

# Reconstructive Surgery for Mycetoma: Is There a Need to Establish an Algorithm?

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**Background:** Mycetoma is a chronic, progressive, disfiguring, and destructive disease. It caused by a variety of microorganisms including fungi and higher bacteria. It is primarily an infection of the skin and soft tissue, most frequently affecting the lower extremity and the hand and spread through fascial planes and lymphatics. **Methods:** Current medical and surgical management are still inadequate and the recurrence rate is high with severe disabilities. **Results:** This review describes some reconstructive techniques that were performed to address essential aspects with regard to mycetoma surgical management that include coverage of large skin and soft-tissue defects left after local excisions, enhancing the rate of chronic mycetoma wound healing, and preservation or restoration of functional status of the affected limbs. **Conclusion:** These applied techniques—which had acceptable preliminary outcome—have to be considered by the surgeons dealing with mycetoma to improve the functional and cosmetic outcomes and to minimize tremendous morbidities and disabilities that are associated with this neglected disease. (*Plast Reconstr Surg Glob Open 2019;7:e2197; doi: 10.1097/GOX.00000000002197; Published online 24 April 2019.*)

# **INTRODUCTION**

Mycetoma is a chronic granulomatous inflammatory disease which is endemic in tropical and subtropic regions.<sup>1</sup> It is caused either by actinomycetes or fungi; in vivo these cause filamentous grain containing chronic granulomata which form sinuses and reach the skin as a purulent discharge.<sup>1-3</sup> Although rarely fatal, mortality can be secondary to abscess formation, cellulitis, osteomyelitis leading to sepsis; the latter is generally caused by superimposing bacterial infections. The foot is the most common site, but the condition can present in a variety of locations with the main complication being bony involvement. This neglected disease is important to plastic surgeons involved in Global Heath Initiatives, as its pathogenesis causes significant disfigurement and disabilities. Patients are mostly rural workers who are unable to access specialist services until such time that there are massive deformity and challenging disabilities.<sup>1</sup> This article aims to present an overview of mycetoma and describes some reconstructive techniques that were successfully performed for various mycetoma lesions.

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# **PATHOGENESIS**

Mycetoma infection can be caused by a variety of fungi or bacteria.<sup>2,3</sup> If secondary to fungi, it is known as eumycetoma—if bacteria it is referred to as actinomycetoma.<sup>2</sup> The most common causative agents include the fungus *Madurella mycetomatis* and the bacteria *Nocardia brasiliensis*, *Actinomadura madurae*, and *Streptomyces somaliensis*.<sup>4</sup> There is traumatic inoculation of the causative organism cutaneously often secondary to innocuous injury.<sup>5</sup>

# **RISK FACTORS**

Causation is not fully understood.<sup>5</sup> A multitude of risk factors have been described including environmental exposure to pathogenic organisms, genetic predisposition to infection, and immunosuppression.<sup>5–9</sup> It predominantly affects adults between 20 and 40 years old, with preponderance to those with contact to the land (students, farmers, and herdsmen).<sup>5</sup> It is agreed that the disease is not contagious between humans or from animal to human.<sup>5</sup> It is considered as a neglected tropical disease by the World Health Organization.<sup>8</sup>

# **EPIDEMIOLOGY**

Mycetoma is distributed in tropical and subtropical regions.<sup>4</sup> It prevails in the mycetoma belt, which stretches from 15° South to 30° North of the Equator<sup>4</sup> (Fig. 1) and includes Sudan, Senegal, Yemen, India, Mexico, Venezuela, and Colombia, with the highest prevalence reported in Africa, with Sudan thought to be the most endemic coun-

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Fig. 1. Mycetoma distribution worldwide.

try in the world.<sup>1</sup> Published prevalence rates are largely estimates and not derived from population-based studies and do not reflect the magnitude of the problem in endemic regions.<sup>4</sup>

# **CLINICAL FEATURES**

The triad of painless subcutaneous mass with multiple sinuses discharging seropurulent fluid containing mycetoma grains is pathognomonic (Fig. 2).<sup>5</sup> During the active phase of the disease, these sinuses discharge grains of variable consistency and size whose color indicates the causative agent.<sup>5</sup> Table 1 illustrates essential clinical and radiological features of eumycetoma and actinomycetoma. In endemic areas, mycetoma should be included as a differential diagnosis of any swelling.<sup>1,5</sup> Differential diagnoses are listed in Table 2. The body parts that contact soil during daily activities are the most frequently affected, with the lower limb accounting for 82% of cases.<sup>1,9</sup> The prevalence of mycetoma in limbs and the often-sizable defects create the need for significant involvement from plastic and reconstructive surgeons, as it can have dire consequences which are challenging to treat. The lack of medical and health facilities, and financial constraints compounded by poor health education in endemic areas meant that most of the studied patients presented late with advanced disease.<sup>10</sup> Mycetoma is recognized to cause several disabilities and deformities,<sup>11-19</sup> and it could be fatal on rare occasions (eg, cranial mycetoma).<sup>5</sup> Rare sites include



Fig. 2. Pathognomonic triads of mycetoma.

abdominal wall, chest wall, spinal cord, facial bones, mandible, paranasal sinuses, oral cavity, the eye, perineum, vulva, and testes.<sup>20–27</sup>

## **Radiological Investigations**

X-rays and ultrasound allow for an assessment of progression of disease. Early features show as a dense shadow or several scattered shadows similar to a granuloma<sup>28</sup> with progression of the disease, and the cortex can be compressed by the granuloma until many punched-out cavities appear.<sup>28</sup> Grains have specific ultrasonic appearances which allow differentiation between eumycetoma and actinomycetoma.<sup>29</sup> Magnetic resonance imaging is used to assess the extent of bony destruction, soft-tissue involvement and might show a pathognomonic "dot-incircle" appearance that indicates the presence of grains.<sup>30</sup>

### Cytological and Histopathological Investigations

Fine needle aspiration cytology allows for confirmation of the diagnosis. It demonstrates a chronic granulomatous reaction with a purulent centre and allows different grains to be observed. Various stains (most commonly hematoxy-lin and eosin) can be used to identify the causative organism and host-tissue reaction.<sup>31,32</sup> Serological and molecular tests are also proved to be valuable in diagnosis with acceptable sensitivity and specificity rates.<sup>33–35</sup>

Table 1.	Illustration of	fessential Clinic	al and Radiolo	gical
features	of Eumycetor	na and Actinomy	/cetoma	

Features	Eumycetoma	Actinomycetoma
Causative	Fungi	Bacteria
agent	Gram positive	Gram negative
Clinical	Large	Smaller
features	Well defined	Numerous
	Capsulated	Poorly defined margins.
X-Ray find-	Single or multiple thick-	Grains demonstrate distinct
ings walled cavities, without hyper-reflective acoustic enhancement		hyper-reflective echoes
Invasion	Less common + late bone invasion	More readily + early bone invasion

# Table 2. Differential Diagnoses of Mycetoma

#### **Differential Diagnoses**

Cutaneous tuberculosis Non-tuberculous mycobacterial infections of the skin Osteomyelitis Folliculitis Actinomycosis Chromomycosis Sporotrichosis Elephantiasis Blastomycosis of the skin Dermatophyte pseudomycetomas Botryomycosis Sarcoma Kaposi sarcoma Podoconiosis Foreign body granuloma

#### **Medical Management**

The treatment and outcome depend mainly on the causative organism, the site, and extent of disease at presentation.<sup>1,5,9</sup> Current medical treatment for mycetoma is generally inadequate; moreover, medications can be expensive with many side effects.<sup>36</sup> Multidrug therapy is recommended to avoid drug resistance and enhance the eradication of disease.<sup>36</sup> A multitude of azole antifungals are available for treatment of eumycetoma.<sup>36</sup> The optimal duration of antimicrobial therapy for treatment of eumycetoma is uncertain and can involve long-term therapy in some cases going up to 12 months with bony involvement. Patients should be followed for at least 2 years after discontinuation of antimicrobial therapy before declaring a cure.<sup>36</sup>

### **Surgical Treatment**

#### Local Excision

Surgical management is dependent on the size of the lesion. Small lesions (<5 cm) without bony involvement require wide local excision with concomitant antimicrobial therapy.<sup>36</sup> Excision should always be undertaken under general anesthetic with a bloodless field using a tourniquet-a bloodless field is paramount to prevent bursting of the capsule—as bursting the capsule can lead to increased rates of recurrence.<sup>36,37</sup> Repeated debridement is required for massive infected disease.<sup>38,39</sup> The Mycetoma Research Center in Sudan have developed guidelines for amputation in cases of mycetoma listed in Table 3.40 Authors have utilized repeated local excisions to prevent amputation,<sup>41</sup> but amputation can be a lifesaving procedure in selected patients and rates range from 10% to 25% in most series.<sup>39,42,43</sup> In Sudan, a homeland of the disease, mycetoma is the third most common cause of amputation after diabetes-related complications and

#### Table 3. Guidelines for Amputation 40

land-mine injuries.<sup>44</sup> Interestingly enough, there is less phantom pain in patients with mycetoma after amputation, and this allows more successful prosthetic fitting as there is an adequate blood supply.<sup>44</sup> The recurrence rate varies from 25% to 50%<sup>5,36</sup> There are multifactorial causes of this high recurrence rate that include widespread disease along the fascial planes, inadequate excisions, lack of compliance with medication, and lack of health education.<sup>36,39</sup>

# RECONSTRUCTIVE SURGERY FOR MYCETOMA

Reconstructive surgery for mycetoma was successfully performed worldwide.<sup>45–50</sup> However, published work on reconstructive surgery for mycetoma is essentially limited in both number and case volume. Various reconstructive surgeries were applied in selected patients and had satisfactory preliminary outcomes.<sup>45–50</sup>

Imaging modalities now would enable accurate detection of the extent of the lesion.<sup>51</sup> Deep structures and bone involvement can be identified and an adequate surgery with safe margins can be well-planned.<sup>45</sup> A radical approach has been suggested for an adequate resection of mycetoma with applying of Enneking Principles for Oncologic Surgery.<sup>52</sup> The rationale of this approach is to manage mycetoma lesions as malignant tumors due to the high recurrence rates after a local resection,<sup>46</sup> although mycetoma is a benign disease with no reported malignant or premalignant potentials.<sup>5</sup>

A series of 26 patients with eumycetoma who underwent wide surgical excisions followed by reconstructive surgeries was reported from Sudan (a homeland of the disease) from 3 referral plastic surgery centers in Sudan.<sup>45</sup> The size of lesions encountered ranged from 8 to 20 cm. The sites of lesions include the foot, knee joint, leg, hand, and gluteal region. A wide local excision was performed for large-sized lesion of >8 cm with a safe margin of 0.5-2.2 cm to eradicate the disease.<sup>45</sup> As large skin/soft-tissue defects were left after excision, various reconstructive techniques were applied, ranging from split-thickness skin grafts (STSGs) to local or advanced flaps (eg, V-Y advancement flaps, gastrocnemius flap, dorsalis pedis flap, and lateral calcaneal artery flap). These procedures allowed closure of the skin and soft-tissue defects and hence the length of hospital stay was reduced. The patients were followed up over 9-12 months and all had good healing and return to ordinary activities but 1 patient had recurrence. According to our initial experience, we do recommend a reconstructive surgery after wide local excision (WLE) for larger-sized mycetoma lesions and when there are difficulties to close the skin primarily.

Reconstruction of a foot defect created by local excision of mycetoma lesion was reported from Israel,<sup>46</sup> where a right foot defect following an adequate excision of mycetoma was reconstructed by a free tensor fasciae latae musculocutaneous flap with a successful tissue transfer. The function of the affected limb was preserved, and there was no recurrence on magnetic resonance imaging during follow-up.<sup>46</sup> Authors justified their reconstructive approach by the fact that transferring a highly vascularized tissue to the mycetoma region-where the infectious process existedafter a radical resection can significantly assist in eradicating residual nests of disease by means of a better delivering of antifungal drugs to the surrounding tissues.46 This newly innovated technique can be effective to reduce recurrence rates. A similar case was reported from Kenya.<sup>47</sup> A solitary left anteromedial leg mycetoma swelling with lytic bone invasion on leg x-rays that led into severe inability to use the limb was successively managed by local excision with reconstruction of the defect by gastrocsoleus flap and STSG in combination with antifungal treatment.<sup>46</sup> The result was complete resolution of the swelling with no clinical and radiological signs of recurrence on follow-up. The patient became ambulant with normal weight-bearing function of his limb. It is important to note that amputation is still considered as a social stigma and has many medical and socioeconomic impacts especially in developing countries that have the greatest burden of mycetoma.<sup>39</sup> Therefore, there is a real need to establish appropriate reconstructive options that would preserve the function of the affected limbs to avoid amputation. Nevertheless, the decision of amputation may be undeniably difficult in some cases as it can be a lifesaving procedure<sup>39</sup> A local V-Y flap was advanced to cover a large gluteal defect following local resection of an eumycetoma in the gluteal region.<sup>49</sup> A latissimus dorsi flap was transferred to cover a large chest wall defect following an excision of mycetoma lesion.<sup>50</sup>

It is interesting to note that STSG after application of topical negative pressure wound therapy (NPWT) to cover skin defects has been successfully described in the literature.53,54 An STSG was used for covering a skin defect after application of topical NPWT to induce the granulation tissue formation following local excision of a Madura foot.53 The lesion involved plantar arch of the right foot and was recurrent and complicated by multiple sinuses formation.53 This modality coupled with medical treatment had resulted in complete disappearance of the lesion at an 18-month follow-up.53 Although graft had developed necrosis, the presence of an adequate granulation tissue in the surgically treated area had facilitated reepithelialization of the wound.<sup>53</sup> The final outcome of the scar was acceptable esthetically and functionally.53 A second case of employing similar technique was reported in Turkey.54 Topical NPWT was used for 2 weeks after local excision of a dorsal ankle Madura and then an STSG was applied.54 It had resulted in complete disappearance of the lesion with full graft take.54 In fact, NPWT, based on the application of subatmospheric pressure has revolutionized the management of chronic wounds complicating burn, chronic infection, exposed bone, and previous wound dehiscence.55,56 This modality facilitates healing by improving the rates of angiogenesis and endothelial proliferation, maintaining the integrity of the capillary basement membrane and reducing microbial burden within the wound.<sup>57</sup> Therefore, it can be a good choice to induce granulation tissue formation and enhancing the rate of healing of chronic mycetoma wounds.<sup>53,54</sup> In addition, it could enhance local inflammatory response and ensure eradication of any residual fungal elements.53 Although the value of this novel technique has not yet been fully proven,<sup>53</sup> it merits attention by workers in the field of mycetoma to examine its efficiency based on larger-scale studies. Secondary bacterial infections represent a constant morbidity with mycetoma.<sup>5</sup> However, the role of reconstructive surgery in reducing the incidence of these secondary infections of mycetoma wounds by covering the skin/soft-tissue defects and enhancing wound healing has not previously been discussed in the literature. This is an important relation to assess in details in future case–control studies.

Therefore, surgical management would supplement medical treatment by debridement or local excisions, therapy enhancing the response to antimicrobial therapy, and can augment functional and cosmetic outcomes by various reconstructive procedures.<sup>58</sup> The use of latter techniques to cover large skin/soft-tissue defects following local excisions, to enhance chronic wound healing rates, and to preserve the function of disabled limbs can be rewarding as shown objectively above, hence there is a genuine need for a consensus on reconstructive approach and formulation on algorithm for reconstructive ladder for the cases that required these management options.

# **CONCLUSIONS**

Mycetoma is a difficult-to-treat disease. A complete cure is not achieved in many patients and recurrence is not uncommon even after adequate treatment.<sup>35</sup> Reconstructive surgery techniques have to be greatly considered by surgeons working in endemic areas. These novel surgical options can be effective in regard to many aspects of mycetoma surgical management. The benefits would include covering large skin and/or soft tissue defects following wide local excisions of massive lesions, preserving or restoring the function of disabled limbs, and enhancing the rate of chronic wound healing.

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