Case Report

Managing Deep Mycoses in Nepal is still a challenge: case report of actinomycetoma, a neglected tropical disease

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Abstract

Deep mycoses are frequent diagnoses in Nepal. Most of the cases remain undiagnosed because of unavailability of specific diagnostic services in our setting. We report a case of deep mycosis, in a 60-year-old male, who presented with multiple nodules and scars on his neck and chest, and was histopathologically diagnosed as actinomycosis.The culture was negative. The patient did not respond to treatment with penicillin initially, however, on starting co-Trimoxazole , the lesions resolved in two months, thus pointing towards diagnosis of actinomycetoma. The patient remained disease free at last follow- up at two years. The case highlights importance of considering actinomycetoma as one of the differential diagnosis of such presentation and also need of developing well set reference laboratory in order to reach to a proper diagnosis.

Key words: Actinomycetoma, cotrimoxazole, deep mycoses, Neglected diseases.

Introduction

Nepal, being agro-based society, it is not uncommon for a dermatologist to see cases of deep fungal infections. Fungal or appropriate tissue culture is rarely done because of its limited availability and hence the diagnosis is based on clinical and histopathological examination most of the time. Because of lack of precise diagnosis, physicians treat these conditions with uncertain outcomes. The commonly diagnosed cases includes sporotrichosis, chromoblastomycosis,and mycetoma in our clinical practice , but the exact data is not available in literature, and this in part could be uncertainity about the diagnosis in absence of tissue culture or DNA amplification tests. We have been treating these cases based on histopathological findings and sometimes we have been fortunate enough to grow the organisms, but very rarely. We report a case of such deep fungal infection, whose diagnosis was directed by clinical presentation and histopathology. The treatment with penicillin and antifungal was unsuccessful initially, and when treated with co-Trimoxazole, in line of actinomycetoma, directed by clinical presentation, the lesions resolved dramatically.

Case report

A 60- year-old immunocompetent male, presented with multiple nodules over upper chest and neck since five years which was recurrent even after complete excision. Biopsy done at other center revealed heavy neutrophilic infiltrates with abscess formation with colonies of branching, filamentous organism positive for Periodic Acid Schiff, Giemsa and Gram stains. Based on clinical presentation and histopathological findings, a diagnosis of actinomycosis was made and patient was started on benzathine penicillin and continued for one month, which did not improve the lesions. Then he was subsequently put on itraconazole for two years and was switched to amphotericin B and lastly to Potassium iodide. The lesions progressed despite treatment for which patient visited our centre.

At our center he presented with multiple nodules over anterior aspect of chest and neck with multiple discharging sinuses (Figure 1). The discharge was pus like and did not contain any granules. X-ray neck and chest did not reveal any underlying bony or pulmonary involvement. A repeat biopsy was performed which was inconclusive and suggested chronic inflammation

only. Culture for both bacteria and fungi were negative. Mantoux test was negative. As the patient had already been treated with penicillin, and antifungal, a therapeutic trial was done with Sulfamethoxazole-Trimethoprim combination based on clinical diagnosis of Actinomycetoma, following which he showed rapid resolution of the nodules within two months (Figure 2). Patient continued the medication for four more months. At two year follow- up there was no relapse and residual deformities.

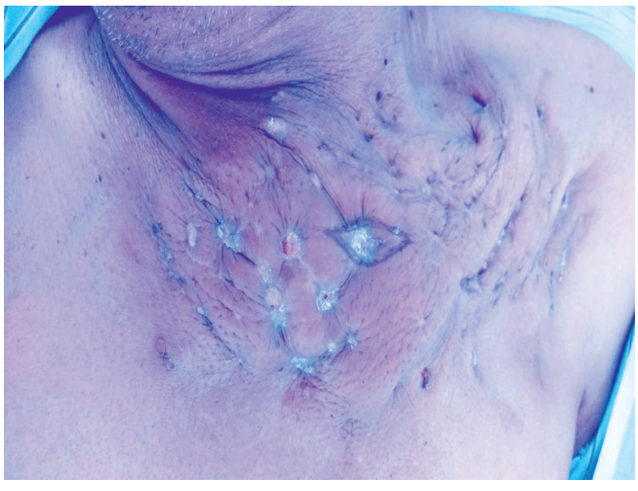

Figure 1 Multiple ulcerated nodules and scars

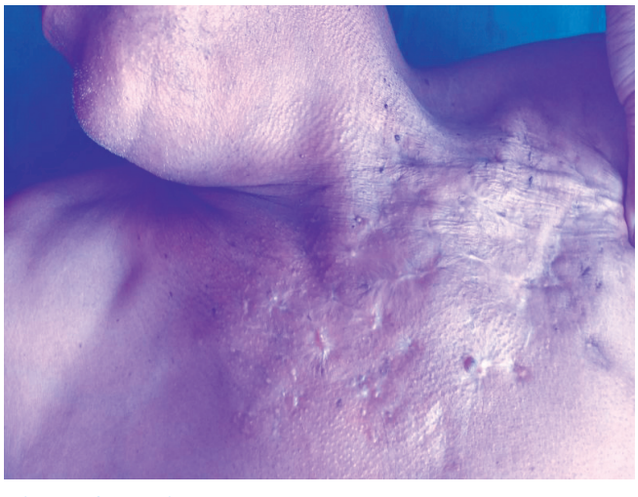

Figure 2 Mutiple healed scars

Discussion

Though we see cases of deep mycoses, there is paucity of literature from Nepal on these neglected tropical diseases, which in part is due to lack of diagnostic facility. Mycetoma , which belongs to one of the deep mycoses, is a chronic and localized granulomatous infection affecting the skin, subcutaneous tissue and bones.[1] They are caused by aerobic bacteria (actinomycetoma) or fungi (eumycetoma). [2] The differentiation is important since the treatment of these etiologies is entirely different and the former responds well to medical therapy whereas the latter also requires surgical intervention. If not detected early, it can cause severe deformity leading to disabilities.[3]

Mycetoma is generally prevalent in many tropical and subtropical areas, mostly between latitudes of 15˚ south and 30˚ north, known as Mycetoma belts.[4] It usually affects those parts that come in contact with soil and foot and legs are affected in more than 80% cases and head and neck is involved in less than 1% [2], which was seen in our case. Mycetoma affecting the trunk is considered to be of poor prognosis because of the proximity of lungs, spinal cord, and viscera.[5]

The clinical charactersistics of actinomycetoma and eumycetoma are similar. However, actinomycetoma is more inflammatory, aggressive and destructive and tends to invade bone faster than eumycetoma .[6] Our patient did not show any underlying bone or viscera involvement. This could be attributed to the fact that he was receiving multiple antimicrobials during the course of his therapy.

Laboratory based diagnostic tools include culture, direct microscopy, cytological, histopathological and immunohistochemical techniques.[7] The diagnosis usually relies on clinical grounds in our setting, which are based on the triad of firm, painless subcutaneous mass, multiple discharging sinuses and purulent or seropurulent discharge that contains grains of various colour and consistency.[2] The clinical findings are confirmed by histopathological examination.

Treatment of both actinomycetoma and eumycetoma is currently based on expert opinion and no standard outlined treatment measure has been given by WHO.[6] Trimethoprim-sulphamethoxazole combination is considered as gold standard for actinomycetomas ,[8] which was also used in our case. Eumycetoma are not amenable to medical therapy alone. In addition to antifungals, preferably Itraconazole, wide local excision of the lesion has to be done if not fully cured by it.[2]

In conclusion, primary infection with mycetoma remains a management challenge. Rapid and reliable molecular techniques are not available in resource limited settings. Its low report and lack of familiarity may predispose patients to misdiagnosis and delayed treatment initiation. Therefore, a high degree of clinical

suspicion is needed for the diagnosis, especially in developing countries like ours.

Conflict of interest: None declared.

References

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