

### A Case Report of Intramuscular Abscess Secondary to *Epicoccum nigrum* infection

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#### ABSTRACT

A 36-year-old man with underlying chronic lymphocytic leukemia had left arm swelling for a duration of 3 months. Clinically, the affected arm was swollen, erythematous and tender. *Epicoccum nigrum* was isolated from the culture of the tissue that was obtained intra-operatively. He was treated and responded to voriconazole therapy. To the best of our knowledge, this is the first case of intramuscular abscess as a result of *E. nigrum* infection in an immunocompromised patient.

**KEYWORDS:** *Epicoccum*, *Epicoccum nigrum*, intramuscular abscess, pyomyositis

#### INTRODUCTION

Intramuscular abscess, also known as pyomyositis, is a disease that is characterized by suppurative lesion within skeletal muscles. *Staphylococcus aureus* has been described as the most common causative agent [1]. However, fungi such as dematiaceous moulds have rarely been described as one of its causative agents.

The term 'dematiaceous moulds' (also known as black moulds) are a fascinating and complex group of fungi characterized by the formation of a dark pigment due to the production of melanin in the cell walls of hyphae or conidia or both [2]. These fungi have a worldwide distribution and are commonly isolated from soil and plants. It has been considered as an opportunistic fungi and a rare causative agent in humans. Saprophytes were historically considered as a rare cause of diseases in humans; however, they are now considered as emerging fungal pathogens [3].

Generally, the clinical spectrum of diseases caused by these fungi include eumycotic mycetoma and chromoblastomycosis, predominantly in hosts with normal immune systems, and phaeoophomycosis,

which is most common among immunocompromised patients [4, 5]. These diseases are frequently diagnosed on the basis of a unique histologic picture of the fungus in tissue, followed by isolation of the fungus in culture and morphologic evaluation.

*Epicoccum* sp. is one of the dematiaceous moulds and has been described as an opportunistic fungal pathogen. It has also been described as one of the pathogens responsible for the acute skin infection in an elderly patient [6].

Here, we report a case of a 36-year-old immunocompromised man with underlying chronic lymphocytic leukemia who presented with *Epicoccum* intramuscular abscess in his left arm.

#### CASE PRESENTATION

A 36-year-old Malay male, who is a known case of having chronic lymphocytic leukemia (CLL), had been previously admitted for prolonged intermittent fever 1 week prior to current admission. He was initially treated as having pneumonia and empirically was started on intravenous Tienam 1 g three times daily.

In the ward, the clinician-in-charge had noticed that his left arm was swollen. On further history taking, it was found that he had experienced the swollen arm for the past 3 months prior to admission, and that it was associated with pain and redness. The swelling was actually precipitated by lifting heavy objects. The swelling became worst for a few days while in the ward. There was no pus discharge or complaint of swelling on other body sites.

On examination, he was afebrile, conscious, alert, not toxic looking and vital signs were stable. The swelling on his left arm was measured about 5 x 5 cm, and was erythematous, warm, soft and fluctuated. No discharge of pus was noted from the swollen area. On systemic review, there were crepitations that was heard on his right lower zone. Thus, he was treated as having pneumonia. There was also hepatosplenomegaly, which most probably was due to his underlying CLL. Other systemic review was unremarkable.

In the beginning, in view of the left arm swelling, he was treated as having left arm cellulitis and referred to the orthopaedic team. A left arm ultrasound was done and it showed an hypoechoic lesion extending from the left mid arm to forearm (anterior part within intramuscular measuring about 9.24cm) which was highly suggestive of a left arm intramuscular abscess. Upon X-ray of the left arm and forearm, it only showed a soft tissue swelling with no evidence of bony lytic lesion. He was then treated with intravenous Cloxacillin 1 g four times daily.

Incision and drainage procedure was done on the swelling. Pus aspirate and tissue were collected for microbiology investigations. Tissue biopsy also was sent for further histopathological examination. However, there was no growth obtained from the pus aspirate culture.

In view of the patient's fever still prolonging despite having been given antibiotics, disseminated fungal infection of soft tissue, lung and spleen was suspected. Thus, he was started with oral Voriconazole 400 mg twice daily and intravenous Amphotericin B 20.8 mg per day empirically.

After a 9-day-incubation of tissue culture, dematiaceous mould was noted on culture media. However, histopathology examination only showed

acute non-specific inflammation with no organism seen. The patient was still febrile with spiking temperature after 1 week of combination anti-fungal therapy (Amphotericin B and Voriconazole). Thus, intravenous Amphotericin B was increased up to 38 mg per day and Voriconazole was reduced to 100 mg twice daily. Intravenous Caspofungin 70 mg stat followed by 50mg/day was added to the list of combination medications.

After 2 weeks, the fungal culture was identified as *Epicoccum nigrum*. Amphotericin and Caspofungin were discontinued. Tablet Voriconazole was changed to 200 mg twice daily for 8 weeks. The patient responded well and was afebrile after 4 weeks of Voriconazole therapy. The post-operative wound was clean with no pus discharge or slough.

### Laboratory investigation

Other relevant laboratory investigations were as follows: Full blood count on admission showed leucocytosis (total white cell count:  $16.3 \times 10^3/\mu\text{L}$ ), severe anaemia (haemoglobin: 7.6g/dL) and thrombocytosis (platelet:  $616 \times 10^3/\mu\text{L}$ ).

Full blood picture showed a left shift which is suggestive of being due to an underlying infection and is consistent with chronic lymphocytic leukemia. Septic workup such as blood culture from peripheral and central blood, urine culture, sputum for acid fast bacilli, and melioidosis serology were unremarkable.

### DISCUSSION

Dematiaceous fungi cause a wide range of diseases, ranging from localized to disseminated infection and allergic diseases [7]. *Epicoccum nigrum* is one of dematiaceous fungi that is commonly found in soil, decayed plants, air, and water. It has mainly been described as the cause of hay fever and allergic manifestations such as skin allergy and allergic fungal sinusitis [8]. Besides that, a few reports have reported that *Epicoccum sp.* has been isolated from air sampling [9, 10].

Its role as an opportunistic pathogen has been described earlier in immunocompromised patients with skin allergy [9]. *Epicoccum sp.* is also known as one of the toxigenic moulds due to its capability of producing toxins, which include flavipin, epicorazine

A and B and indole-3-acetonitrile, which has antibiotic-like-substance properties [11]. Furthermore, *E. nigrum* are capable of synthesizing extracellular fungal polysaccharides known as epiglucan [12].

This pathogen has also been reported to be as one of the causative agents in a case of an elderly patient who presented with acute skin infection in the lower part of the leg with co-infection with *Aspergillus flavus*, *Emericella nidulans*, and *Pestalotiopsis sp.* In another case report, *E. nigrum* had been isolated in renal bezoars of a young male patient with history of percutaneous nephrolithotomy to remove his renal stones as the treatment of his renal calculi and severe hydronephrosis [11].

With regards to our case, the underlying immunocompromised state of chronic lymphocytic leukemia has played a role as a predisposing factor in the disease process. The possible explanation on disease acquisition would be that the underlying immunocompromised state has changed the nature of the saprophytic fungi to pathogenic fungi in humans. According to the literature, *E. nigrum* has been found to colonize the nasal sinus [8]. This could possibly explain its route of entry into the lungs and body systems. Other possible explanations pertaining to this case would be primary localized infection which probably has disseminated to the lungs. However, there was no strong evidence to conclude fungal pneumonia in this patient. Besides that, there was no history of skin trauma that provides an entry of this saprophytic pathogen to correlate with the possibility of it being the source of infection.

Choice of treatment and appropriate management will depend on the clinical presentation, whether as a localized or disseminated disease. Localized infection can be treated with just excision alone. Systemic or disseminated illness may be difficult to treat with anti-fungal therapy and is associated with a high mortality rate. Generally, triazoles such as Voriconazole, Posaconazole, and Itraconazole have the most consistent *in vitro* activity against dematiaceous moulds [7].

Combination therapy has been reported to have synergistic effects in some species. However, there is lack of data specifically against *E. nigrum* [7]. Combination therapy was also reported to be effective

in combating the *Epicoccum* infection. A previous case of *E.nigrum* pyelonephritis was successfully treated with combination therapy of Amphotericin B and Voriconazole for 2 weeks duration [11]. Based on the current findings and previous published cases, Voriconazole should be considered as it has been found to be helpful in the treatment of *Epicoccum* infections.

## CONCLUSIONS

In conclusion, the present case showed that *E. nigrum* should be considered as one of the causative agents in an opportunistic infection. Proper and adequate therapy is crucial in the management of the patient.

## Conflict of Interest

Authors declare none.

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