

### Subcutaneous facial mycosis in a child due to *Madurella mycetomatis*

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#### Abstract

Mycetoma is a chronic, granulomatous, subcutaneous, inflammatory disease caused by true fungi (eumycetoma) or filamentous bacteria (actinomycetoma). Eumycetoma usually affects adult males involving limbs and other exposed body parts. Children represent the least commonly encountered age group with this disease. A case of subcutaneous facial mycosis due to *Madurella mycetomatis* in a three year old child was diagnosed at the Microbiology department Armed Forces Institute of Pathology Rawalpindi, which to our knowledge is the first case reported of its kind. Early diagnosis and timely medical therapy lead to favourable outcome without any surgical intervention.

#### Case Report

A three-year-old child, an Afghan refugee living in the slums of Rawalpindi, Pakistan was brought to the outpatient department of Rawalpindi General Hospital by her parents with the complaint of a small pea size swelling on her left cheek. It had grown gradually in size with a brownish discolouration of overlying taut skin. The swelling was non-fluctuant, non-pulsatile, rubbery in consistency, and non-adherent to the underlying tissues. The child started to have fever, which was low-grade intermittent in nature, without rigors and chills occurring at night. She lost appetite and started losing weight and lost six kilograms in three months. She was referred to the Dermatology department of Rawalpindi General Hospital where a tissue biopsy was taken. A provisional diagnosis of Embryonal Rhabdomyosarcoma was made but tissue biopsy showed only non-specific chronic inflammation. Her symptoms aggravated over subsequent months, leading to the involvement of the lower side of her left eye orbit occluding corresponding visual field. She was referred to the ENT dept of Combined Military Hospital Rawalpindi for evaluation of possible involvement of her paranasal sinuses. On careful clinical examination the paranasal

sinuses were found free from the effects of the overlying pathology. She was also referred to the surgeon for possible surgical treatment. There was no bony involvement on radiological investigations. A tissue biopsy from the lesion for histopathology was taken and she was referred to the Armed Forces Institute of Pathology, Rawalpindi for further investigations.

On examination she had mild pallor with no lymph node enlargement and all vital signs were normal. She had haemoglobin of 9.9 g/dl with a White blood cell count of  $12.9 \times 10^9/L$  (66% Neutrophils, 18% lymphocytes, 12% Monocytes, 4% Eosinophils). Her erythrocyte sedimentation rate was 125 mm for first hour. A fine needle aspiration from the site was performed at the Microbiology department for both bacterial and fungal cultures. The aspirate was examined, showing no granules and no fungal hyphae on a wet mount. It was cultured on blood agar and



Figure 1. Yellow fluffy powder like growth with visible aerial hyphae.

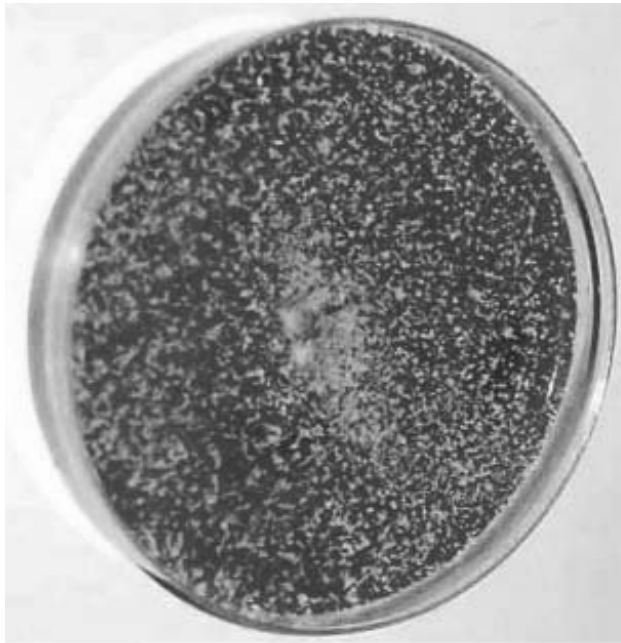


Figure 2. Small breadcrumb yellowish colonies.

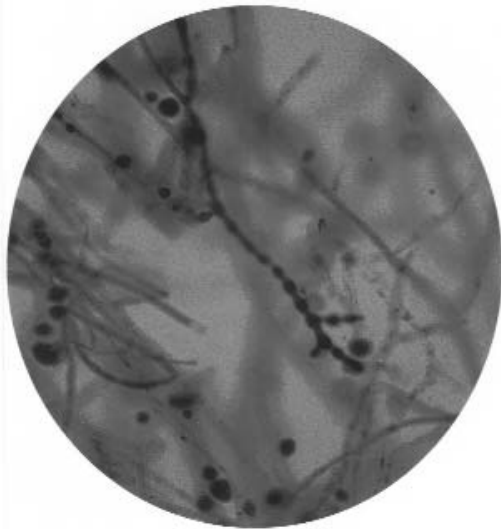


Figure 3. Septate hyphae with lateral phalides.

MacConkey agar for bacterial culture and incubated at 37° C for 3 days. The fungal cultures were done on Sabouraud dextrose agar with chloramphenicol (without cyclohexamide) incubated at 22°C for five days. The Sabouraud agar revealed yellowish fluffy powdery growth with visible aerial hyphae after 72 hours of incubation (Figure 1). The Blood agar plates were further incubated, showing small breadcrumb yellowish colonies after 96 hours of incubation, which showed hyphae on a wet mount (Figure 2). Microscopic morphology in Lactophenol blue preparation revealed septate hyphae with lateral phalides. Round to oval conidia were also seen. The growth was identified as *Madurella mycetomatis* (Figure 3).

Keeping in view the possible threat of involvement of the cavernous sinus and poor response of the fungus to therapy, it was decided to treat the disease aggressively. It was planned to start the treatment with Voriconazole but due to lack of availability as an alternative, intravenous Amphotericin-B 0.5 mg/kg of body weight was started. The dosage was further increased up to 1.0 mg/kg of body weight after two weeks. This treatment was supposed to be given to the patient for at least three months; however, the patient developed phlebitis after 3 weeks and had to be put on oral Itraconazole 100mg/day, which was continued for six months. Her renal profile and liver function tests were constantly monitored and they all remained normal. Patient showed very good response to the treatment and her symptoms regressed quickly. No relapse was seen on subsequent follow up for 6 months.

## Discussion

Subcutaneous mycosis is a chronic fungal infection of the skin and subcutaneous tissues characterized by tumefaction, abscess formation, draining sinuses, and presence of grains within the abscesses and fistulae.<sup>1</sup> *Madurella mycetomatis* is known to be one of the most common causes of eumycetoma.<sup>2</sup> Adult males with more outdoor activity are the usual victims of this disease, commonly involving areas like limbs and other exposed body parts. Eumycetoma in children is not a common occurrence and the facial involvement has not been yet reported in any of the age group. Clinically diagnosis of mycetoma is made on the basis of presence of sinus tracts, or discharge of characteristic granules<sup>2</sup> however, this was an atypical presentation of maduromycosis marked by the absence of black grains in the pus, or any overlying sinus tracts.<sup>3</sup>

Confirmation of diagnosis of eumycetoma is based on isolation and identification of fungus in a mycology lab.<sup>4</sup> In most of the cases more sophisticated tests are not required and routine fungal culture and histology is sufficient. Techniques such as fine needle aspiration cytology, ultrasonography, Computed tomography, histology or immunodiagnosis are also very helpful in diagnosis.<sup>5</sup> Bone involvement is a major complication and must be investigated by radiology.<sup>6</sup> An X-ray of the orbit and the paranasal sinuses of the child revealed no bone involvements.

Secondary bacterial infections pose a major threat to the patients suffering from eumycetoma subsequently increasing the disability and pain.<sup>7</sup>

Treatment of eumycetoma is difficult and poses a challenge and may include long term antifungal therapy however the relapse rates are higher.<sup>1</sup> Surgical debridement

in conjunction with long-term antifungal therapy is often necessary but in chronic progressive cases amputation may become a last possible option.<sup>1</sup> Voriconazole is among the newer promising extended-spectrum triazoles antifungal agents for the treatment of eumycetomas nevertheless its toxic effects must be taken into consideration before use.<sup>8</sup>

This child was treated with intravenous Amphotericin-B considering the nature of spread of the lesion. She responded well and the swelling subsided within 3 weeks. The child was also given itraconazole to which there was a good clinical response. Timely diagnosis and treatment not only cured the child effectively but also prevented the dreaded complications and possible surgical intervention.

## References

1. Brownell I, Pomeranz M, Ma L. Eumycetoma. *Dermatol Online J* 2005; 11:10.
2. McGinnis MR. Mycetoma. *Dermatol Clin* 1996;14:97-104.
3. Meis JF, Schouten RA, Verweij PE, Dolmans W, Wetzels JF. Atypical presentation of *Madurella mycetomatis* mycetoma in a renal transplant patient. *Transp Inf Dis* 2000; 2: 96-8.
4. Singhvi A, Joshi KR, Sharma JC, Vyas MC. Immunoglobulin profile in mycetoma. *Ind J Pathol Microbiol* 1996; 39: 203-6.
5. Fahal AH. Mycetoma: a thorn in the flesh. *Trans R Soc Trop Med Hyg* 2004; 98:3-11.
6. Develoux M, Dieng M T, Kane A, Ndiaye B. Management of mycetoma in West-Africa. *Bull Soc Pathol Exot.*2003; 96:376-82.
7. Rao GM, Devanandam K, Janaki M, Lakshmireddy K. Unusual sites of mycetoma. *Indian J Surg* 2004; 66:46-7.
8. Mancini N, Ossi CM, Perotti M, Clementi M, Digiolo DB, Schaenman JM et al. Molecular mycological diagnosis and correct antimycotic treatments. *J Clin Microbiol* 2005; 43:3584-5.