INTRODUCTION

Scytalidium dimidiatum (synanamorph Nattrassia mangiferae) is a pigmented mould.1 It is mainly a plant pathogen especially affecting fruit trees but it can also cause human infections.2,3 Infection can result from direct or indirect contact with contaminated soil or plants. In immunocompetent host, it may cause chronic superficial skin diseases and onychomycosis whereas in immunocompromised patients, it causes deeper infections such as subcutaneous abscesses, mycetoma and fungemia.3,4 Superficial infections caused by Scytalidium dimidiatum are difficult to be distinguished from those caused by the dermatophytes.

We present a case of invasive fungal infection caused by Scytalidium dimidiatum in a young individual involving left orbital area, orbital cavity and maxillary sinus (left) leading to proptosis of left eye. The importance of the case is that the infection was non-traumatic in nature in an immunocompetent individual.

CASE REPORT

A 19-year-old soldier presented with gradual painless protrusion of left eye with blurring of vision early in the morning that improved as the day passed. The symptoms were gradually increasing for 3 months. There was no history of trauma and the only notable point in the personal history was contact with mango trees in his native town.

On examination, his vital signs were within normal limits. There was 8 mm proptosis of the left eye with inferolateral dystopia, and a firm mass was felt at left superonasal orbital area. Visual acuity was normal and extraocular muscles movements were intact. Examination of chest, heart, abdomen and extremities revealed no abnormality.

Blood complete picture including erythrocyte sedimentation rate, urine routine examination, X-ray chest, thyroid profile, fasting and postprandial plasma glucose and liver function tests were within normal limits. C-reactive protein was less than 6 mg/L and Mantoux test was positive. A contrast enhanced computed tomography scan of the head revealed an aggressive mass (6.0 x 4.5 x 2.0 cm) involving left orbit (displacing the left eye ball inferolaterally), maxillary antrum and subcutaneous tissue overlying left maxilla (Figure 1). Magnetic resonance imaging of the skull revealed intra- and extra-conal peri-bulbar and retro-bulbar enhancing mass along with left intracranial extension.

Initially lymphoma was suspected, subsequent histopathological examination of biopsy specimen revealed chronic granulomatous disease on Hematoxylin and Eosine stain, whereas PAS stain showed septate fungal hyphae (Figure 2). Cytology for malignant cells was negative. Tissue for bacterial and fungal culture was taken and patient was put on systemic antifungal, inj. amphotericin B 25 mg I/V OD, along with inj. hydrocortisone 100 mg I/V and symptomatic treatment.

ABSTRACT

Scytalidium dimidiatum is mainly responsible for human skin and nail infections but the mould has also been reported for invasive infections in immunocompromised individuals. We report a young immunocompetent individual diagnosed with invasive non-traumatic Scytalidium dimidiatum infection involving the left orbital cavity and maxillary sinus.

Key words: Scytalidium dimidiatum. Invasive fungal infection. Immunity.
Biopsy specimen was inoculated onto MacConkey Agar incubated at 37°C in air, blood agar incubated at 37°C both aerobically and anaerobically and chocolate agar at 37°C with 5 % supplemented CO₂.

Part of biopsy specimen was also inoculated aseptically onto Sabouraud’s dextrose agar (SAB), SAB with chloramphenicol (100 mg) and SAB with actidione (0.05 mg/L) and chloramphenicol. The plates were placed inside polythene zippers and incubated aerobically at 22°C and 35°C.

The culture plate without cycloheximide subsequently started showing growth in 7 days that was identified later as *Scytalidium dimidiatum* by its colonial and microscopic features. The colonies on SAB were floccose, grey at first, becoming dark brown to black and abundant growth of grayish black mycelia, with grayish black reverse (Figure 3). Microscopic examination by cellophane tape preparation demonstrated septate, branched, subhyaline to dark brown hyphae that fragmented to form cylindrical, non-septate or single septate arthroconidia (Figure 4). It could easily be differentiated from *Scytalidium hyalinum*, a non-melanin producing species with whitish floccose colonies, arthrospores are single walled and vary in shape from elongated narrow to short wide cylinders.

![Figure 3: Typical colonies of Scytalidium dimidiatum on SDA.](image)

![Figure 4: Lactophenol preparation from the culture growth showing septate, branched and fragmented hyphae.](image)

The isolate was sensitive to amphotericin B (MIC=0.5 µg/mL) and itraconazole (MIC of 1 ug/mL) whereas fluconazole was in intermediate range. The antibiotic susceptibilities were determined by E-test.

After two weeks, treatment was discontinued as the patient had to proceed on emergency leave. Two weeks later, he reported back with increased proptosis. Review computed tomography revealed increased dimensions of the mass. Injection Amphotericin B was restarted and re-evaluated. His immune status (immunoglobulins, lymphocyte subsets, neutrophil function test, cANCA, pANCA) was found to be normal. HIV, Brucella, Toxoplasma, Tuberculosis serology and PCR for *Mycobacterium tuberculosis* were negative.

Surgery was planned and left orbital and maxillary exploration was done for debridement and debunking of the mass. Histopathology of tissue specimen again revealed chronic granulomatous inflammation and tissue fungal culture yielded growth of *S. dimidiatum*.

Postoperative recovery was smooth. Amphotericin B treatment was continued for 3 months and afterwards the patient was shifted to oral itraconazole. For the initial one month, he was given 300 mg itraconazole daily in 3 divided doses, then 200 mg for one month followed by 100 mg OD dosage. Presently, he is on oral treatment for the last 6 months and has shown marked clinical and radiological improvement.

**DISCUSSION**

*Scytalidium dimidiatum* was formerly known as *Hendersonula toruloïdes*. It is a thermotolerant, melanin producing dematiaceous mould thriving in plants mainly of the genera Plantus and Pinus. Infection can result from direct or indirect contact with contaminated plants or soil, predominantly in tropical and semitropical regions like South America, India, Southeast Asia, Caribbean and West Africa. It has been identified as pathogen for a variety of trees including Citrus, *Magnifera indica* (mango), Arbutus and others in North America, Arizona and Colombia. The present case belonged to an area that is famous for mango farming. The nature of contact with mango trees was probably incidental while the patient was on holidays followed by delayed onset of the symptoms.

*S. dimidiatum* tends to form dark pigmented colonies and recent work has established the production of melanin. Growing evidence suggests that melanization extends the survival of *Scytalidium* species in the environments by protecting the mould from ultraviolet radiations, extremes of temperature, free radicals like oxygen and nitrogen. It also produces keratinase, an enzyme that helps skin invasion.

*Scytalidium dimidiatum* has mainly been implicated in superficial skin and nail infections but can also involve other sites following accidental inoculation. It has been reported to cause mycetoma, subcutaneous abscesses, fungemia, and endophthalmitis following trauma usually in immunocompromised patients. Previous studies from Pakistan have reported *Scytalidium dimidiatum* as a cause of onychomycosis at around one percent and has also been implicated in tissue infections.

The extent and duration of neutropenia are major threats for the development of invasive *Scytalidium dimidiatum* disease in immunocompromised persons. AIDS, diabetes mellitus, chronic corticosteroid therapy, transplants and chronic renal failure have been the underlying morbidities in patients with deep tissue infections and maxillary sinusitis. Our case, as such, was thoroughly investigated for any underlying immune-deficiency but was normal.

*Scytalidium dimidiatum* is considered very difficult to treat not only in deeper infections but also in superficial infections. The clinical response to treatment is usually
poor despite *in vitro* susceptibility to several antifungal agents like amphotericin B, ketoconazole, miconazole, itraconazole, within the limit of attainable serum and tissue levels. Treatment outcome for deep infections has been unpredictable as no single agent has established efficacy. Amphotericin B has been reported useful in patients of facial lesions with underlying immunosuppression. However, effective treatment of invasive disease in immunocompromised patient requires management of neutropenia along with administration of antifungal agent. Voriconazole may prove a substitute option for treating invasive disease patients with poor renal functions. Our case responded well to amphotericin B followed by long-term oral itraconazole. Due to young age of the patient with fully intact renal functions and absence of any co-morbidity, amphotericin B could be administered for 3 months.

The message is that deep fungal infections are very much prevalent in our set-up but remain undiagnosed to underdiagnosed due to lack of diagnostic facilities or expertise. The scenario is even more important for case like this as it demands a multifaceted treatment approach like removal of predisposing factors, surgery and prolonged antifungal therapy.

**REFERENCES**