SUBCUTANEOUS PHAEOHYPhOMYCOSIS CYST IN COMORBID PATIENTS:
REPORT OF TWO CASES

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Abstract:

Phaeohyphomycosis is a rare fungal infection caused by a fungus belonging to a heterogenous group of fungal species. They are morphologically pigmented, branched and septate. A 60-year-old male presented with painless swelling of size 8 x 5 x 6.5 cm over the right leg 8 cm below the right knee towards the medial aspect for a duration of 4 months while another male patient of 76 years presented with similar swelling in right leg 5 cm above the medial malleolus towards the medial aspect for the past 2 months. On

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local examination they were firm, non-fluctuant and non-tender. Clinically the first case was diagnosed as soft tissue sarcoma later being lipoma. The cyst was sent for histopathological examination and the cut section showed thick purulent material with foamy histiocytes, neutrophilic infiltration, foreign body giant cells and necrotic debris along with fungal elements. Surgical excision with antifungal treatment is done in both the patients. On follow 6 months follow-up, the fungal infection shows no signs of recurrence.

**Keywords:** Phaeohyphomycosis, fungal infections, pigmented fungi, soft tissue sarcoma, lipoma

**Introduction:**

Of various organisms that cause opportunistic infections in immunocompromised patients, fungi are more common. Among them, pigmented fungi belonging to more than 80 genera and species were found to cause Phaeohyphomycosis, which is a subcutaneous fungal infection common in tropical and subtropical countries. Ajello et al. coined the term “phaeohyphomycosis”. (1) The infection usually follows a traumatic penetration of the fungal elements following a thorn prick, wooden splinter or contaminated soil. Inoculation of fungal elements without being associated to foreign body does not cause infection. (2) It mainly affects skin, cornea, subcutaneous tissue and deep tissues like the brain. (3) The infection typically presents as localised and nontender nodular swellings. The diagnosis is mainly made by fine needle aspiration cytology and histopathological examination which shows dematiaceous yeast like cells, pseudohyphae like elements or pigmented, branched and septate hyphae or in combination of these forms. (3) Fungal elements are identified using special stains like Periodic Acid Schiff (PAS), Gomori Methenamine Silver (GMS) and Fontana - Masson (MF) staining. Here we have retrospectively studied
two cases with comorbidities showing subcutaneous cystic swellings which turned out to be phaeohyphomycosis.

Case History:

Case 1: A 60-year-old male patient, a wage worker, presented to the outpatient department during the month of October, 2019 with the complaints of painless swelling over the right leg 8 cm below the right knee towards the medial aspect that was insidious in onset, gradually progressive, size of 2.8 x 5 x 6.5 cm, irregular in shape, smooth surface and normal surrounding surface. It was warm, firm in consistency, not fluctuant and not tender. No discharge was seen. Patient had multiple right inguinal lymph nodes. Patient had an alleged history of sustained injury to the right leg following a hit with a wooden block 6 months ago following which he noticed the swelling only 4 months later. Other systemic examinations were found to be normal. An MRI of the right leg showed a well circumscribed heterogeneous complex multiloculated soft tissue multilocular nodule. [Fig.1- a, b, c] The swelling was in the subcutaneous plane of leg and the underlying bone did not show any sclerotic or cortical lesions. The above findings lead to a clinical diagnosis of benign soft tissue sarcoma with the differential diagnosis such as fibroma, lipoma, ganglion. Incidental left calcaneal lipoma was found in this patient.

Case 2: A 76-year-old male, a farmer, presented to the outpatient department during the month of February, 2020 with the complaints of painless swelling over the right leg 5 cm above the medial malleolus towards the medial aspect that was insidious in onset, gradually progressive, size of 2.2 x 1.7 x 1 cm, oval in shape, smooth surface. It was not warm, cystic in consistency. It was not fluctuant and not tender. Discharge was not present. No obvious lymph nodes were present. Patient had an alleged history of thorn prick in the right leg 9 months ago following which he noticed the swelling for the past 2 months. Other systemic examinations were normal. On USG, thick walled multilocular collection with thick echogenic debris was noted. The swelling was in the subcutaneous plane of leg without any invasion into the muscular plane and the underlying bone did not show any sclerotic or cortical lesions. Soft tissue
swelling was the clinical diagnosis based on the above findings and the differential diagnosis being fibroma, lipoma, ganglion.

All the blood investigations were normal. Both the patients were a known case of Type - II Diabetes Mellitus and Systemic Hypertension and are on medication.

[Fig.1]: a. Axial, 1b. Coronal, 1c. Sagittal sections of MRI – right leg showing the multi locular cystic swelling

**Pathological findings:**

<table>
<thead>
<tr>
<th>Case no</th>
<th>Clinical diagnosis</th>
<th>Size of the excised specimen (cm)</th>
<th>FNAC</th>
<th>Gross Findings</th>
<th>HPE</th>
<th>Fungal morphology</th>
</tr>
</thead>
</table>
1. Soft tissue sarcoma
   
   2.4 x 4.4 x 6.6
   
   Cellular smears showed numerous multinucleated giant cells, epithelioid cells, cyst and pigmented macrophages. On GMS & PAS stain, yeast and branching hyphal filaments seen in giant cells and in the background of neutrophilic and necrotic debris. No atypical cells seen.

2. Lipoma
   
   2.5 x 1 x 0.6
   
   External surface - grey brown and smooth
   
   Cut surface - cystic areas filled with brownish fluid measuring 1.5 x 1 cm along with the yellowish and grey brown areas.

   Fibro collagenous cyst walls with extensive granulation tissue with sheets of macrophages and foreign body giant cells and necrotic material. In between the giant cells and neutrophilic background, yeast and pigmented, branching and septate hyphae were visualised. On subjecting the tissue sections to special stains like PAS and GMS stains, the septate hyphae with branching that resembled species of phaeohyphomycosis was found [Fig.2 - a, b, c]

   Pigmented, thin septate, branched hyphae with bamboo stem like constrictions along with thick walled spores [Fig.3 - a, b, c]

[Table - 1]: Table showing the pathological findings of the patients

![Image](image1.png)
[Fig.2a, b, c] – H&E staining showing the fibro collagenous wall of cyst with foamy histiocytes (100X)

[Fig.3a – FNAC showing pigmented fungal hyphae with acute angle branching on PAS stain (40X)
Operative findings:

Under aseptic conditions, the soft tissue was easily separated from the surrounding tissue and removed completely. Muscles and the blood vessels of the area were intact. The cyst was completely excised and sent for histopathology examination by immersing it in 10% formalin. Following that primary closure was done. Clinically the infective aetiology of the lesion was not considered, hence the specimen was not sent for microbiological culture.

The postoperative period was uneventful and the patients recovered well. Both the patients were given oral itraconazole (100 mg/d) for two weeks. They remained asymptomatic and no recurrence was noted during follow up at 3 months and 6 months after the surgery.

Discussion:

Around 30 different species of the genus Exophiala, a dematiaceous saprophytic mould from the family Herpotrichiellaceae were found to cause phaeohyphomycosis. (4) These fungal species cause both subcutaneous and systemic lesions. (5). The infection usually occurs following the invasion of the fungal
elements into the subcutaneous tissue after an accidental penetration by a thorn prick or wooden splinter. (6) In the present study both the patients were documented involving lower limbs while some patients may have disseminated disease. (7) Immunocompromised patients are at higher risk to develop this infection however it may rarely occur in healthy individuals. (8) In the present study, two patients with Type - II diabetes mellitus and systemic hypertension for more than 5 years and on treatment developed the infection. The swelling caused by phaeohyphomycosis infection is usually painless and involves regional lymph nodes similar to the findings in the case of the two patients. Histopathology of phaeohyphomycosis shows inflammatory changes like foamy histiocytes, multinucleated giant cells, sheets of macrophages, necrotic material in the neutrophilic background along with the brown pigmented septate hyphae. Gomori - Methenamine Silver and Periodic acid Schiff stains show the fungal wall structure. Fine needle aspiration cytology helps in differentiating the swelling with fungal or neoplastic origin. (9) Since the clinical diagnosis in most cases was soft tissue swelling like fibroma or lipoma the excised specimen was sent only for biopsy and not for culture. The tissue specimen must be sent for routine culture, acid fast bacilli and fungal culture. However, the infection is rare, it is vital in the management of the same in the avoidance of recurrence. Treatment involves simple surgical excision along with antifungals Itraconazole - 200mg, twice daily for two weeks along with monitoring of liver enzymes. Cryosurgery or electrocautery can be done in case of smaller lesions. (10,11) The patients should be followed up for at least 4 - 6 months to look for any recurrence.

Conclusion:

Fungal infections are more commonly seen among immunocompromised patients. They can also be seen in patients with comorbidities. Subcutaneous phaeohyphomycosis is one of the rare types of fungal infections. Here we reported two patients with Type - II diabetes mellitus and systemic hypertension who developed phaeohyphomycosis. FNAC should be done in these cases as it aids in the identification of fungal elements.
eliminating the infective aetiology and also helps in sending the aspirate for culture during the surgery. High suspicion with proper care is necessary for these lesions as early identification and proper treatment prevents recurrence and further complications.

**Declaration of patient consent:**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient had given their consent for their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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**References:**


