr-old boy is reported. Evaluation of cell mediated immunity revealed cutaneous anergy to PPD and low level of T-Lymphocytes. B-Cell count and immunoglobulin G(IgG) were increased and there was a low level of serum iron. The patient was put under treatment with oral administration of Ketoconazole and ferrus sulfate, which resulted in some clinical improvement.

INTRODUCTION

Chronic Mucocutaneous Candidiasis is a rare syndrome (1). Patients show persistent candida infection of the mouth, skin and nails (2). It is clinically characterized by red, raised, serpiginous and scaly lesions, often with marked hyperkeratosis (3); the lesions are not tender or painful (3). It usually starts in infancy or early childhood (2). Its pathogenesis is not fully understood, but it is frequently associated with a depression of

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cell-mediated immunity (CMI) (4). In some cases, there is associated endocrinopathy (especially hypoparathyroidism, hypoadrenalism, hypothyroidism, or diabetes mellitus)(5). The disease is difficult to treat and is often resistant to conventional therapy (6). Ketoconazole is an imidazole derivative that has been shown to have a wide antifungal spectrum activity against candida species (7). Several cases of Mucocutaneous candidiasis successfully treated with ketoconazole have been reported (7,8,9,10,11). In this paper a case of Mucocutaneous candidiasis in which candida albicans was the causal agent is reported. the patient was treated with oral ketoconazole and ferrus sulfate (for hypochromic microcytic anemia).

MATERIALS AND METHODS

A 13 years old boy from kerman, was admitted to Dermatology Ward of Loghman Hospital, Beheshti University of Medical Sciences, Tehran, in January 1989, for crusted nodules and productive cough (Figure 1). Past history revealed that he was the ninth child of his family and had no abnormality at birth. From the first months of his life, the patient had had recurrent respiratory tract infections and chronic diarrhea. His mucocutaneous lesions appeared first at 3 years of age. Family history failed to show any similar problem in his parents or siblings.

Physical Examination: the patient was ill and pale and his weight (22.5 Kg) and height (118 cm) were below normal. In the right eye there was an irregular opacity on the cornea, which was due to previous keratitis and had impaired the patient's vision. On auscultation there were rales and ronchi in both lungs. Heart sounds were normal. Spleen was palpable below the costal margin. No peripheral adenopathy was palpable. Clubbing of all fingers was present. His intelligence seemed normal and there was no abnormality in neurologic examination. Examination of the skin showed scaly, crusted nodules and plaques on tip of the nose and ears, with papules on backs of the hands. Examination of oral mucosa revealed white patches on the mucosa of the cheeks fissured tongue. Also angular cheilitis was present.

Laboratory Investigation: The blood, urine analysis and all immunological, immunoelectrophoresis and hemoglobinelectrophoresis tests were normal except the following tests:

Ca=7 (9-11), Alk ph=26 (5-15), ET (T cell)=39% (60 ± 12), EAC (B cell)=41% (22 ± 5), NBT= positive, IgG=24 mg/ml (6-17 mg/ml), IgM=1/5 mg/ml (0.3-1.4 mg/ml)Chest X-Ray= Patchy infiltration in both lung field.

Mycological Examination: The patient was referred to the Mycology Department, School of public Health, Tehran Medical Sciences University, for further investigation. A fresh preparation of the crusts collected from the lesions with potassium hydroxide showed budding cells and pseudohyphae (Figure 2).

Histological section of a biopsy of the nose lesion showed abundant branching, septate mycelia (Figure 3). A portion of the biopsy and crust materials was also cultured on Sabouraud dextrose agar (5), Sabouraud dextrose agar containing Chloramphenicol and Cycloheximide (SCC), brain heart infusion agar (BHI) and blood agar. Sets of culture were incubated at 25 and 37 . colonies on (S), (SCC) and blood agar media became visible after 3 days of incubation at 25 and 37 (Figure 4). they were creamy and white. After the API system test, the isolate was identified as candida albicans.

Investigation of thyroid and parathyroid hormones was not possible.

Treatment: The patient was put on Nizoral (Ketoconazole), 50 mg/day, Ferrus sulfate, one tablet daily, and antibiotic (for respiratory infection). After a week of Treatment the patient left the hospital and until now has not returned for follow-up.

DISCUSSION

Chronic mucocutaneous candidiasis is likely to occur in individuals with an impaired immune response mechanism (6). In some cases the defect is in both the humoral and cellular immune systems (12).

In addition to congenital defects of CMI, there are several nutrients, mainly, glucose and iron, that are important for growth of candida (4).

Iron abnormalities affect host resistance to candidiasis. Low serum and tissue levels of iron and hypochromic anaemia are seen in familial chronic mucocutaneous candidiasis

(13). In our patient humoral antibody function was normal, except for increased immunoglobulin G (IgG). Endocrinologic investigations for thyroid and parathyroid function were not made, but liver function tests were within normal levels. Serum iron was at the lower level of normal range. Cell mediated immunity revealed impaired delayed hypersensitivity reaction to PPD and low levels of T. lymphocytes (by T. rosette forming cell). Medical history showed that the patient had suffered from recurrent respiratory tract infections and chronic diarrhea.

Keratoconjunctivitis has also been reported in chronic candidiasis patients (3). In this patient an irregular opacity on the cornea, which was due to previous keratitis and had impaired his vision, was observed. Treatment of mucocutaneous candidiasis is difficult. Ketoconazole is a well absorbed oral imidazole which has been used for treatment of mucocutaneous candidiasis in several cases (7,8,9,10,11). In the present case, after a week of treatment with Ketoconazole, the patient left the hospital with some clinical improvement, but until now he has not returned for follow-up.

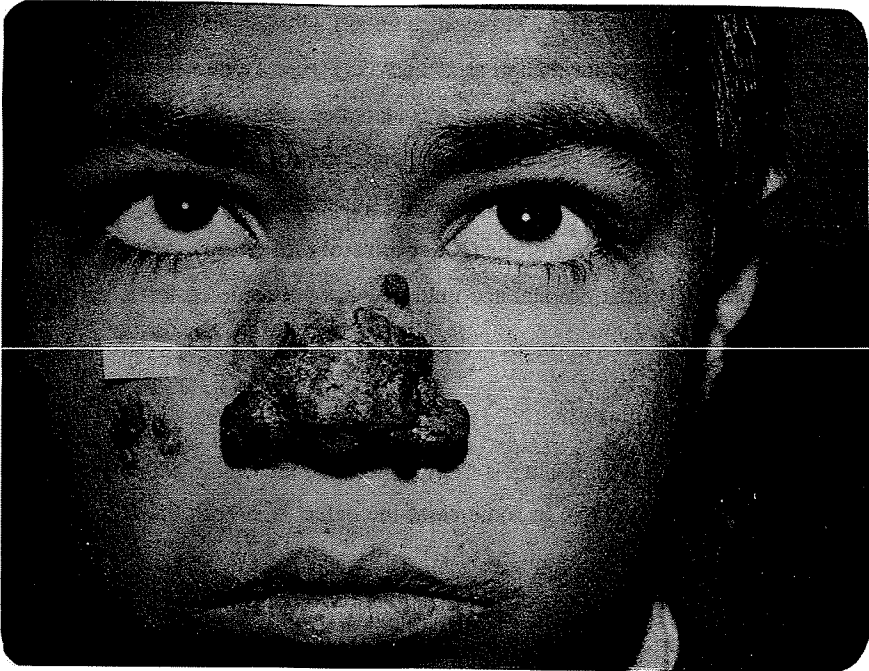


Figure 1- Crusted nodules and plaques on the skin of the nose and cheek.

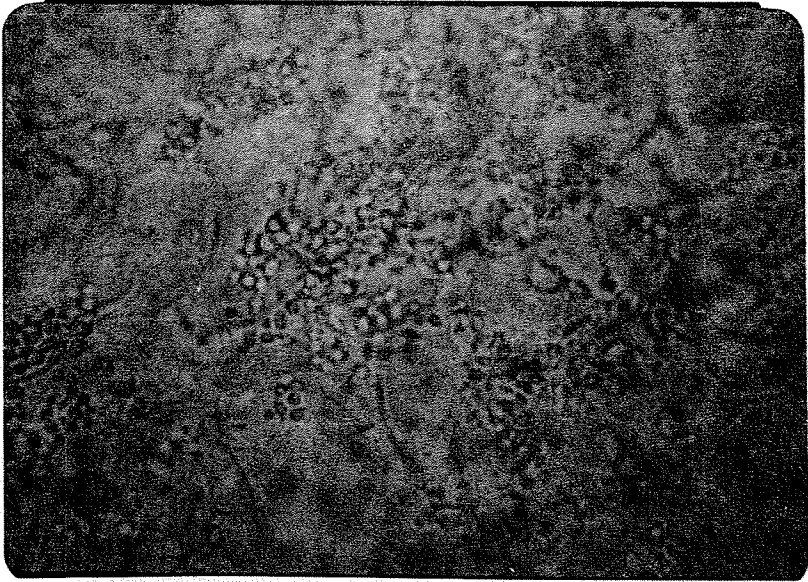


Figure 2- Potassium hydroxide preparation of the nose lesion showing budding cells and pseudohyphae.

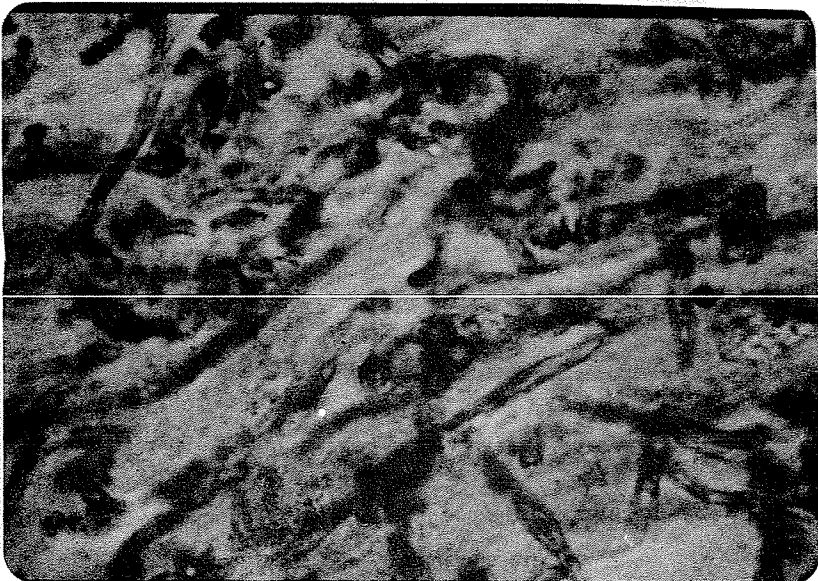


Figure 3- Histological section of the nose lesion showing abundant branching, septate mycelium (H&E x 1000).



Figure 4-3 day old culture of the nose specimen on blood agar and Sabouraud dextrose agar at 37 and 25^{°C}.

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