Final report
Project name:
Mapping the geographical distribution of Mycetoma in Sudan, a pilot study
Mycetoma research centre-University of Khartoum

- **Introduction** :

Mycetoma is a common health problem, endemic in many tropical and subtropical regions characterized by devastating deformities, disabilities and y, high morbidity [1, 2]. It has serious socioeconomic bearings on patients, families, and communities [3,4, 4]. To date, its global incidence, prevalence, and route of infection are not well characterized, as is its susceptibility, resistance, and response to medical treatment [5-7]. This data scarcity is badly reflected in the available diagnostic and treatment modalities which proved shown to be suboptimal [8, 9].

Early-stage detection of mycetoma and subsequent medical intervention lead to more favorable prognosis and minimization of disease complications and recurrence sequel, and its economic burden [10, 12]. However, mycetoma patients showing up to the Mycetoma Research Center, (MRC), WHO Collaborating Center on Mycetoma, University of Khartoum, Khartoum, Sudan only when the disease has progressed to a point where mobility and activity participation has been severely affected curtailed [13,14].

The available diagnostics and treatments for mycetoma are far from satisfactory; they are either invasive toxic and or expensive for patients and health authorities in endemic areas [15, 16]. While there is a wide array of causative agents of the disease, the fungal form; eumycetoma, the most prevalent type encountered in Sudan proved to be more difficult to treat and results in higher morbidity [17, 18].

Furthermore, worldwide, there are neither o preventive nor or control measures nor or programs for mycetoma [19]. The disease surveillance, especially in Sudan, is limited to targeted prevalence studies, anecdotal case reports, and passive case detection. The absence of a standardized, centralized mycetoma surveillance system has far-reaching effects on how existing interventions are delivered in a cost-effective, evidence-based manner. In this project, the study team had tested a new approach for mycetoma services decentralization based on available health system structure and minimum requirement of mycetoma management and control services.

- **Objectives**
- **General objective:**

1. To transfer capacity to community health workers to implement a rapid epidemiological assessment of mycetoma
2. To determine the prevalence of mycetoma in Sudan
3. To estimate the population at risk of mycetoma in Sudan

- **Specific objectives:**

1. To estimate the overall burden of mycetoma in Sudan;
2. To identify the spatial distribution of mycetoma in Sudan;

3. To identify demographic, ecologic, environmental and socioeconomic factors that influence the spatial distribution of mycetoma in Sudan;
4. To build the capacity within Sudan to undertake large scale epidemiological/spatial mapping studies of other endemic diseases in Sudan

- **Data & Methods**
  - **Study design :**
- This community-based cross-sectional study was conducted at Eastern Sennar Governate, Sennar State, Sudan in the period 2016-2017.
- **Study settings and population:**
- The Governate is 400 km south Khartoum, the Sudan Capital, has 292 villages with a total population of 219,800 inhabitants. In this study, 19 villages in the Governate were surveyed for early mycetoma patients' detection. One village; Wad EL Nimear, the highest endemic village, was studied in depth. During the study period, four medical and health mobile missions form the MRC to the study villages were conducted. The mission team consisted of three consultants surgeons, one physician and two consultants radiologists, four one surgical registrars, two one radiology registrars and one anesthesiology registrars, one molecular biologist, one epidemiologist, one environmental consultant, one pharmacist, five medical officers, three surgical scrub nurses, two anesthesiology assistants, four laboratory technologists, two nurses, 10 medical students, one photographer, two fine artists and one musician. The study was implemented in partnership with Federal Ministry of Health, Sennar State Government, the local administrative authority and local community leaders and activists to assure the services sustainability and the study outcome execution.

  Data collection:

  For early case detection and management, house to house total coverage survey of 19 villages in the Eastern Sennar Governate was done. The data were collected by well-trained teams of medical officers, house officers, medical students, health care providers and community activists using digital pre-designed validated closed-ended questionnaires in smart tablets. Computer Assisted Patients Identifier (CAPI) computer application designed for this study was used. To validate the study questionnaire and the computer application, a pilot study was conducted prior to the data collection in a nearby village.The collected data included the suspected patient's demographic characteristics, lesions photographs, the suspected patient's locality geographic coordinates (latitude, longitude, altitude) and the neighborhood photographs. The CAPI was connected to the MRC, Data Centre system and the data analysis was done spontaneously.

  All the suspected mycetoma patients from the Governate were referred to the Wad Onsa Mycetoma Regional Center (WOMRC). The WOMRC was established in 2015 as joined efforts between the Federal Ministry of Health,

Ministry of Health, Sennar State, MRC and the local community leaders to manage mycetoma patients locally in their region. The centre consists of small surgical operation complex, two wards, pharmacy, laboratory and ultrasound and out-patient suites. At the centre, the patients were managed by the MRC mobile mission team, and in between MRC mission visits, they were treated by the surgical team from the regional Sennar Teaching Hospital and the local resident doctor at in the WOMRC. The diagnosis of mycetoma was established by careful clinical examination and lesion ultrasound examination by mobile ultrasound machine conducted by the consultant radiologists. All patients underwent surgical excisions under general or spinal anesthesia by the consultant's surgeons and surgical registrars. The histological examination of the surgical biopsies and grains culture and PCR were performed at the MRC. Some patients with massive lesions were referred to the MRC for further assessment and management. In between the MRC mission the patients were followed up by telemedicine facility communicating the WOMRC and the MRC. All the investigations and treatment were provided free of charge. System for medicines procurement, delivery and storage was designed.

The confirmed mycetoma patients' information was entered in a predesigned patient's management records. That included patients' demographic characteristics, diagnostic tests results, management decisions, follow-up plan and final patient treatment result. This information was regularly checked and updated throughout the patient management and follow-up period.

Health Care Providers Training

More than 300 care providers; medical assistants, nurses and public health officers were trained on different aspects of mycetoma, and that included patients' care and referral indications, community population health education and disease advocacy. The instructional training instructional methods included presentations, group discussions, clinical sessions and ultrasound diagnosis demonstration. Suspected patients' referral card was designed the distributed to the trainee. The improvement in knowledge, attitude and practice (KAP) of the trainees were assessed by pre and post-training tests.

The training sessions were conducted at Singa Town, Sennar State capital, WOMRC and in various East Sennar Governate villages. The training also included a group of medical students from the local University; Sennar University, as well as the University of Khartoum University and other health institutes on early detection, referral and management of mycetoma patients were conducted at WOMRC.

Local Community Involvement

Several meetings with the local villages' leaders, villagers and community activists ties were conducted at the Wad Onsa village leader's home, the village mosque and WOMRC to explain the study objectives and to gain their confidence and support. They were actively involved in the mycetoma advocacy activities and awareness raising. The local Red Crescent volunteers

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were training in mycetoma advocacy and took a good role in improving the local environment and hygiene in the affected villages.

A toktoko (big motorcycle) was donated by the MRC to a community activist at Wad EL Nimear village for transporting patients and their medicines between the different villages and for mycetoma advocacy. In appreciation of excellent, active and energetic involvement in mycetoma advocacy and awareness, three Mycetoma Ambassadors from the Sennar State were selected.

Health Education & Advocacy

The study Health Education Team was led by social workers with several fine artists, musicians and community volunteers.

The health care providers, community leaders and activists, school teachers and medical students from the local university were trained to conduct health education and advocacy sessions, and they had conducted that in different villages to raise the disease awareness. Health education and advocacy material were designed and produced.

Several health education sessions and activities were carried out for early case detection, and active case finding was conducted. The activities included small group discussions, school visits sessions, video films watching, and interactive open theatre drama.

“Mesaket Story”, a drama film documented a mycetoma patient journey from minor infection which was neglected till limb amputation was produced and was shown to more than 1000 individuals at WOMRC and other villages.

Subsidiary project activities:

Community Environmental Improvement Campaigns

The methodology was designed in three steps:

1- Developing evidence base, identifying key campaign messages and strategies

2-Intervention - conducting the community education campaign;

3- Assessing education impacts and evaluating campaign effectiveness.

- These campaigns were conducted to improve the community environment, sanitation, and hygiene to reduce the mycetoma transmission risks were organized by the State Government, official local authorities, community leaders and activists, community and Red Crescent volunteerss in collaboration with the study team. The thorny trees and bushes, thorny animals cages, animal dungs, dirtdirt and rubbish, were removed and burnt.
- To improve the village hygiene, reduce the contact with the animals and their excreta and to eradicate the thorny cages, 72 modern animal cages were constructed outside Wad EL Nimear village, one of the highly endemic villages in the locality. This project was conducted by a kind donation from an engineering company as its a social reasonability activity. The new animals cages were distributed free of charge to the villagers. The study team had distributed around 400 new shoes to the

school pupils at Wad EL Nimear village to improve the personal hygiene and to reduce the risk to develop mycetoma.

- **Results**
- **Patients' Management**
- In this study, 758 suspected patients from the surveyed villages and referred by the Governate health care providers were seen at the WOMRC. Of them 220 patients underwent surgical treatment to confirm the diagnosis of mycetoma. There were 134 males (60.9%)and 86 female (39.1%). Their ages ranged between 2 and 70 years. Most of them were students 68(30.9%), housewives 46(20.9%), farmers 35(15.9%). The geographical distribution was uneven, but Wad El Neamir village had the highest prevalence.
- The ultrasound diagnosis of these patients was eumycetoma 157(71.4%), foreign body granuloma 16(7.3%) and others.
- The surgical procedures ranged from wide local excision 218 (99%) to amputation 2 (1%). All patients had uneventful postoperative recovery. The operatives findings included mycetoma lesions 192(87.3%), foreign body granulomas with thorns 17(7.7%), fibroma 2(1%) and others. That was confirmed by surgical biopsies histopathological examinations, grains cultures and in few by PCR. The final diagnosis of these patients was eumycetoma 189(85.9%), foreign body granuloma 17(7.7%), actinomycetoma 3(1.4%) The patients were followed up in the centre. Thirty-seven patients (16.8%) had a recurrence, and the cause was multifactorial which include massive lesion, patients' non-compliance with treatment or others. Twenty-five patients (11.4%) were lost to follow-up.
- **Patients' Information System**
- Mycetoma confirmed patients' information was entered in the pre-designed patient's management records. These records included full details of the patient's demographic characteristics, diagnostic tests results, the management offered, follow-up and final patient treatment result. This information was regularly checked and updated throughout the patient management and follow-up period.
- All these data were systematically reported to the MRC in quarterly basis through two types of reporting format; hard and a digital one through a telemedicine facility at the WOMRC and MRC. The reported information was systematically entered in the pre-designed data analysis software for further analysis and systematically checked for information accuracy.
- Data from management teams, diagnostic services and inventory was crosschecked and discussed regularly to improve recording and reporting process.
- **Free Medicines Management**

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- Medical treatment was provided free of charge to all patients. A system for medicines procurement, delivery from the MRC, storage at the WOMRC and dispensing was designed and tested in the study.
- **Community Mycetoma Advocacy Activities**
- The advocacy team had conducted seven big health education sessions and several small group sessions on mycetoma clinical presentation, treatment and control. They also organised ten training sessions for different community activists and school teachers and that included small group discussions, school activities, video films watching, and interactive open theatre drama. Different health education materials in the form of posters, leaflets, video film, audios clips were used.Experts in watercolour, oil painting and a photographing had contributed massively to mycetoma advocacy and awareness through their production of high-quality paints, photographs and videos captured from the Governate.

- **The Social study**
  To study the social background of the population in the affected villages in the study area, their KAP to mycetoma and socioeconomic impacts, 40 students from the Department of Social Sciences, at the University of Khartoum had spent two weeks at Wad Onsa village. They surveyed in depth ten villages in the locality. Opened ended questionnaire and focus group discussion were used to obtain the data. The KAP study showed low levels of health education level among the studied population. It was clear that mycetoma is considered as a social stigma, particularly among females. The villagers believed that the WOMRC had many positive impacts on their lives.

- **Discussion** :
- Although the MRC records showed that EL Gazeria and White Nile states are the commonest endemic states in the country and Sennar state is the third one yet the present study was conducted at the later state due to extreme and strong political and community commitments to support it and to implement the research outcome. The response to the study team request to pledge the study was quick and positive from both the senior state officials and civil society members as they were aware of the mycetoma medical and socioeconomic negative impacts and bearings.
- The establishment of a village specialised mycetoma centre to deliver an integrated medical and social services at Eastern Sennar locality, the heart of an endemic area, is a unique experience that is not reported previously. The centre was established as joint efforts of the Federal and Sennar Ministries of Health and the local community, and this is also an exceptional experience. This study had revealed numerous positive impacts of the centre on the local communities. It had improved the local population health education and disease awareness, had provided local decentralised mycetoma services based

on the existing health system with minimum requirements for their management. The telemedicine facility at the centre had helped in the management and follow-up of the patients, by reducing the financial and geographic burdens on the patients and families, had reduced the patients' follow-up dropout rate. Although the cost of the telemedicine establishment is high in developing countries, however, on the long run, it will be more economical for the patients, families and health authorities in mycetoma endemic regions.

- As there are still no disease control or elimination programmes worldwide due to lack of the basic disease epidemiological characteristics, early case detection and management remain the only available methods to reduce the disease incidence and prevalence and its community impacts. It is clear that WOMRC had tremendous bearings on that by offering early case detection facilities and free decentralised medical, health and advocacy services.
- The mycetoma onset and progress are usually slow and painless, and the patients are of low socio-economic and health education levels and hence they are different from patients with other deadly diseases, e.g. malaria, cholera, leishmaniasis, where patients have no other choice than to report early and follow medical instructions. Furthermore, mycetoma patients with early lesions are different from patients with large disabling massive mycetoma lesions. Early small lesions are usually painless not interfering with their normal daily activities, some of them consider it at this stage as a normal event. These patients usually have many other more pressing social and economic problems than their mycetoma, e.g. the short seasonal farming sessions, raising children in poor conditions, so their early lesions are not priority, and in fact, they believe that treatment will delay undertaking these urgent issues, and that explains the late presentation with massive lesions (Fahal AH, personal communication).
- From this study, it is clear that this holistic management had addressed all these issues. The community engagement activities had led to active early case detection and this fact is supported by the high number of patients with early disease were seen at the WOMRC, and these patients were amenable to treatment and had good outcome.
- Patient access to free treatment at the village level as seen in this study had reduced the patients' delay in starting treatment, decrease the patients' geographical and financial burdens, treatment interruption, and patients' follow up drop out that can induce drug resistance. For this, it is vital to ensure that the free mycetoma treatment services are sustainable and available.
- The medical assistants, the health care providers, are the backbone for the management of mycetoma patients in Sudan. At Sennar State, most of them were trained on the different aspects of mycetoma care and management. That was essential as most of them used to operate on the mycetoma patients under local anaesthesia and suboptimal conditions with poor surgical experience

leading to the recurrence rate we documented in many reports [7, 10]. Recurrent disease is usually associated with wide local disease spread. Hence it is difficult to cure and thus the repeated surgical excisions that cause numerous deformities and disabilities [10].

- A well-trained multidisciplinary team on mycetoma was developed in Eastern Sennar Locality. It consisted of a trained surgeon, surgical theatre attendants, anaesthetic assistants, pharmacy assistant, ultrasound technicians, nurses, information technology expert, statistical clerks, community leaders and activists. That is essential to provide comprehensive, holistic management for the mycetoma patients.
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- Currently, the definite route of infection in mycetoma is an enigma. However, it is clear that mycetoma incidence is high in areas with poor hygiene, where people live in close contact with animals and their dung, thorns, dirt, mud and poor environmental conditions. Hence this study aimed to improve Wad EL Nimear village, one of the highly endemic villages in the locality, the living and hygienic standards. The local villagers were encouraged to improve that. For that many advocacy and awareness campaigns were conducted and the new modern healthy animals' cages were constructed and donated to them free of charge. To support all these, the local Governate authorities had issued a law banning the presence of animals in inside the village. This kind donation of the animals' cages by the engineering company is due to intensive mycetoma awareness and advocacy in the community.

- In this study, the community leaders and activists were actively involved in conveying messages to their community in their own culture and traditions, and that is important to accept this holistic disease management procedures. Likewise, the local villagers have engaged actively in promoting their health and improvement of local environmental conditions that believed to be the main source of transmitting mycetoma.

- In conclusion, the holistic, comprehensive management approach implemented in this study had improved the mycetoma patients' quality of care in the studied endemic area. More early disease was detected and treated, the treatment interruption rate was decreased that had led to increased cure rate and decreased the recurrence and hospitalisation rates. That will eventually lead to decrease in the amputation and disability rates. The results obtained from this study suggest that such study can be expanded to other endemic areas in the country. The MRC as a WHO Collaborating centre on Mycetoma communicating the present experience to the WHO to share the experience with other endemic countries to ensure good mycetoma management, prevention and control.

•The study of the environmental factors and the distribution of Mycetoma

In collaboration with a special team of environmental studies we managed to collect coordinates of the endemic villages. Many data were collected for the environmental indicators (soil and vegetation) which were analyzed in spatial dimension to illustrate the situation at each area. Environmental samples were collected from different parts of sennar state according to the patients coordinates .Samples were collected from soil (two depths; surface and 30cm depth), thorns, trees, walls and others. These samples will be processed for DNA extraction for meta-genomic analysis.

The Geographical mapping of Mycetoma the White Nile state (Umm garr Island):

The Mycetoma Research Centre (MRC), University of Khartoum in collaboration with the White Nile State Ministry of Health had conducted a surgical campaign and epidemiological field study to the White Nile state (Umm Garr east villages).

The objectives were:

2. Geographical mapping of Mycetoma in the White Nile state
3. Mycetoma awareness raising and advocacy.
4. Assessment of Mycetoma burden in the White Nile state.

For early case detection and management, house to house total coverage survey of the villages in the Umm Garr east was done. The data were collected by well-trained teams of medical officers, house officers, medical students, health care providers and community activists using digital pre-designed validated closed-ended questionnaires in smart tablets. Computer Assisted Patients Identifier (CAPI) computer application designed for this study was used. The collected data included the suspected patient's demographic characteristics, lesions photographs, the suspected patient's locality geographic coordinates (latitude, longitude, altitude) and the neighborhood photographs. The CAPI was connected to the MRC, Data Centre system.

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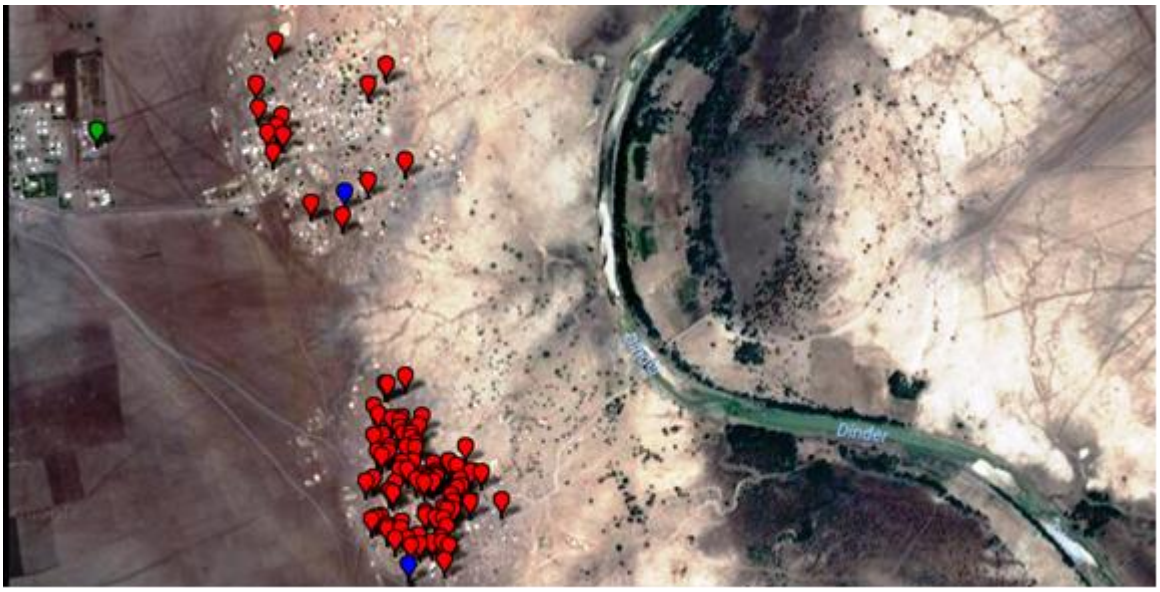
Picture No 1:
Tele-medicine facilities at the Mycetoma research centre

Picture No2:
Group of artist and social workers during the trip to Sinnar for Mycetoma advocacy



Map 1: Coordinates of Mycetoma patients in Wad Onsa and Wad EL Nimear villages –Sinnar state