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Subcutaneous Phaeohyphomycosis of Scalp Caused by Rhytidysteron Rufulum

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Abstract: ***The term "phaeohyphomycosis" is used to describe cutaneous, subcutaneous and systemic infections caused by a rare group of heterogenous dematiaceous (brown pigment producing) fungi. Rhytidhysteron is a dematiaceous fungus, which has been recently found to be causing human infections. Till date only five cases of infection with Rhytidhysteron rufulum have been reported in the literature. An adult immunocompetent male presented with a subcutaneous nodule over the scalp after an alleged history of trauma. The diagnosis was made by direct microscopy, histopathology and culture. The isolates were confirmed by PCR based its sequencing. The diagnosis is often missed due to lack of knowledge regarding the fungi causing the infections and physicians should have a high degree of clinical suspiscion followed by clinicopathological and microbiological examination. Molecular studies may be required to identify a fungus if growth in artificial culture media fail. This case highlights the importance of molecular techniques for identification of nonsporulating pathogenic fungi. We report a case of subcutaneous phaeohyphomycosis of scalp in an immunocompetent male caused by a dematiaceous fungus belonging to genus Rhytidhysteron.***

Keywords: phaeohyphomycosis, dematiaceous fungi, Rhytidhysteron rufulum, rare fungi, phaeohyphomycosis of scalp

1. Introduction

The nomenclature phaeohyphomycosis was introduced by ajello et al. in 1974. [1] The term phaeo has been derived from the greek word phaios, which means grey and refers to the colour of these fungi in vivo and/or in vitro. [2] Phaeohyphomycosis can be classified into superficial, cutaneous and corneal, subcutaneous and systemic rapidly fulminant illness. [2] Subcutaneous phaeohyphomycosis usually results from the traumatic inoculation of the fungus or following wound contamination and occurs mostly in adult males above 30 years with some degree of immunosuppression. [1]

Though 100 genera have been attributed to cause phaeohyphomycosis [2], the common ones are curvularia, alternaria, bipolaris, exophiala, cladophialophora etc. [2] Recently many rare melanized fungi like Rhytidhysteron spp. have been identified as etiologic agents of phaeohyphomycosis due to the availability of molecular techniques. [2]We present this case of subcutaneous phaeohyphomycosis due to the rarity of the etiological agent and the unusual site.

2. Case Report

A 40 year old male patient presented with an asymptomatic swelling in the back of the head since two years after an alleged history of fall. It was pea sized when it started, and gradually increased to attain the present size. He sustained a cut injury on the back of the head which was left to heal by secondary intention leaving a scar. He had no evidence of underlying diseases or immunodeficiency.

Dermatological examination revealed a soft, smooth, oval, skin coloured swelling of size 7x5cms with a linear scar in the occipital region [Figure 1]. The swelling was uniformly cystic in consistency and freely movable on palpation. Slip sign was negative. Skin over the swelling was pinchable. No warmth/ tenderness. No punctum/discharge. Surrounding skin was normal. No regional lymphadenopathy.

Routine laboratory tests were within the normal range.

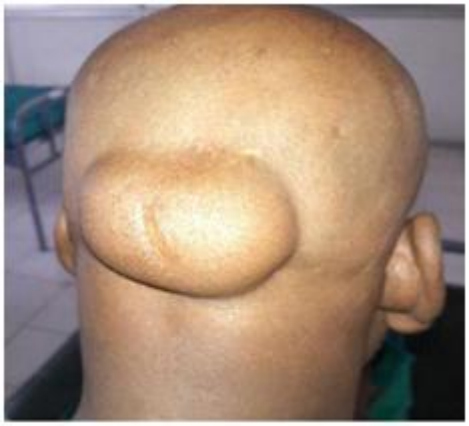

Figure 1: Soft, smooth, oval, skin coloured swelling of size 7x5cms with a linear scar in the occipital region

USG occipital region showed two well defined thick walled cystic lesions below the aponeurium layer of scalp measuring 4.2x1.6cms + 3.2x1.2cms with thick echogenic debris.

CT scan showed normal study of brain parenchyma and well defined soft tissue density lesion in midline in the subcutaneous location.

FNACshowed slender irregularly acute branching septate hyphae with brownish black pigmentation seen in suppurative background with sheets of neutrophils along

with histiocytes and multinucleated giant cells [Figure 2a,2b].

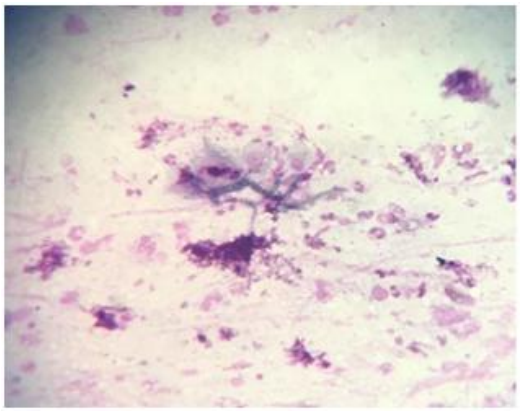

Figure 2 (a): Slender Pigmented irregularly branching septate hypae against inflammatory background (GIEMSA, 100X) - for low power

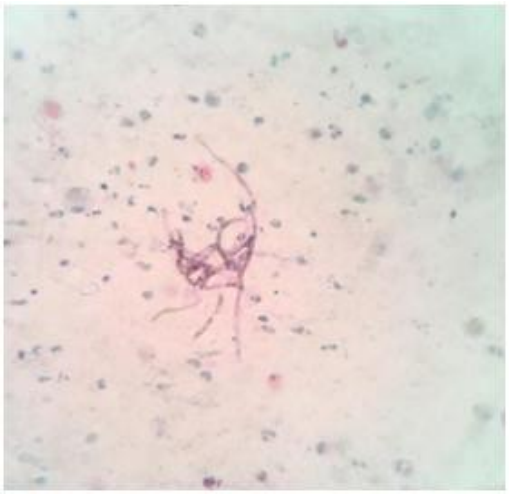

Figure 2 (b): Slender Pigmented irregularly branching septate hypae against inflammatory background (Papanicolaou, 100X) - for low power

The swelling was surgically excised[Figure 3] . Pus was sent for gram stain, KOH examination and culture.

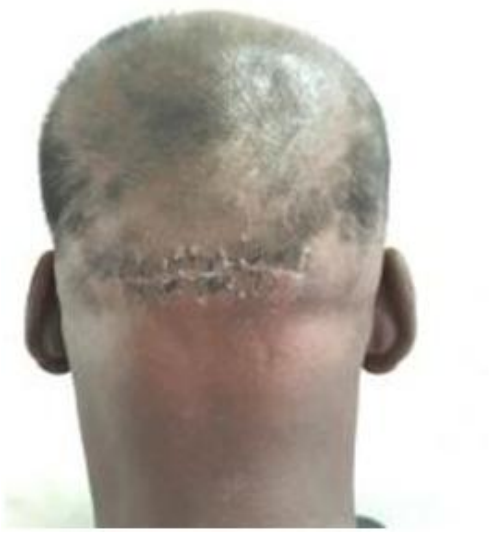

Figure 3: After surgical excision of the swelling

Gram stain showing septate, thick walled, dematiaceous hyphae in the aspirated pus (100x) [Figure 4].

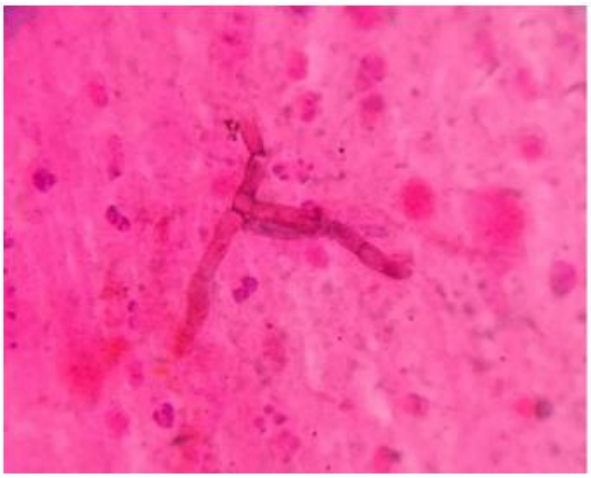

Figure 4: Gram stain showing septate, thick walled, dematiaceous hyphae in the aspirated pus (100x)

Biopsy: It showed cystic lesion which has epitheloid cell granulomas with giant cells and acute inflammatory infiltrates admixed with branching, irregularly septate fungal hyphae with brownish pigmentation.[Figure 5a, b, c]

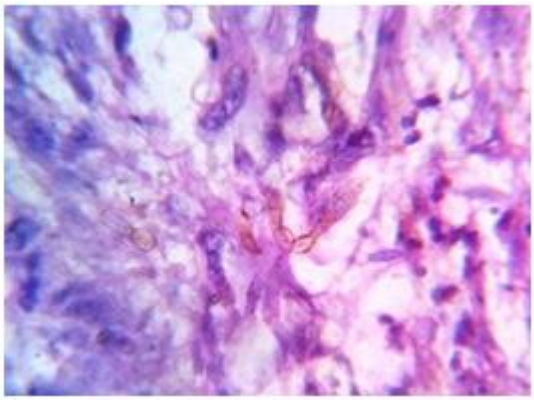

Figure 5(a): Slender Pigmented irregularly branching septate hypae (H&E, 400X) - for high power

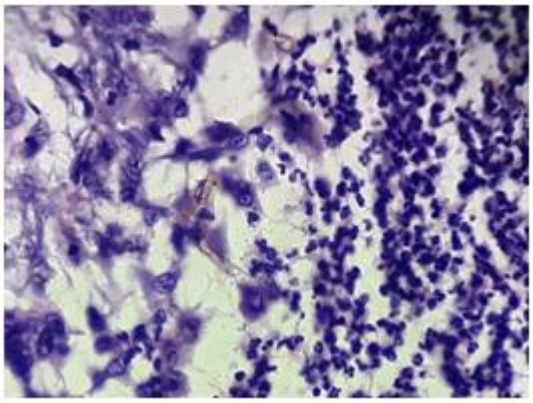

Figure 5 (b): Slender Pigmented irregularly branching septate hypae (H&E, 400X) - for high power

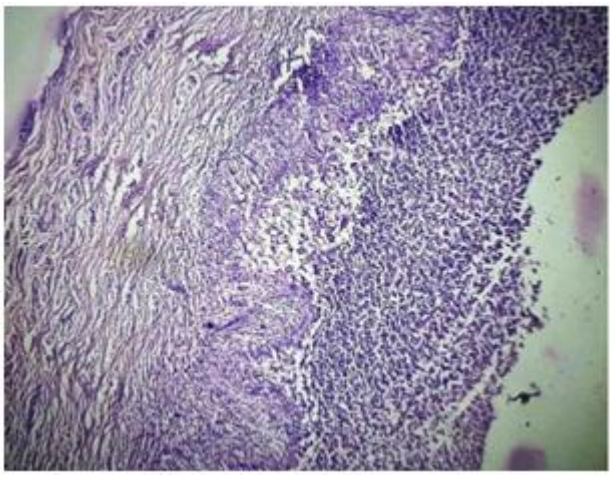

Figure 5 (C): Fibrocollagenous cyst wall with granulomatous inflammation and central abscess formation (H&E, 100X) - for low power

Culture: On sabouraud's dextrose agar showing grayish black, floccose colonies with black reverse of rhytidisteron rufulum. [Figure 6a,b].

Figure 6 (a): Sabouraud's dextrose agar showing grayish black, floccose colonies with black reverse of rhytidisteron rufulum

Figure 6 (b): Sabouraud's dextrose agar showing grayish black, floccose colonies with black reverse of rhytidisteron rufulum

As the culture failed to sporulate, the specimen was sent for molecular analysis and the final identification of the causative organism was achieved by PCR based ITS sequencing. He was started on itraconazole 200mg bd per day and the patient was lost to followup.

3. Discussion

Subcutaneous phaeohyphomycosis is characterized by papulonodules, verrucous, hyperkeratotic or ulcerated plaques, cysts, abscesses, pyogranuloma, non-healing ulcers or sinuses. In india, commonly associated genera are exophiala, phialophora, cladosporium, curvularia, fonsecaea and alternaria.[3]

The case of phaeohyphomycosis described here is peculiar since its etiologic agent is a species of Rhytidhysteron that is recently recognized as a human pathogen. The genus Rhytidhysteron belongs to the family patellariaceae (ascomycota) [4]and has four species: R. Rufulum, R. Hysterinum, R. Opuntiae and R. Dissimile. The most conspicuous and determined morphological character to differentiate species in Rhytidhysteron is ascospore septation. [5]R. Hysterinum has one septate ascospores. [5] R.rufulum has three and R.dissimile five septations. [6] Rhytidhysteron opuntiae has 1–3 (4–5) septate ascospores, with one longitudinal septum in mid cells. [5] The natural habitat of R.rufulum is known to be wood of a wide variety of living or dead dicotyledonous plants. Although worldwide in distribution, it is mainly prevalent in the tropical and subtropical climates. [4]

Infection is probably introduced by traumatic inoculation or inhalation of the etiologic agent. Trauma is responsible for initiating infection in this case.

In the literature, only five cases of infection due to Rhytidhysteron rufulum have been described. Most of the cases of phaeohyphomycosis give history of trauma but the reports without trauma are also not unknown. No explicit history of trauma was given in cases reported before unlike in our case, where there is history of trauma.out of the 6 cases, 4 cases [including our case] were immunocompetent. This suggests that Rhytidhysteron sp. Is not an opportunistic pathogen and can cause infection in both immunocompetent and immunocompromised patients.

The lesions are commonly seen on exposed parts of the body such as feet, legs, hands, arms and back. But in our case, scalp is involved. Scalp involvement is reported in only one case report in 2014 [7]

All the cases reported till date for this fungus are from north to west india only. Our patient belong to tamilnadu.

Laboratory diagnosis includes isolation of fungal hyphae on KOH mount and culture on sabouraud's dextrose agar. [3] Histopathologically, the lesions show the presence of brown-coloured fungal hyphae and yeast-like elements with giant cells or in the background of a granulomatous inflammatory reaction. [1] Molecular studies are necessary to identify a causative fungus if attempts to grow it in artificial culture media fail. [1]In our case, the histological and culture findings supported the diagnosis but final identification of the causative organism was achieved by molecular sequencing.

No specific management protocol exists. Surgical excision alone has been successful in a number of cases, but oral systemic therapy with an azole antifungal agent is frequently used in conjunction with surgery. [8] Itraconazole has been the preferred choice as the invitro susceptibilities to this drug of most strains of dematicaeus fungi is high. [8]

4. Conclusion

Due to the fact that so many different species may be involved, together with the failure of some primary isolates

to sporulate, molecular methods must be included in the diagnostic workup.

Due to the rarity of the disease and lack of large controlled studies of antifungal agents, further studies on antifungal susceptibility profile should be performed to establish management protocol for this group of mycoses.

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