

Improving mycetoma case detection through the training of community health workers in Northern Uganda: protocol for a stepped-wedge cluster-randomized trial

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Abstract

Introduction: The World Health Organization acknowledges the need for countries to incorporate neglected tropical disease care into their routine health care system. However, low detection rates and late presentation of mycetoma to health facilities have been observed in endemic countries, including Uganda.

Objective: To empower community health workers (CHWs) in Northern Uganda to recognize and refer suspects of mycetoma to health facilities.

Design: This will be a stepped-wedge cluster-randomized trial based in Gulu and Pader districts over a period of 9 months with sequential crossover from intervention phase to the control phase at different time points until both districts are exposed to the intervention.

Methods and Analysis: The study will leverage on the ongoing partnership between Northern Uganda Medical Mission and the Uganda Ministry of Health that has trained over 300 CHWs in Gulu and Pader. The study evaluation will be done using the RE-AIM (Reach, Effectiveness, Adoption, Implementation, and Maintenance) framework. The expected outcome of the study is increased detection and referral of suspects of mycetoma. Data will be analyzed using STATA 17.0 and Friedman statistics or Analysis of Variance to determine increase in case identifications and referrals.

Ethics and Registration: The study was approved by Mulago Hospital Research and Ethics Committee (MHREC 2406) and registered with Pan African Clinical Trial Registry (PACTR202301534749787).

Dissemination: The results from this trial will be published in a peer-reviewed journal. In addition, the findings will be shared at conferences, with funders, and at other research meetings.

Keywords: community-health workers, mycetoma, neglected tropical disease, Uganda

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Introduction

Mycetoma is a chronic debilitating granulomatous infection that is highly prevalent and endemic in the tropical and subtropical regions of Africa, Asia, Europe, and Latin America.¹ It is associated with significant morbidity, social stigma, deformities,

and disabilities, which could result into premature termination of occupation and inability to complete school among children.^{2–4} Despite these undesirable outcomes, the accurate global burden of mycetoma is still unknown.⁵ This affects the mobilization of funds and development of

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strategies to counter the deleterious effects of mycetoma. Several countries have carried out single center epidemiological studies to estimate their true prevalence and incidence of mycetoma. For example, using a retrospective cross-sectional chart review, researchers in Uganda estimated that the average incidence of mycetoma was 0.32/100,000 persons per decade, while its average prevalence was 8.32/100,000 persons per decade, translating to about 3683 people living with mycetoma.⁶ These findings align with the estimated prevalence of 0.11–1/100,000 population in Uganda by 2018 as reported by the World Health Organization (WHO).⁷ A systematic review and meta-analysis which reviewed 49 published articles on mycetoma and considered 8763 mycetoma cases, found that the average global number of reported cases of mycetoma was 127 cases per annum.¹ The numbers reported underestimate the true prevalence and actual burden of mycetoma in the endemic regions such as Uganda.

Several factors could be attributed to the under-reporting of mycetoma cases by affected countries. These include the chronic and painless nature of the disease, its association with the people of low socioeconomic standing,⁸ and inadequate public and health worker awareness of the diseases.^{5,9} These factors make affected persons reluctant to seek health care, in fact, a descriptive cross-sectional survey conducted among 7789 households in Sennar State, Sudan found that about 77% of the participants with mycetoma did not seek professional help while those that sought help had to incur exorbitant costs to access treatment.¹⁰ In addition, 66% of mycetoma cases in Sudan had first consulted a traditional healer for mycetoma treatment¹¹ which further delays their contact with the health system. Even when mycetoma patients report to the health facility early, the absence of fast, reliable, and cost-effective diagnostic tools as point-of-care tests to establish a timely diagnosis of mycetoma hampers efforts to diagnose and initiate treatment early.^{5,8}

In 2016, during the 69th World Health Assembly, the WHO designated mycetoma as a neglected tropical disease (NTD) and mandated member states to expedite and reinforce strategies for enhancing early detection and treatment of mycetoma cases.¹² This initiative also sought to train healthcare workers and promote community awareness about mycetoma cases.¹² Moreover,

the WHO roadmap for NTDs 2021–2030 emphasizes an integrated approach to mitigate the morbidity, disability, and psychosocial impacts associated with debilitating skin NTDs like mycetoma.¹³ As part of this integrated approach, it is expected that 8 and 15 countries adopt and implement integrated skin NTD strategies based on local endemicity in 2025 and 2030, respectively.¹³ The strategies include social mobilization, community education on NTDs, active case finding to detect and contact suspected cases early, and prioritized health care worker training for better diagnosis, treatment, and care. The primary goal of these measures is to strengthen the health system within the framework of universal health coverage. This ensures effective detection, referral, and provision of care for mycetoma, achieved through sufficient laboratory capacity to enable appropriate and timely diagnosis, as well as the establishment of surveillance systems for case detection and reporting.¹³ During the first WHO global meeting on skin-related NTDs, which took place in March 2023, mycetoma was among the skin NTDs targeted for control, with emphasis on placing the patients' needs at the center and providing training to front-line health workers to deliver better care.¹⁴

In light with these efforts, community health workers (CHWs) who are the first point of contact between the patient and health system in Uganda provide a reliable workforce that could enable Uganda to address the undetected burden of mycetoma. Despite the WHO launching a mobile application to facilitate diagnosis of skin NTDs,¹⁵ a small proportion (less than half) of CHWs in Uganda own smart mobile phones,¹⁶ and therefore there is limited access to the application. However, when CHWs are trained to detect illness in the community, their impact on case detection and referrals as well as community awareness is significant.¹⁷ Community-based interventions, such as training local medical and health personnel on early detection and management of mycetoma, as well as mycetoma advocacy by the local community, have shown positive results in Sudan.¹⁸ In this intervention, 758 suspected mycetoma cases were referred for diagnosis, with 220 confirmed cases, leading to improved quality of care for mycetoma patients through early disease detection and treatment, as well as an improvement in

their knowledge, attitude, and practices toward mycetoma within the community.¹⁸

In the proposed trial, CHWs in Northern Uganda, which may be an endemic region in Uganda,⁶ will undergo training in mycetoma case detection. This will implement a public health strategy to address the burden of mycetoma in Uganda, as it is in other endemic countries such as Sudan, following the sixth international conference on mycetoma in Khartoum in 2019. Additionally, it will improve case detection and bolster the national surveillance system and registry on mycetoma as well as strengthen the mycetoma preventive measures.

Objectives of the trial

- (1) To assess the baseline knowledge and practices of CHWs in Northern Uganda on mycetoma
- (2) To provide basic training to the CHWs on the clinical presentation of mycetoma
- (3) To evaluate the impact of training of the CHWs on case detection and referral of suspected cases of mycetoma

Methods and analysis

Trial design and framework: Stepped-Wedged Cluster-Randomized Controlled Trial which is essential for rapid adoption and uptake of outcomes will be conducted in Gulu and Pader districts. The proposed study design allows us to utilize a phased approach that involves a continuous evaluation of key process and outcome indicators.¹⁹ Additionally, our approach is well suited for the proposed project since the intervention will likely do more good than harm and because it may not be possible to deliver the intervention to all participants at the same time.²⁰

Assignment to the intervention: The districts will be clusters, and the crossing-over from one participating district (cluster) to another (from the control to the intervention phase) will follow a sequence. The districts will be allocated to the intervention or control group randomly and allocation will be determined by the holder of the sequence who is situated off site. Blinding of the CHW may not be possible because of the nature of the study while the risk of bias will be minimized by using an objective primary outcome.

The districts will start the trial at the same time point with one acting as controls until they have been randomized to move from the control to intervention arm. Crossing-over will take place every 12 weeks of exposure to the intervention until all districts have been exposed to the intervention.

Study setting

The study will be conducted in two districts: Gulu and Pader. We shall leverage on the ongoing partnership between the Northern Uganda Medical Mission (NUMEM) and the Uganda Ministry of Health that focuses on developing and expanding CHWs training. This collaboration has trained over 300 CHWs in all subcounties in the above districts. Multiple villages encompass a subcounty, and most villages have at least one CHW. The CHWs are reassessed on skill and didactic competencies annually. With repeated competency in all skills scoring >85% for two or more years, CHWs can choose to become advanced (lead) CHWs. The lead CHW becomes a trainer for new CHWs to ensure the sustainability of the CHWs program.

Study population

All CHWs who are trained by the NUMEM and Ministry of Health partnership and meet the eligibility criteria will be recruited using simple random sampling.

Inclusion criteria

- Both males and females.
- Above 18 years of age.
- Have consent to participate.

Exclusion criteria

- Those who reside outside the two districts will be excluded.

Sample size estimation

No previous study has been conducted to evaluate the impact of training CHWs in enhancing the case detection of mycetoma. Therefore, for this trial, we will recruit and train 50 CHWs in Gulu and Pader, given the resources we have at hand. A total of 22 and 28 CHWs will be selected from Gulu and Pader districts, respectively. The

selection of CHWs is based on the estimated populations of 171,215 and 200,500 in Gulu and Pader districts, respectively.

Intervention

Recruitment

A community engagement meeting will be held between the prospective participants and the study team to introduce the study to them and address any concerns that may arise. CHWs who show interest will be selected and randomized to participate in the study.

Formative activities

Prior to the exposure of the districts to the intervention, a formative baseline assessment of the knowledge of CHWs about mycetoma will be carried out. An investigator-developed questionnaire based on fungal disease books, peer-reviewed publications, the WHO, and Global Action for Fungal Infection briefings (<https://www.gaffi.org/wp-content/uploads/Mycetoma-briefing-paper-final-September-2018.pdf>) will be used to collect this data from randomly selected CHWs.

Intervention group

The proposed intervention is training of CHWs in mycetoma case identification. The CHWs from the intervention district will undertake a 3-day training covering all aspects of mycetoma. This will be based on a curriculum developed using existing training materials on mycetoma and related fungal NTD by the Leading International Fungal Education (<https://en.fungaleducation.org/courses/>). In addition, the training guide for front-line health workers on skin-related NTDs provided by the WHO will be utilized to train and guide CHWs in the identification of suspected mycetoma cases. The training will be delivered by two trained clinical mycologists/clinicians using a combination of PowerPoint presentations, tutorials, pictorials, and group discussions.

Control group

The control group will continue undergoing the continuous training offer by the partnership between the NUMEM and Ministry of Health of

Uganda. They will not undergo any intervention until the point of crossing-over.

Evaluation framework and data collection tools

Evaluation will be done at 3-, 6-, and 12-week intervals. We will evaluate the intervention using the RE-AIM (Reach, Effectiveness, Adoption, implementation, and Maintenance) framework.²¹ Reach: the number of CHWs who are willing to participate in this study. This will be collected using an investigator-designed questionnaire. Effectiveness will be the impact of our intervention on knowledge, practices and mycetoma case detection. This variable will be collected using data extract sheets from the health facility, pre- and post-training assessment tools as well as investigator-designed questionnaire. Adoption: number of CHWs willing to initiate the program. Data collection tool will be an investigator-developed questionnaire. Implementation: proportion of the intervention conducted, consistency of implementing the intervention by the CHWs, time and cost of training.

Data collection tool: Pre- and post-training assessment tools as well as an investigator-designed questionnaire. Maintenance: extent to which program becomes routine practice in the selected districts which will be done using a data extraction sheet retrieving data on referrals and case identification.

Data Extraction sheet: These will be standardized and provided to each CHW for notification and referral of suspects. A copy of the case report form will be kept by the CHW and another by the designated health facility.

Outcomes

Primary outcome will be implementation. Under the implementation process, we will consider RE-AIM.

Effectiveness will be the secondary outcome: Under effectiveness, we will consider change in the number of mycetoma cases reported and referred by the CHWs and changes in the level of knowledge and practices about mycetoma among CHWs. These will be considered at baseline and after exposure to the intervention. Based on the estimated prevalence of 8.32 per 100,000 persons

per decade,⁶ approximately 12,024 individuals will need to be screened by the CHWs to detect one case of mycetoma in the target population. Furthermore, suspected cases identified by CHWs will be referred to the nearest health facilities with basic laboratory diagnostics such as microscopy and ultrasound scans to assess and confirm the extent of mycetoma lesions. Confirmed cases will be managed at the district hospitals by healthcare professionals to ensure appropriate care and treatment.

Data management and monitoring

The generated data will be field edited for accuracy and consistency before it is uploaded to the online cloud. Paper-printed questionnaires will be used to collect the data and all research assistants will be required to upload the data to the online server daily. Quality assurance and fidelity checks will be done biweekly by the study investigator. All data from the project will be kept confidential with limited access to key personnel from the research team. All data will be anonymized prior to analysis and kept in the database. This will be managed by the research team and will be destroyed after 7 years. All de-identified data generated from this study will be available to researchers who submit methodologically sound proposals to the investigators after the trial.

Data analysis

We will perform a before-and-after analysis of scores of the participants using GraphPad Prism 8.03 by Dotmatics, Boston, Massachusetts. The impact of the training will be assessed by baseline determination of cases recognized and reported by the CHWs before the training and 3, 6, and 12 weeks after the training. We will analyze the data basing on the principle of the intent to treat. Categorical data will be presented using counts and percentages while continuous variables will be presented as mean and standard deviation or median and interquartile range as appropriate. A trend analysis will be conducted using Friedman statistics or Analysis of Variance to determine an increase in case identifications and referrals. A $p < 0.005$ will be considered statistically significant.

Dissemination

Project output will be published in peer-reviewed journals and presented at local and international

conferences. We will provide feedback to the CHWs, funder, and policymakers. In addition, blogs and newspaper articles will be published to disseminate our findings widely to both the scientific and nonscientific community.

Declarations

Ethics approval and consent to participate

The study has been approved by the Mulago Hospital Research and Ethics Committee (MHREC-2406). Written informed consent will be obtained from all participants before they are enrolled in the study. Participant data generated from this study will be kept confidential and accessible only to the key investigative team.

Consent for publication

Not applicable.

Author contributions

Winnie Kibone: Conceptualization; Methodology; Supervision; Visualization; Writing – original draft; Writing – review & editing.

Andrew Weil Semulimi: Conceptualization; Supervision; Visualization; Writing – original draft; Writing – review & editing.

Richard Kwizera: Conceptualization; Supervision; Visualization; Writing – original draft; Writing – review & editing.

Felix Bongomin: Conceptualization; Methodology; Supervision; Visualization; Writing – original draft; Writing – review & editing.

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Competing interests

The authors declare that there is no conflict of interest.

Availability of data and materials

Not applicable.

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