Neurological Manifestations of Mycetoma

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Abstract

Introduction:

Mycetoma is a chronic specific granulomatous progressive and disfiguring subcutaneous inflammatory disease. It is caused by true fungi (Euomycetoma) or by higher bacteria (Actinomycetoma). Mycetoma mainly affects lower limbs, followed by upper limbs, back and rarely head and neck. Mycetoma is mainly transmitted through trauma with infected sharp objects.

Objectives:

To determine the neurological manifestations of mycetoma.

Methodology:

A descriptive cross-sectional community based study included 160 patients with mycetoma seen in White Nile state.

Results:

Almost 160 patients were included in the study, 90% of them were male. Two patients presented with entrapment neuropathy, one presented with proximal neuropathy, one has peripheral neuropathy, one has dorsal spine involvement presented with spastic paraplegia with sensory level, one has cervical cord compression, and one patient has repeated attacks of convulsion.

Conclusion:

Although it is rare, clinicians should highly suspect neurological involvement in mycetoma patients.

Introduction

Carter first recorded mycetoma, also known as Madura's foot, in 1861. It is one of the most overlooked health problems in the world. It is commonly seen in tropical and subtropical region, usually occurs within 15 south to 30 north of the equator.[1, 2] Mycetoma is a chronic specific granulomatous, progressive and disfiguring subcutaneous inflammatory disease. It is caused by true fungi or higher bacteria, so it is usually classified as true fungi and actinomycetes, respectively. [2] The most common causative agents include the fungus *Madurella Mycetomatis* and the actinomycetes *Nocardia Brasiliensis*, *Actinomadura Madyrae*, *Streptomyces Somaliensis*, and *Actinomadura Pelletieri*. Male predominance is a constant findings in Mycetoma. No age is exempted but it commonly affects adults between 20-40 years of

age, who represent the earing members of the society especially under-developed countries. The organisms are usually present in the soil in the form of grains. This infecting agent is then implanted into the host tissue through a breach in the skin reduced by trauma caused by sharp objects eg. thorn in areas where Mycetoma is frequent the habit of going pair foot is common. [3] The trial of painless subcutaneous mass, sinuses formation, purulent and seropurulent discharge that contain grains is pathognomonic of Mycetoma. It may spread to involve the skin and deep structures eg. nerves and bones resulting in destructin, deformity and loss of functions. [4,5]

Neurological manifestations of Mycetoma:

Neurological manifestations of mycetoma are very rare. Known neurological manifestations of Mycetoma include spinal cord myelopathies, brain space occupying lesions, rarely mycetoma can present with neuropathy, myositis, proximal myopathy and cranial nerves involvement. Most of the research on the neurological manifestations of Mycetoma were case reports or case series.

Spine Mycetoma: Mycetoma of the spine leads to destruction of vertebrae and compression of the spinal cord which eventually can lead to quadriplegia or paraplegic [4, 13] .

Head and neck Mycetoma: Head and neck Mycetoma is a rare, severe and debilitating disease, with a low cure rate and high drop-out percentage [15-16-17]. In Sudan 49 cases of head and neck Mycetoma were reported by the Mycetoma research center during the period (1991-2014). Majority were caused by Actinomycetoma. The most common presentation was headache followed by seizure and hemiplegia. CT scan revealed osteo-sclerotic rather than osteolytic lesions.

Problem statement and justification:

In relation to the neurological manifestations of Mycetoma several problems arise including:

Mycetoma SOLs, and myelopathy are the only investigated manifestations until now, while other neurological manifestations like peripheral neuropathy, proximal myopathy, and cranial nerves involvement were not investigated. This problematic scene points to the importance of conducting a cross sectional study to determine the different varieties of neurological manifestations of Mycetoma.

Objectives:

To investigate the presence of neurological manifestations in Sudanese Mycetoma Patients.

Methods And Materials

Study design:

Descriptive cross sectional community based study.

Study area: White Nile State, 250 kilometers to the south of Khartoum.

Study population and Sampling: 160 patients with Mycetoma. The patients gave verbal consent to participate in this study

Data collection methods

Data was collected using Standardized questionnaires including clinical history, neurological examination, and investigations (urine, complete Haemogram, Blood Urea, Serum Creatinine, Spinal MRI, Brain MRI, CPK, Muscle biopsy, Nerve Conduction Study and EMG).

Data management and analysis

By using SPSS (statistical package for social science) software (version 20).

Results

Out of 160 patients with mycetoma 95% were male and 5% were female. Age distribution of our studied group ranges from 18 years to 78 years (80% of our patients are between 20–40 years). All of our patients are farmers. All of our patients are of low socioeconomic status and health education level. Lower limbs were affected in 80% of our patients, followed by upper limbs 15% while the trunk, back, neck and head constituted 5%. Almost 95% of the patients had painless lesions. 70% of our patients had history of discharge containing grains and the color of the grains was mainly yellow. One patient presented with entrapment neuropathy, one presented with proximal neuropathy, one patient has peripheral neuropathy, one patient has dorsal spine involvement presented with spastic paraplegia with sensory level, one of our patients has cervical cord compression, while one patient has repeated attacks of convulsion due to tumor-like mass caused by fungal infection affecting the right cerebral hemisphere.

Discussion

Like what was mentioned in the literature, our study showed that males were affected more than females because mycetoma infection commonly affects farmers, field labourers and herdsmen whose occupation involves direct contact with the soil. [6] Although no age is exempted from the infection with mycetoma, the majority of our patients range from 18–45, this is similar to what was reported by other researchers worldwide. [6] Foot was found to be the most predominantly affected part of the body, followed by the hand and rarely other parts of the body like thigh, trunk, back, neck and the head. [7] Considerable number of our patients (90%) had an obvious history of local trauma. The characteristic triad of a progressive painless subcutaneous swelling at the site of previous trauma as well as the nodules may suppurate and drain through multiple sinus tracts and these sinuses may close transiently after discharge during the active phase of the disease. Fresh adjacent sinuses may open while they are connected with each other, with deep sterile abscesses and with skin surfaces. Mycetoma is usually painless in nature, it was suggested that Mycetoma produces substances that have anesthetic action. [8] At a late stage of the disease the pain may become negligible due to the nerve damage by the tense fibrous tissue reactions, endarteritis obliterans or poor vascularisation of the nerve. Pain may

be produced by the expansion of the bone with Mycetoma granuloma and grains or it may be due to secondary bacterial infection. For unknown reasons the tendon and the nerves usually are curiously spared until very late in the disease process, this may explain the rarity of neurological and atrophic changes even in patients with long standing Mycetoma. One of our patients presented with entrapment neuropathy, nerve conduction study showed an evidence of focal demyelination of the median nerve at the wrist and mild acute denervation in the abductor brevis muscle, consistent with carpal tunnel syndrome, there is no obvious explanation but it may be due to the damage caused by circulating immune complex.[9,10,11] Proximal myopathy was observed in one patient,(creatinine phosphokinase was very high,muscle biopsy showed an evidence of myositis and electromyogram study confirmed the present of myopathy),the myopathy most probably due to damage of the muscle with Mycetoma and coexisting secondary bacterial infection or it may be due to circulating immune complex deposition as part of a local immune response to mycetoma infection. One of our studied group presented with spastic paraplegia due to direct destruction of the two dorsal spinal vertebrae by Mycetoma.

One of our patients is A 65 year's Sudanese female presented with upper and lower limb weakness; this was preceded by multiple discharging sinuses on both sides of the neck. The grains expressed through the sinus were yellow in colour. Some of the sinuses closed transiently after discharge. First the condition was painless then the patient experienced neck pain. Clinical examination showed an evidence of spastic quadriplegia, while local examination of the neck showed that the skin over it become attached and stretched, smooth, shiny and there are areas of hypo and hyperpigmentation. There are subcutaneous masses with discharging sinuses; some of the old ones were healed completely. There is an area of local hyperhidrosis confined only to the site of the lesion and the skin around it. Cervical x-ray showed evidence of soft tissues swelling and destruction of cervical vertebrae C4 and C5. Cervical MRI showed extensive neck pyomyositis, epidural extension and cord compression. The primary symptoms of a tumor, sinuses, and grains flecked discharge provided enough information to diagnose Mycetoma although the species of fungi at the root of infection is identified by staining the discharge grains and inspecting them with microscope.

Like what was mentioned in the literature mycetoma commonly affects the foot and rarely affects the head. [12,13]

One of our patients is a 45 years Sudanese male working as a farmer presented with progressive left sided weakness, both upper and lower limbs, this was associated with severe headache, nausea and vomiting. During the course of the disease he experienced three attacks of generalized convulsion. He claimed that he had a history of discharge containing yellow grains from a sinus in the temporal region of the head, also he mentioned that he has no history of local trauma at the mycetoma site. The patient has had the disease for five years. On clinical examination he was unwell, not pale, jaundiced or cyanosed. Systemic examination revealed no abnormality,the abnormalities were confined to the central nervous system where he has left sided hemiparesis, Examination of the head showed both active and healed sinuses, yellow grains discharged from the sinuses were noted, local hyperhidrosis around the mycetoma lesions were detected, also he has cervical lymphadenopathy, the regional

Lymph nodes were tender and attached to the skin, brain MRI examination showed intracranial extension of the disease. Although bloodstream spread in mycetoma is rare, it was reported and in such a situation the skin and subcutaneous tissue were normal.[14, 15] Spread along the lymphatics to the regional lymph nodes can occur especially with actinomycetoma. Intracranial mycetoma is associated with serious complications and poor outcomes.[16,17]

Conclusion

Although it is rare clinicians should highly suspect neurological involvement in mycetoma patients. Mycetoma infection(wither bacterial or fungal) can cause peripheral or central nervous system damage(At the level of formation of papule and discharging sinuses which can lead to entrapment neuropathy, muscle involvement by the mycetoma itself or coexist bacterial infection, direct destruction of the bone which can cause nerve damage or cord compression. A rare manifestation due to spread of infection from the skull to the brain causing hemiplegia and convulsion.

Declarations

Declaration of Conflicting Interests

The authors declared no potential conflicts of interests with respect to the authorship and publication of this article.

Ethical Considerations

Ethical approval was obtained from the State Ministry of Health, Khartoum, Sudan

Availability of data and materials

The materials datasets used and/or analyzed during this study are available from the corresponding author on reasonable request.

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Author contributions

All authors participated in planning the study, data collection, results and discussion sections.

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