

# Mycetoma of the Neck: An Extremely Rare Presentation of a Rare Disease

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**Abstract:** *Mycetoma is an uncommon chronic granulomatous subcutaneous infection. The disease is commonly seen in the foot and uncommon to involve other parts of the body. Mycetoma of the head and neck is a rare entity. Head and neck mycetomas are associated with increased morbidity and even mortality if not treated. Ultrasonography is a non - invasive method which helps in early diagnosis of the disease. Here we present an extremely rare case of mycetoma of neck. A 58 - year - old male presented with a painful swelling in the anterior aspect of the neck with a sinus discharging seropurulent pus for 15 months. Ultrasonography (USG) of the neck revealed the features of mycetoma and the diagnosis was confirmed on histopathology. Fungal mycetoma of the head and neck region is a rare condition. The diagnosis is generally made when the lesions are advanced. Early diagnosis by ultrasonography or cross - sectional imaging leads to better prognosis and minimizes the number of surgeries required.*

**Keywords:** Mycetoma; neck; Ultrasonography

## 1. Background

Mycetoma is a unique neglected tropical disease. It is endemic in tropical and subtropical regions, also known as the “Mycetoma belt” (Sudan, Somalia, Senegal, India, Yemen, Mexico, Venezuela, Colombia, and Argentina). Mycetoma is commonly seen in the foot. The disease is less common in other parts of the body and is even rarer in the neck. In the neck, anterior neck involvement is very rare as compared to the posterior neck involvement along with the skull base and occipital region. Head and neck mycetomas are associated with increased morbidity and even mortality if not treated early (2). Ultrasonography helps in early detection of the disease and leads to a better prognosis. Here we present a rare case of mycetoma of the neck in a 58 - year - old male.

## 2. Case Report

A 58 - year - old male, farmer by occupation, presented with the complaints of a soft, painless, gradually increasing swelling (Figure 1) with a discharging sinus (Figure 2) in the front of the neck, after a trivial trauma to the neck 15 months back. There was no history of pain, fever, weight loss/weight gain, palpitations, hair loss or giddiness. No history of diabetes or hypertension. No other comorbid conditions were present. Physical examination revealed a soft, boggy, non - tender lump in the anterior aspect of the neck with a sinus discharging pus. All haematological and biochemical parameters were normal. The patient has taken treatment with antibiotics before presenting to our hospital.

Ultrasonography (USG) of the neck revealed multiple conglomerate hyperechoic lesions with hyperechoic centres (Figure 3) suggestive of a typical dot in circle sign (Figure 4).

These lesions are located in the subcutaneous tissues of the anterior neck. Thyroid gland is not involved by the lesions. Based on these findings, diagnosis of mycetoma was given. FNAC was done from the discharging sinus, which showed nonspecific inflammatory changes. Later biopsy was done which demonstrated sulfur granules in H & E stain (Figure 5) and Gomori methenamine - silver stain showed the presence of numerous thin filamentous mycelia radiating outwards (Figure 6). The granules are composed of branched, Gram - positive filaments of *Actinomyces*.

## 3. Discussion

Mycetoma is a chronic, localised, gradually progressive granulomatous subcutaneous infection characterised by subcutaneous swellings and formation of discharging sinuses. It can be caused by true fungi (eumycetoma) or by the bacteria *Actinomyces* (*Actinomycetoma*) (2) (3). This disease is known to occur in the farmers and in the rural population with low socioeconomic status especially in the tropical regions. Mycetoma is generally seen in the extremities after a trivial trauma as extremities are prone to thorn injury in the fields (1).

Mycetoma commonly affects adults in the age group of 20–40 years, with men being more commonly affected than women with a ratio 3.5: 1 (4).

The disease mainly affects the feet as they are more prone to trauma in comparison to other organs in the body such as the hands, head, neck, chest, shoulders and arms (5). On a series of 50 cases of mycetoma involving the other organs, reported by Dieng et al. only 1% involves the head and neck (6) (7).

Patients usually present with painless lumps and discharging sinuses. The pathological process includes the formation of abscesses, draining sinus tracts, fistula formation and can

progress to severe deformity and disability if not treated (5). Another characteristic feature of mycetoma is the formation of aggregates of the organism (grains) in the tissues. Presence of these black grains in the discharge is characteristic of fungal mycetoma (6) (8). Head and neck mycetomas are associated with increased morbidity and even mortality if not treated (2).

Radiographs may be normal in the early stages of the disease or may show soft tissue swellings. In chronic cases, the infection may spread to the bones and show periosteal reactions, osteoporosis, sclerotic or lucent lesions on radiographs.

Ultrasonography is a non - invasive, readily available method which helps in the early diagnosis of the disease. USG shows multiple round hypoechoic lesions containing hyperechoic centres suggestive of typical “dot - in - circle” sign. These hyperechoic centres correspond to the grains (5).

Other investigations that can help in the diagnosis include MRI which may also show “dot - in - circle” sign.

CT helps in the better delineation of the bone involvement.

Early diagnosis and treatment are important to avoid complications and to decrease the morbidity. The treatment is mainly surgical in case of fungal mycetoma and is medical in case of actinomycotic mycetoma (6) (9).

#### 4. Conclusion

Fungal mycetoma of the head and neck region is a rare condition. The diagnosis is generally made when the lesions are advanced. Early diagnosis by ultrasonography or cross - sectional imaging leads to better prognosis and minimizes the number of surgeries required.

#### Figure



**Figure 1:** Front view shows swelling over the neck



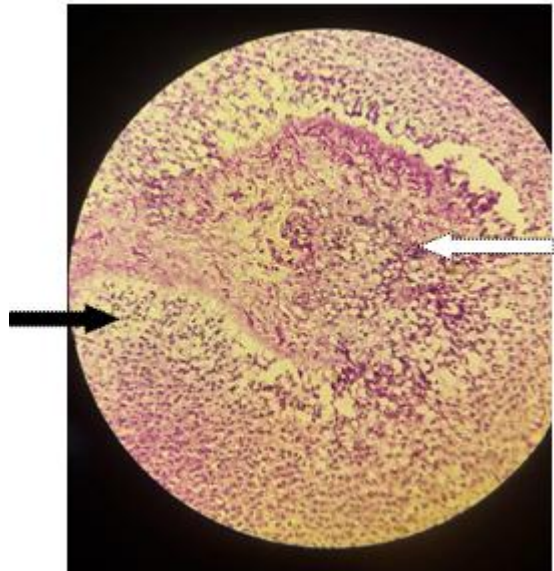
**Figure 2:** Swelling over the neck with a discharging sinus (white solid arrow)



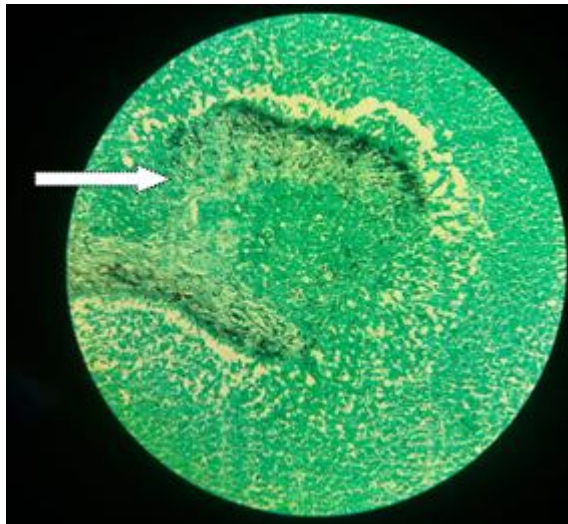
**Figure 3:** USG of the neck shows conglomerate hypoechoic lesion with multiple hyperechoic lesions within (white solid Arrow)



**Figure 4:** USG of the neck shows typical dot in circle sign (white solid arrow)



**Figure 5:** H&E stain demonstrating sulfur granules (white solid arrow) from a sinus/abscess in the neck. The granules are composed of branched, Gram - positive filaments of *Actinomyces*. Surrounding the abscess are inflammatory infiltration of neutrophils (black solid arrow)



**Figure 6:** Gomori methenamine - silver stain showing the presence of numerous thin filamentous mycelia radiating outwards (white solid arrow)

### Abbreviations

USG: Ultrasonography; CT: Computed tomography; MRI: Magnetic resonance imaging; FNAC: Fine needle aspiration cytology;

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