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Review Article

Reconstructive surgery for mycetoma: Preliminary algorithm and a systematic review

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ABSTRACT

Background: Surgical treatments of mycetoma are a cornerstone in management. However, while doing a wide surgical excision of mycetoma lesion, surgeons think about how to close the skin defect, which can be closed primarily, left to heal by secondary intension, by skin grafts or local flaps. In this review, we demonstrate the various applications and changes of mycetoma reconstruction after surgical excision.

Methods: This is a systematic literature search and review conducted to determine articles presenting mycetoma reconstruction options. Articles were identified, and the time of publication, type of study, time of study, and country of study were checked. Additionally, all patients in those articles were included. Patients' names, sex, clinical presentation, and management were identified.

Results: A total number of 9 articles fulfilled our inclusion criteria; 8 of them are case reports, and 1 is a case series. The first mycetoma reconstruction case was published in 1959. The country of publication varies from tropical and non-tropical countries. The total number of patients found in those articles is 34 patients, most of whom are male. The causative organism is mainly eumyce-

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toma. The site of mycetoma lesions is varied with variable sizes. The reconstruction options used were skin graft and local or regional flaps, where only 1 case underwent a free flap for reconstruction.

Conclusion: Reconstruction of mycetoma should be considered following mycetoma surgery in small or large size defects if skin closure is not feasible and there is no indication for amputation.

Introduction

Mycetoma is a chronic, progressively destructive granulomatous disease affecting the skin, subcutaneous, connective tissue, muscle, and bone. It is caused by certain fungi (eumycetoma) or bacteria (actinomycetoma).[1] In 2016, the World Health Organization added mycetoma to the list of 18 priority neglected tropical diseases; this is an important step toward control of the disease.[2] Mycetoma mainly affects poorer populations in remote rural areas in tropical and subtropical countries in the so-called "mycetoma belt" regions, including Sudan, Somalia, Senegal, Yemen, India, Mexico, and Venezuela.[3] Other countries with reported mycetoma include Egypt, Mauritania, Kenya, Niger, Nigeria, Ethiopia, Chad, Cameroon, Djibouti, and Somalia in Africa and Colombia, Argentina, and Brazil in Latin America.[3]

Mycetoma is characterized by a typical triad of painless subcutaneous tumour-like swelling, multiple draining sinuses and fistulas, and grainy discharges, which are the colonies of bacteria or fungi, with different colours, sizes, and textures, depending on the species of the causative microorganism(s).[4] The diagnosis of mycetoma can be made based on the clinical trial of symptoms in endemic regions. Grains are first grossly examined for their colours, size, and consistency; then smeared on clear slides; and then examined under a light microscope for fungal hyphae and spores in case of eumycetoma and branching filaments for actinomycetoma.[5] Sero-diagnostic tests, including immunoblots, indirect hemagglutination assays (IHAs), immunodiffusion (ID), counter immunoelectrophoresis (CIE), and enzyme-linked immunosorbent assays (ELISAs), were developed to test both actinomycetoma and eumycetoma.[6]

Either x-ray, ultrasonography, computed tomography (CT), or magnetic resonance imaging (MRI) is required to reveal a mycetoma's site, size, extent, bone invasion, and relationship with surrounding tissues and to plan surgical operation.[7]

The subcutaneous tissues can be obtained with a punch biopsy, local surgical excision, and fine-needle aspiration (FNA). FNA specimens can be either imprinted on slides for further DME or processed for cell blocks and further processed as biopsy specimens for histopathological assessment.[8]

Treatment of mycetoma depends mainly on causative organisms, fungi or bacteria.[9] Usually, actinomycetoma is treated by using anti-bacterial treatments such as cotrimoxazole, whereas eumycetoma is treated by using anti-fungal treatments before and after surgical excision.[10] After surgical excision, the resulting defect is closed with primary closure if feasible. Sometimes, if the defect cannot be closed primarily, reconstruction may be necessary.[11]

This research was conducted to review articles demonstrating mycetoma reconstruction and to highlight the characteristics of the patients with mycetoma. Additionally, we look for reconstruction of for mycetoma case after excision.

Methods

This research was conducted to determine the surgical opinions, options, and modality treatment of mycetoma. An electronic systemic search was conducted by examining PubMed and Google Scholar

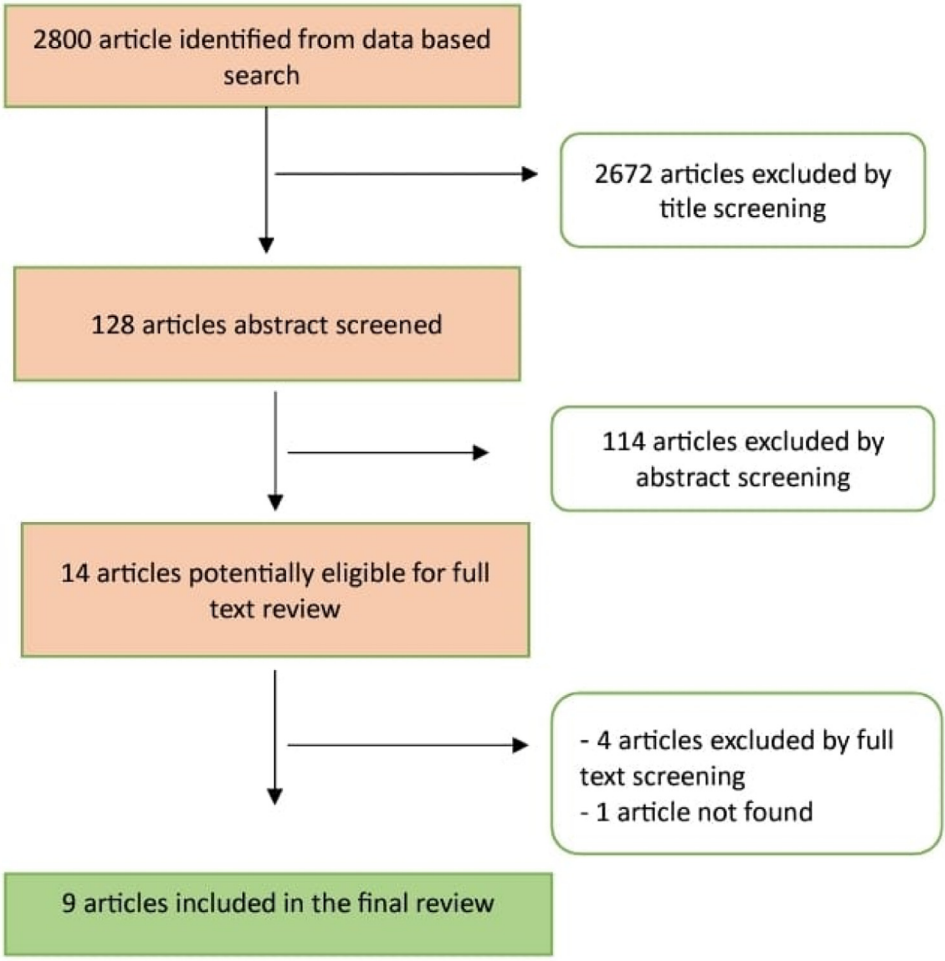

Figure 1. Inclusion process for articles reviewed in this study.

websites. The words mycetoma, eumycetoma, and surgical management were used for selecting the articles.

Inclusion criteria were any article describing the surgical management with reconstruction of mycetoma written in English. Any article irrelevant to mycetoma or not concentrating on surgical treatment with reconstruction was excluded. Any article not describing patients' characteristics or surgical management of mycetoma with reconstruction was excluded.

Review of the article titles was conducted by 2 co-authors and followed the inclusion and exclusion criteria. Subsequently, each abstract was screened and reviewed, and, if needed, the article was reviewed to extract the data. The details about the type of article and subject of publication were shown in Figure 1.

After reviewing, all articles were tabulated, and the detailed personal data, clinical presentation, and reconstruction types were determined. Analysis was conducted for the presentation of the data. The patient's characteristics will be extracted to draw a road map algorithm to select the patients who are suitable for reconstruction.

Table 1
Included published articles on mycetoma reconstruction

	Country	Study design	Patient's characteristics	Site of mycetoma	Mycetoma	Bone involvements	Reconstruction	Medical treatments	Follow-up
Ali[12]	India	Case report	A 55-year-old male farmer	Left gluteal	Eumycetoma		V-Y advancement flaps	-	4 weeks
Maina[13]	Kenya	Case report	A 34-year-old male	Left leg	Actinomycetoma and eumycetoma	-X-ray -No, just periosteal reaction	Gastrocnemius Flap	-Itraconazole and ciprofloxacin -Itraconazole and penicillin	8 months
Tamir[14]	Israel	Case report	A 28-year-old male	Right foot	Eumycetoma	-MRI -Bone not involve	Free musculocutaneous flap, based on the lateral circumflex vascular bundle, through the lateral approach	-Ketoconazole	12 months
Al-Kathiri[15]	Oman	Case report	A 37-year-old male	Right gluteal	Actinomycetoma	- X-ray - Bone not involve	Skin graft	Dapsone and rifampicin	2 years
Kalender[16]	Turkey	Case report	A 59-year-old male	Right ankle	Actinomycetoma	- CT and MRI - Bone not involve	Skin graft and NPWT	-	3 months
Estrada-Chavez[17]	Mexico	Case report	-A 21-year-old male farmer -recurrence	Foot	Eumycetoma	-X-ray and CT -Bone not involve	Skin graft and NPWT	-Ketoconazole -Itraconazole	18months
Conway[18]	United States of America	Case report	A 44-year-old male	Left foot	Eumycetoma	-	Cross leg pedicle flap	-	-
Mestre[19]	Portugal	Case report	-A 43-year-old female, borne in Cape Verdi* -recurrence	Right foot	Eumycetoma	-MRI -Bone not involve	Skin graft	With itraconazole	16 months
Abdelrahman[20]	Sudan	Case series	-Total (n=26) -The mean age was 28 years (range, 18–56 years) -male to female ratio of 4.2:1) -Recurrence 9 patient	Foot (n=9) Hand (n=5) Leg (n=3) Knee/thigh (n=2) Gluteal (n=3)	Eumycetoma	X-ray MRI No bone involvement	-Skin graft (n=14) - V-Y advancement flaps (n=5) grastrocnemius flap (n=2) dorsalis pedis pedicled flap(n=1) ateral calcaneal artery skin flap (n=1) skin undermining and tension-free suture(n=3)	Antifungal therapy	18 months

Cape Verdi* Portugal tropical African island.

Table 2
Characteristics of patients who underwent mycetoma reconstruction

Variables		Total	Percentage
Sex	Male	27	79.4
	Female	7	30.6
History of mycetoma	No	23	67.6
	Yes	11	32.4
Site of mycetoma	Gluteal	5	14.7
	Leg	4	11.8
	Foot	14	41.2
	Hand	5	14.7
	Knee and thigh	4	11.8
	Arm	2	5.9
Types of mycetoma	Eumycetoma	31	91.2
	Actinomycetoma	2	5.9
	Actinomycetoma and eumycetoma	1	2.9
Bone involvement	No	34	100
	Yes	0	0
Reconstruction	Local V-Y advancement flap	6	17.6
	Regional flaps	5	14.7
	Free musculocutaneous flap	1	2.9
	Skin graft	18	52.9
	Cross leg pedicle flap	1	2.9
	Undermining and tension-free suture	3	8.8

Results

The total number of included articles in this study was 9; 8 of them are case reports, and 1 is a case series. They are published from different countries, which include tropical and non-tropical countries. Four articles had been published from Asia, 2 articles from Africa, 2 articles from North America, and 1 article from Europe. The first article that described surgical excision of mycetoma and reconstruction was published in 1959. Five articles in this review were published in the previous decade. The other detailed descriptions of those manuscripts are shown in Table 1.

In this review, we studied and evaluated 34 patients who had been diagnosed with mycetoma and undergone wide surgical excision with reconstruction. In these reviews, males predominate over females. The mean age was 21 years, ranging between 18 and 59 years. Eleven patients (32.1%) had a previous history of mycetoma with recurrence lesions. The commonest sites of mycetoma among those patients are foot, hand, and gluteal, with 11, 5, and 5 rates, respectively. The causative organisms are eumycetoma in 31 patients, whereas only 2 patients had actinomycetoma. All patients who underwent reconstruction of mycetoma had no bone involvement. The commonest reconstruction done is skin graft, local flap, and regional flap. All these detailed patient characteristics are presented in Table 2.

As a result of Table 2 and the details of patients' descriptions, an algorithm was concluded. This algorithm will demonstrate the management steps to treat patients with mycetoma and the reconstructive option needed to close the defect (Figure 2).

Discussion

Abdelrahman et al. stated a question—"Is There a Need to Establish an Algorithm for Reconstruction of Mycetoma surgery?"—while presenting a number of patients with mycetoma who underwent wide surgical excision and reconstructions.[21] In that article, the authors highlighted the obligation of an algorithm while managing a mycetoma by using wide surgical excision with or reconstruction.[21] As a result, an algorithm was necessary for mycetoma surgical excision to get adequate, accurate, and acceptable treatments. This research review was conducted to rule out and draw an algorithm flowchart to guide surgeons while treating mycetoma. It can support the health provider during providing treatments and to refer for further management.

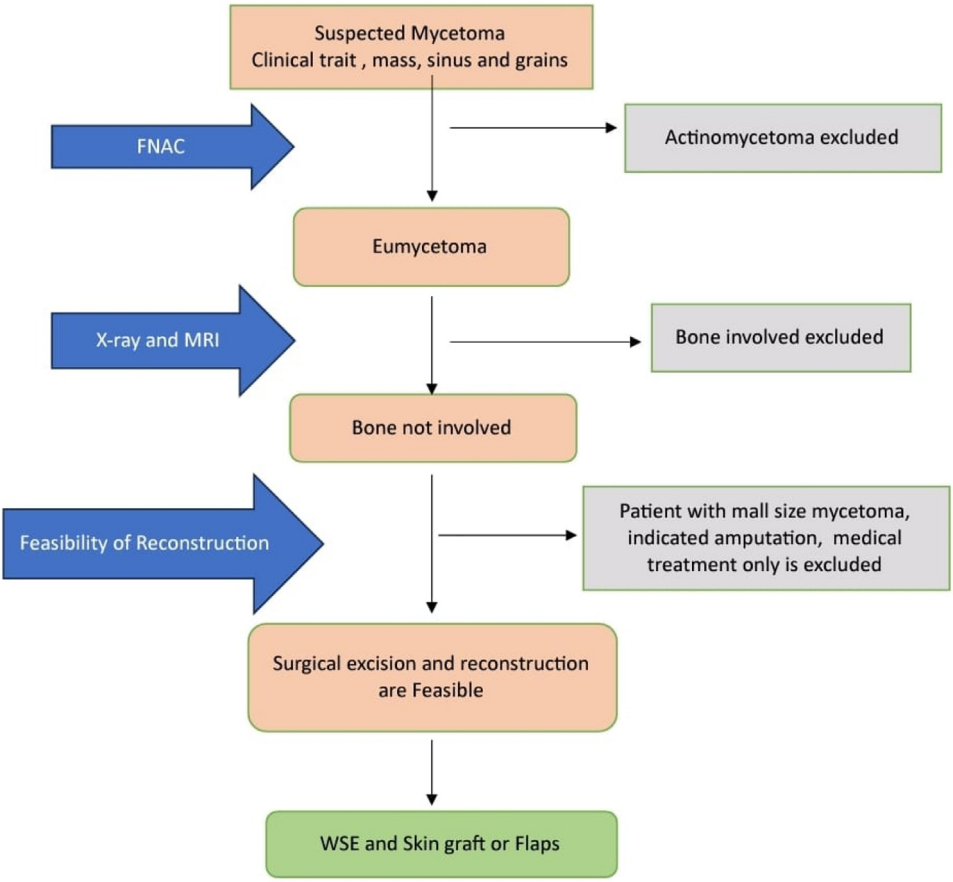

Figure 2. Reconstructive surgery algorithm for mycetoma.

Patients with mycetoma should be thoroughly assessed clinically. Past medical history, dominant hand, and social background of any patients should be obtained.[11] Clinical examination of the lesion site, size, shape, sinus, swelling, redness, and evidence of secondary infection is required.[11] As we present in this algorithm, the lesion should be determined with cytological or histopathological tools. FNA cytology is considered an initial confirmatory tool; it is easy, rapid, and inexpensive in full hand. Additionally, the fixation of cells in blocks can be considered a diagnostic tool.[22]

The aim of surgery is to excise the mycetoma lesion completely and then close the defect.[10] Closure of the skin can be obtained primarily.[10,11,23] Factors that determine the primary closure are mass less than 5 cm, mass size of 5-10 cm, and that the surgeon can excise the lesion and close the defect.[10,11] If the lesion is more than 10 cm, it is better to reconstruct the defect to close the wound.[20,21]

Bone involvement was evaluated before any surgical decision. Usually, this was done by a plane x-ray with anterior and posterolateral views.[7,24] Sometimes, MRI and CT scans are useful to describe the soft tissue involvement as well as the bony involvement.[25] Any patients who had bone involvements should not be offered excision and reconstruction.[10,20] The recurrence of mycetoma lesions in cases of bone involvement is very high. It is useless to do major surgery or reconstruction in those patients.[24]

Reconstruction of mycetoma should follow the reconstructive ladder, which begins from the primary suture till the free flaps. In this review, the reconstruction varies from skin under mining to free flaps. As a result, we should offer a reconstruction for any excised mycetoma lesion when the closure

cannot be obtained. If amputation is indicated, it is better not to reconstruct. Some patients had a lesion in a difficult organ where surgery is challenging, such as the brain[26] or perineum.[27]

Conclusion

Surgical mycetoma essentially depends on wide surgical excision and primary closure. While closure is not achieved, reconstruction is mandatory and advisable. There are some contraindications for reconstruction, such as a patient not being a candidate for surgery, bone involvement, and precluded amputation.

Declaration of competing interest

The authors declare no competing interests.

Availability of data and materials

The manuscript contains all pertinent information.

Author contributions

MDAG, MYB, and AMA contributed to the conception, design, and data extraction; MSMS, GMAA, AMIA, and AEKA evaluated the methodological quality of the included articles, participated in data analyses, interpretation, and writing the first draft of the article. All the authors read, commented on, edited, and approved the final submitted manuscript.

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Institutional Review Board Statement

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Ethics approval and consent to participate

The ethical approval and consent to participate in this study are not applicable, because, as stated in the title, it is a systematic review and meta-analysis with no direct participation of study subjects as in a primary study.

Consent for publication

Not applicable.

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