European Journal of Plastic Surgery (2019) 42:385–390
https://doi.org/10.1007/s00238-019-1502-z

ORIGINAL PAPER

Reconstructive surgery for mycetoma: a case series

Mohamed Abdelrahman[1,2] · Irfan Jumabhoy[2] · Eltaib A. Saad[1] · Gamal M. Abdulla[3]

Received: 17 October 2018 / Accepted: 10 January 2019 / Published online: 19 January 2019

Abstract

Background Mycetoma is an under recognised but significant disease endemic in various countries and associated with high morbidity. Treatment is a combination of antimicrobial therapy, with surgery often reserved for refractory or delayed cases where functional sequelae are often ignored. This case series aimed to provide preliminary evidence for the role of reconstructive surgery in treating mycetoma.

Methods Twenty-six cases of eumycetoma suitable for reconstruction post-excision were identified between 2013 and 2016 in three centres in Sudan. The choice of reconstruction was based on consensus of the treating team, and relevant end-points noted including patient satisfaction, mobility and complication rate.

Results Mycetoma lesions affected primarily the limbs (n = 23), with three cases involving the gluteal region. A range of reconstructive options was used including skin grafting (n = 14), local flaps (n = 5) and regional flaps (n = 4). Three cases were closed primarily. No complications were noted, and disease recurrence was not found in any of the patients. Subjective interviewing revealed adequate patient satisfaction with cosmesis, and all patients reached a post-operative mobility status at least equivalent to their pre-morbid state.

Conclusions This study demonstrates the feasibility of reconstructive surgery in selective patients with mycetoma as part of their treatment protocol.

Level of Evidence: Level IV, therapeutic study.

Keywords Mycetoma · Reconstruction · Tropical surgery · Neglected disease

Introduction

Mycetoma is a chronic granulomatous inflammatory disease of the skin and subcutaneous tissue, commonly described according to causative organism as either eumycetoma (i.e. fungal) or actinomycetoma (i.e. bacterial) [1]. The World Health Organisation (WHO) recognises mycetoma as a badly neglected disease, without accurate data on its incidence and prevalence, or any formal programme for prevention and surveillance [2]. Although it occurs worldwide, it is endemic in certain sub-Saharan and South American countries such as Sudan, Somalia, Mexico and Venezuela—the so-called 'mycetoma belt' [1]. Whilst early disease is amenable to cure through a combination of medication and surgery, little attention has thus far been paid to options for reconstructive surgery for what is often a debilitating and disfiguring surgery.

There are more than 70 organisms implicated in causing mycetoma, most of which are soil inhabitants and are transmitted via traumatic inoculation e.g. via a thorn prick [3, 4]. Mycetoma clinically presents with a classic triad of a hard subcutaneous swelling, multiple sinuses with painless seropurulent discharge, and the presence of grains of various colours and sizes (representing colonies of the causative organism) [5]. The foot is the most commonly involved site, but mycetoma may affect virtually any part of the body [6]. There is a male preponderance (sex ratio 3–4:1), and most cases are seen in individuals in their third and fourth decades [7]. Owing to the painless nature of the disease, lack of health facilities in endemic areas, and low levels of patient health education, late disease presentation is often encountered where the infection involves the underlying bone and muscle and is more resistant to medical treatment [8].

✉ Mohamed Abdelrahman
myhrahman@gmail.com

[1] Department of Surgery, Faculty of Medicine, University of Khartoum, 102, 1111 Khartoum, Sudan

[2] Department of Plastic Surgery, Nottingham City Hospital, Nottingham, UK

[3] Department of Surgery, Faculty of Medicine, University of Gezira, Wad Madani, Sudan

Diagnosis of the disease is made in combination with clinical findings and identification of the causative organism through fine-needle aspiration (FNA) cytology. If equivocal, histopathological and Gram stain examination of tissue biopsies, as well as molecular diagnostic techniques, will identify the causative fungal and bacterial species. Imaging techniques such as plain radiographs, ultrasound and magnetic resonance imaging (MRI) are useful adjuncts in characterising the extent of the lesions, including involvement of deeper bony or muscular structures.

Treatment regimes vary widely, but in general comprise a prolonged course of either an antibiotic or antifungal agent, and in the case of eumycetoma patients, surgical excision. Indications for surgery include small, well-localised lesions, in massive lesions to reduce organism load, in medication refractory cases, and to counter certain complications such as secondary bacterial infection and sepsis [9]. However, little consideration has thus far been given to reconstructive options post-excision, nor the associated complications, impact on disease recurrence or overall patient prognosis.

As such, the aim of this study is to characterise the reconstructive surgical options and post-operative sequelae for eumycetoma patients.

Methods

Ethics

This study was approved by the institutional review board. All participants gave their written consent, and individual patient data was anonymised to preserve confidentiality.

Patients

A total of 26 patients were recruited between the period of 2013 and 2016 within three centres in endemic states in Sudan. The inclusion criteria included a formal diagnosis of eumycetoma, confirmed clinically, radiologically and cytologically, in adult patients over 18 years. These patients were deemed to require surgery as part of the treatment strategy, and the most suitable reconstructive option chosen by consensus by a team of plastic surgeons. Exclusion criteria included paediatric patients, those with incomplete data sets or who were lost to follow-up, and those for whom reconstructive surgery was not indicated.

Diagnosis

All patients were diagnosed as eumycetoma based on a combination of factors. Clinical susceptibility was based primarily on residency in an endemic area, history of discharge of black grains and history of previous mycetoma infection as well as supportive physical examination findings. FNA cytology of lesions was performed using 10% potassium hydroxide in order to classify grains, with subsequent culture on Sabouraud's Dextrose Agar 2% and Mycobiotic Agar. If insufficient, histological examination of tissue biopsy was performed using haematoxylin-eosin, Grocott's methanamine silver, Periodic acid-Schiff and Gram-Brown-Brenns. All patients had plan film radiographs taken of the affected region, with further imaging using ultrasound or MRI as needed.

Treatment

All patients were treated according to a standard protocol at the local institutions. This consisted of 200–400 mg/day of Itraconazole, chosen for its comparatively low rate of systemic toxicity and side effects, for a minimum period of 6 months. The decision to then operate was made by the multidisciplinary team. All patients initially underwent wide local excision (WLE) under a regional/general anaesthetic technique, and proceeded on to reconstruction based on the consensus of the operative team. A tourniquet was used to allow for a bloodless field. Generous long incisions were made to facilitate initial excision, with a wide rim of healthy tissue (between 1 and 2 cm) excised as a safety margin. Choice of reconstructive technique was based on the principles of the reconstructive ladder, as well as anatomical site of defect.

Follow-up

The patients were followed-up monthly at the outpatient clinic to assess cosmetic and functional outcomes post-operatively, the presence of any complications, concordance with medical treatment and disease recurrence.

Statistical analysis Data included in this study were analysed using descriptive statistics with the Statistical Package for the Social Services (SPSS), version 20.0.

Results

A total of 26 eumycetoma cases were included in the present study; 21 were male, and 5 were female (male to female ratio of 4.2:1). In nine (34.6%) patients, this was a recurrent diagnosis of eumycetoma, whilst in the remaining, it was their initial presentation. The mean age was 28 years (range 18–56 years). Four patients had a single concomitant comorbidity, with two having two or more comorbidities. The commonest comorbidities included hypertension and type two diabetes mellitus.

The time from onset of clinical signs to diagnosis and medical treatment ranged from 4 to 27 months (mean 13 months). The distribution of eumycetoma lesions were as follows: 9 (34.6%) affecting the foot, 5 (19.2%) affecting the hand, 3 (11.5%) affecting the leg, 4 (15.4%) affecting the thigh/knee, 2 (7.7%) affecting the arm and 3 (11.5%) affecting the gluteal

region. Diagnosis was established through FNA cytological examination in 23 (88.5%) cases, whilst direct microscopy was required in 3 (11.5%) cases. The causative organism was established in 22 (84.6%) cases, with *Madurella mycetomatis* representing the most common organism in 17 (65%) cases. All patients underwent plain radiography of the affected site, with additional imaging performed via ultrasonography or MRI in 8 (30.7%) and 2 (7.7%) of the cases, respectively.

The average time of antifungal therapy prior to operative intervention ranged from 6 to 13 months (mean 8.7 months). Post-WLE, reconstruction was performed using skin grafting in 14 (53.8%) patients (Fig. 1). Local flaps, primarily V-Y advancement flaps, were utilised in 5 (19.2%) patients (Fig. 2), whilst regional flaps were used in 4 (15.4%) instances. These included two patients who underwent a grastrocnemius flap for left thigh and knee lesions (Fig. 3), a dorsalis pedis pedicled flap for a right foot lesion, and a lateral calcaneal artery skin flap. Three (11.5%) patients had their soft tissue defects managed with primary closure after sufficient undermining to allow a tension-free closure.

Follow-up was performed on a monthly basis for the first year. All patients were still concordant with clinical therapy at this time. No complications were reported, with a 100% successful skin graft and flap take. No cases of recurrence were noted up to the time of writing of this report. Wound cosmesis was reported to be at least satisfactory on subjective questioning of the patients, and all patients reached a functional outcome that at least matched, if not bettered, their pre-operative state, as assessed by the specialist physiotherapist (Table 1).

Discussion

To date, and to our knowledge, this is the largest published case series of patients with eumycetoma that have been managed with both medical therapy and reconstructive surgery. In this study, 26 patients with confirmed eumycetoma underwent medical therapy followed by a combined wide local excision and immediate reconstruction. The rationale for selecting eumycetoma patients was based on the relative paucity of effective antifungal therapy for these cases, which often necessitates surgical intervention, compared with actinomycetoma [10]. However, surgery has historically been performed for disease control, with little consideration given to post-operative function or cosmesis.

The higher male to female ratio identified in our study sample is in line with the reported literature, as is the mean age of the patients included [7]. The presence of existing co-morbidities have not been shown to be associated with more severe or atypical forms of mycetoma, however, in this case, were of general interest from an anaesthetic and peri-operative point of view. Similarly, the distribution of lesions in our study, with the majority affecting the foot, is supported by various case series. In fact, over 80% of mycetoma cases are known to affect the limbs, which is possibly explained by the fact that patients in endemic areas often walk barefoot and thus inosculate themselves with the disease [11].

FNA cytological analysis of grain specimens has been shown to be as reliable as formal histopathological assessment of tissue biopsies, given the characteristic colour, size and consistency of individual grains [12]. *M. mycetomatis*, the commonest eumycotic agent, are often large and stain black. Other organisms responsible for eumycetoma include *Madurella grisea, Pseudoallescheria boydii* and *Leptosphaeria senegalensis*. The use of radiographic assessment greater than plain radiographs is generally reserved for cases where there is a strong clinical suspicion of bony or muscle involvement, delineating the anatomy prior to potential surgical reconstruction or in cases of equivocal diagnosis.

Given the varied total treatment duration for eumycetoma reported in the literature, a consensus approach was taken to determine length of antifungal treatment prior to surgery. In principle, the use of antifungal therapy should reduce the acute disease burden, whilst inducing the formation of an adequate fibrous capsule around the lesion and so facilitate surgical dissection [9]. However, this alludes to a larger problem in the treatment and surgical management of mycetoma i.e. a lack of published guidelines and standardised peri-operative treatment protocols. For instance, there is no defined margin at which a WLE should be performed, with examples in the literature varying between 0.5 to over 2 cm [9, 13–15]. In this study, the reconstructive decision was based on our extensive

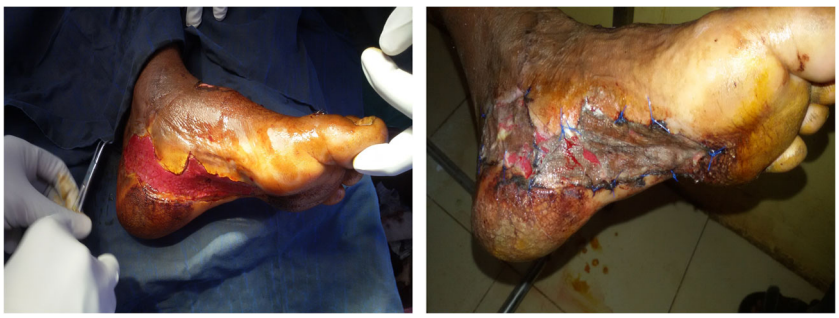

Fig. 1 Right foot mycetoma treated with wide local excision (left) and immediate application of split skin graft (right)

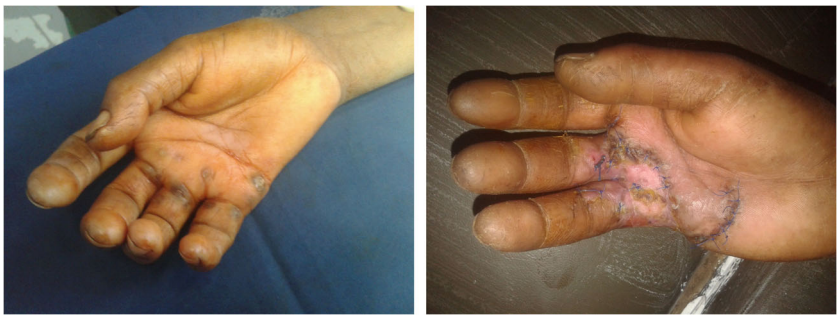

Fig. 2 Right hand mycetoma (left) treated with wide excision and local V-Y flap (right)

experience in dealing with mycetoma patients over the last 25 years [8].

Tamir et al. performed the first published reconstruction for a patient with mycetoma; a 28-year-old patient with a 3-year history of right foot eumycetoma resistant to medical therapy underwent two-stage resection and reconstruction with a free tensor fascia lata (TFL) musculocutaneous flap [13]. More impressively, the patient remained disease-free a year after, and was ambulant on the affected leg. Since then, scant publications have reported varying degrees of success in reconstruction using a range of techniques from skin grafting, to local and regional flaps [14–17]. However, almost none of these reports included a functional assessment post-operatively, and certainly, none included a patient measure of outcome satisfaction.

This is significant for two reasons: firstly, as has been widely noted, farmers and other manual workers are the individuals most commonly affected by this disease, and so the ability to preserve function is integral to their livelihood, which in turn will ease the socioeconomic burden associated with the disease. Limb amputation remains a relatively common surgical treatment and so any effort to preserve limb function with radical dissection and reconstruction will further reduce patient morbidity and the considerable social stigma associated with amputations. This is particularly evident in the four cases of regional flap reconstruction we performed. Weight-bearing status was preserved, and the patients were ambulant soon after, returning to a near normal baseline function within 3 to 4 months. Secondly, patient satisfaction with treatment is likely to result in greater treatment adherence, and hence the reduced likelihood of disease recurrence. There are other posited benefits of reconstruction; Tamir et al. suggested that the transfer of a highly vascularized tissue to the infected region may improve loco-regional blood flow and hence delivery of anti-fungal drugs to the area, thus helping eradicate residual nests of disease [13].

In our case series, there was no reported disease recurrence within the follow-up period, and patients were generally free of surgical complications. This is in contrast with the existing literature, but can be explained by the fact that most cases of surgical excision outside our centres are performed under local anaesthesia and by an inexperienced surgeon in a rural setting with poor surgical facilities. This is one of the main predictors of disease recurrence, along with the presence of

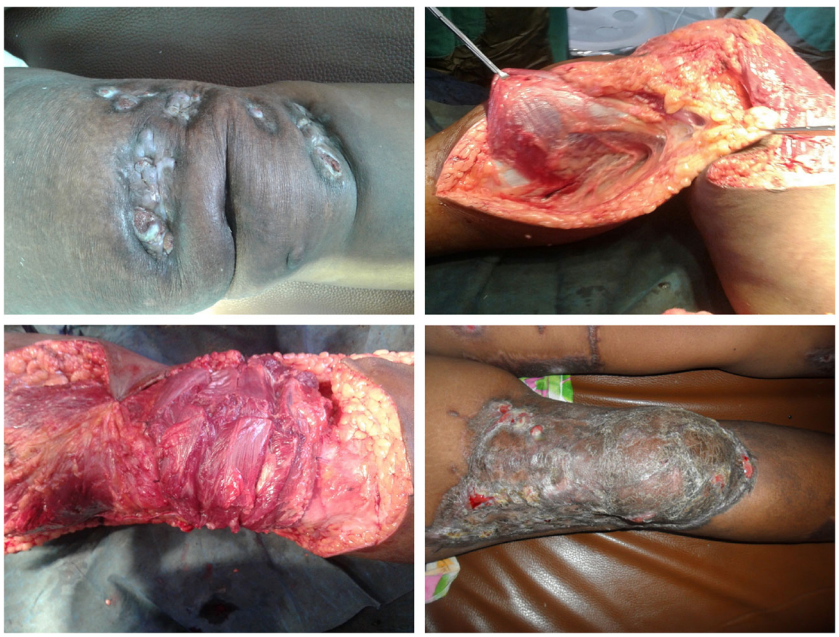

Fig. 3 Right knee mycetoma (upper left) treated with wide excision and regional gastrocnemius flap (upper right). Intra-operative image final flap position (bottom left) and healed defect (bottom right)

Table 1 Summary of the post-operative patient-reported measures

	Strongly agree	Agree	No opinion	Disagree	Strongly disagree	Total
I am satisfied with how my wound looks, after surgery	7 (27.0%)	16 (61.5%)	3 (11.5%)	0	0	26 (100%)
I am satisfied with how my wound feels, after surgery	2 (7.7%)	19 (73.1%)	5 (19.2%)	0	0	26 (100%)
I feel that my function is impaired after surgery	0	0	1 (3.8%)	17 (65.4%)	8 (30.8%)	26 (100%)
I feel that I am able to do at least as much now, compared with before surgery	18 (69.2%)	4 (15.4%)	4 (15.4%)	0	0	26 (100%)
I feel confident in returning to my previous level of function, after surgery	20 (76.9%)	3 (11.5%)	3 (11.5%)	0	0	26 (100%)

Percentages provided in brackets (%)

disease greater than 10 years, the presence of extra-pedal disease, family history, and certain demographic factors such as occupation and geographic location [18].

This case series arguably demonstrates the merits of surgical reconstruction for mycetoma patients. However, despite the first case report of reconstruction being published over 20 years ago, there has been little widespread acceptance of its role. Part of the explanation for this is that mycetoma is endemic in resource-poor countries, where the resources or infrastructure may not permit for reconstruction. However, good medico-surgical management may reduce the recurrence rates of mycetoma, which will inevitably reduce costs associated with prolonged courses of medication, as well as the economic impact of further morbidity. Additionally, as mentioned previously, a lack of consensus on the most suitable reconstructive and peri-operative approach means that routine adoption of reconstruction may be delayed. Within our centres, we are developing a treatment protocol to attempt to address some of the variability in disease presentation, as well as how to successfully match the reconstructive approach.

Conclusion

Mycetoma is a neglected and often underreported disease, associated with significant morbidity and mortality in those countries in which it is endemic. Treatment with antibiotics or antifungal medication is often insufficient in preventing recurrence and is associated with a high dropout rate. Whilst wide local surgical excision has been well established for localised lesions, larger lesions have often necessitated aggressive debulking, often tending towards amputation at the expense of function. Reconstruction, particularly of large skin and soft tissue defects is a viable option to restore both function and cosmesis, as evidenced in this study. However, further work needs to be undertaken to determine the optimal timing of surgery, peri-operative protocol, and ideal reconstructive technique to ensure its wider application.

Compliance with ethical standards

Conflict of interest Mohamed Abdelrahman, Irfan Jumabhoy, Eltaib A. Saad and Gamal M. Abdulla declare that they have no conflicts of interest.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from all individual participants included in the study.

Publisher's note Springer Nature remains neutral with regard to jurisdictional claims in published maps and institutional affiliations.

References

1. Fahal AH, Hassan MA (1992) Mycetoma. Br J Surg 79(11):1138–1141
2. Zijlstra EE, van de Sande WWJ, Welsh O, Mahgoub ES, Goodfellow M, Fahal AH (2016) Mycetoma: a unique neglected tropical disease. Lancet Infect Dis 16(1):100–112
3. Bonifaz A, Tirado-Sanchez A, Calderon L, Saul A, Araiza J, Hernandez M et al (2014) Mycetoma: experience of 482 cases in a single center in Mexico. PLoS Negl Trop Dis 8(8):e3102
4. Fahal AH (2004) Mycetoma: a thorn in the flesh. Trans R Soc Trop Med Hyg 98(1):3–11
5. Hay RJ, Fahal AH (2015) Mycetoma: an old and still neglected tropical disease. Trans R Soc Trop Med Hyg 109(3):169–170
6. van de Sande WW (2013) Global burden of human mycetoma: a systematic review and meta-analysis. PLoS Negl Trop Dis 7(11):e2550
7. Welsh O, Vera-Cabrera L, Salinas-Carmona MC (2007) Mycetoma. Clin Dermatol 25(2):195–202
8. Fahal A, Mahgoub el S, El Hassan AM, Abdel-Rahman ME (2015) Mycetoma in the Sudan: an update from the Mycetoma Research Centre, University of Khartoum, Sudan. PLoS Negl Trop Dis 9(3): e0003679
9. Suleiman SH, Wadaella el S, Fahal AH (2016) The surgical treatment of mycetoma. PLoS Negl Trop Dis 10(6):e0004690
10. Welsh O, Al-Abdely HM, Salinas-Carmona MC, Fahal AH (2014) Mycetoma medical therapy. PLoS Negl Trop Dis 8(10):e3218
11. Fahal A, Mahgoub el S, El Hassan AM, Jacoub AO, Hassan D (2015) Head and neck mycetoma: the Mycetoma Research Centre experience. PLoS Negl Trop Dis 9(3):e0003587

12. Alam K, Maheshwari V, Bhargava S, Jain A, Fatima U, Haq EU (2009) Histological diagnosis of madura foot (mycetoma): a must for definitive treatment. J Global Infect Dis 1(1):64–67
13. Tamir G, Adler A, Hagler J, Hauben DJ, Goldberg I (1995) Mycetoma of the foot - surgical treatment with free flap reconstruction. Eur J Plast Surg 18:124–126
14. Mestre T, Vieira R, Coutinho J (2015) Mycetoma of the foot–diagnosis of the etiologic agent and surgical treatment. Am J Trop Med Hyg 93(1):1–2
15. Gismalla MDA, Abdulla GM, Ali MM, Mohamed SM (2016) Wide surgical excision and reconstruction of eumycetoma in Gezira Mycetoma Centre. Glob J Med 16(2):14–17
16. Maina AM, Macharia JT (2012) Alleviating a Nomad's anguish: successful treatment of a case of leg mycetoma-a case report. Case Rep Orthop 2012:753174
17. Ali A, Khadda S, Yadav AK, Parmar A, Sakhrani JK, Kapoor A (2015) Giant eumycetoma gluteal region: excision and V-Y flap advancement. Int J Sci Rep 1(2):146–147
18. Wadal A, Elhassan TA, Zein HA, Abdel-Rahman ME, Fahal AH (2016) Predictors of post-operative mycetoma recurrence using machine-learning algorithms: the Mycetoma Research Center experience. PLoS Negl Trop Dis 10(10):e0005007