

Knowledge, Attitudes and Practices Regarding Onchocerciasis and Community-Directed Treatment with Ivermectin: A Cross-Sectional Study in South Sudan

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Abstract

Introduction: Despite decades of ivermectin mass drug administration for the control of onchocerciasis (river blindness) in South Sudan, it continues to cause significant ocular burden in endemic foci. The objective of this study was to assess the community's Knowledge, Attitudes, and Practices (KAP) about onchocerciasis and Community Directed Treatment with Ivermectin (CDTI) in Raga and Deim Zubeir, Western Bahr el Ghazal state (South Sudan).

Methods: A community-based survey was conducted from 21st March to 2nd April 2023 in 11 villages of Raga and Deim Zubeir parishes. From among village residents, consenting persons with eye diseases and their carers were randomly recruited and interviewed about onchocerciasis and about CDTI using a structured questionnaire.

Results: A total of 932 persons participated in the study. The majority (93.8%) of participants had heard of onchocerciasis, 83.4% knew that transmission of onchocerciasis is by blackfly bites, and 33.5% identified that blindness is the main sign of onchocerciasis. Furthermore, 77.8% of participants perceived onchocerciasis as a preventable disease and 84.0% upheld that CDTI is useful. However, 12.3% reported that their relatives had missed ivermectin treatment during the last session. Of the 530 (56.9%) participants who had missed CDTI more than twice in the past, 89.4% gave as reason being absent at the time of drug distribution. A large proportion (55.6%) of participants also reported experiencing side effects after ivermectin intake.

Conclusion: The study community had relatively good knowledge about onchocerciasis and CDTI was considered to be useful to prevent onchocerciasis. However, a considerable proportion of persons still missed ivermectin treatment during CDTI campaigns due to their absence, suggesting that increased awareness and better timing of CDTI may contribute to increasing ivermectin uptake in these villages.

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Introduction

Onchocerciasis (also known as river blindness) is a neglected tropical disease caused by the filarial worm *Onchocerca volvulus*, which is transmitted by the bites of infected female blackflies of the genus *Simulium* [1]. Onchocerciasis is currently present in 31 African countries, the Arabian Peninsula (Yemen) and the Americas (two remaining foci in the Amazon area) [2]; it is the world's second leading cause of preventable blindness [3]. It is estimated that 99% of the 20.9 million *O. volvulus*-infected individuals live in Africa. Of these, 14.6 million are considered to have onchocerciasis-induced skin disease and 1.15 million to have vision loss [1]. Increasing epidemiological evidence also suggests that onchocerciasis is able to induce epilepsy [4,5]. Onchocerciasis was first reported in Sudan as early as 1908 [6]. South Sudan is amongst the highly endemic countries for onchocerciasis in Africa, with the disease being endemic in around half (49%) of the country [7]. The most highly endemic foci of onchocerciasis in South Sudan are in Western Equatoria, Northern and Western Bahr el Ghazal areas, and part of Central Equatorial [8].

The main strategy for onchocerciasis control and elimination is Community-Directed Treatment with Ivermectin (CDTI) [9]. However, as ivermectin is only a microfilaricidal drug that eliminates microfilaria in the skin and temporarily sterilises female adult worms in the nodules but without killing them. Mass drug administration with ivermectin needs to be given to the targeted population for at least 15 years which reflects the lifespan of the adult worms [1]. Moreover, to eliminate onchocerciasis, a CDTI coverage of at least 80% of the ivermectin administered to eligible people in the targeted community is needed [10]. Pregnant and breastfeeding women (less than one week postpartum), and children under the age of five years (<90 cm or <15 kg) are not eligible for ivermectin because of lack of safety data.

Using CDTI, the African Programme for Onchocerciasis Control (APOC) was successful in eliminating onchocerciasis as a public health threat in several African countries [11]. However, in many onchocerciasis-endemic areas in Africa, particularly those that have experienced periods of insecurity and where CDTI coverage has been sub-optimal or interrupted, there is still high onchocerciasis transmission and a high prevalence of onchocerciasis-associated morbidity including Onchocerciasis-Associated Epilepsy (OAE) [12]. This is the case in many areas in South Sudan [13,14]. Only after the ceasefire of 1995, Non-Governmental Organizations (NGOs) such as HealthNet International initiated mass drug treatment administration with ivermectin [15]. The national onchocerciasis control programme of South Sudan, established in 1996, initiated CDTI activities by 2004 [16]. However, frequent security concerns hampered the effective yearly implementation of CDTI. Hence, onchocerciasis-related morbidity (dermatitis, eye disease, and epilepsy) remain prevalent in these areas with high ongoing transmission. Following an assessment by the Health Department of the Catholic Diocese of Wau (Western Bahr el Ghazal state) in 2021, it was discovered that many parishes within Raga County had a high number of blind people and people with epilepsy. The affected parishes requested the health department to conduct a surgical eye care campaign for the blind people and to provide treatment for persons with epilepsy. Eventually in March 2023, an ophthalmological research team (headed by KLLS, ophthalmologist) visited the area to determine the causes of eye diseases in Raga and Deim Zubeir. The results of these ophthalmological investigations will be published elsewhere. Given that oncho-

cerciasis could be a main cause of ocular disease in Raga County, the research team also decided to document the community's knowledge, attitudes, and practices towards onchocerciasis and CDTI in the visited villages. The findings from such a study could lead to optimized health education strategies to increase the intake of ivermectin during the CDTI program, thereby improving onchocerciasis elimination prospects in those sites.

Methodology

Study setting

The study was conducted from 21st March to 2nd April 2023 in the Raga County, Western Bahr el Ghazal state of South Sudan. In 2022, the United Nations Office for the Coordination of Humanitarian Affairs (UN-OCHA) estimated the population of Raga County at 59,638 [21]. The county's headquarters is Raga town. The population of Raga County consists of various ethnic groups, including the Fertit people, who are predominantly farmers (sorghum, millet, maize, vegetables) and traders. Cattle herding and fishing are also practiced. The area has faced hardships due to conflicts, displacement, and limited access to basic amenities such as healthcare, education, clean drinking water and infrastructure. In Raga County, there are six fast-flowing rivers and many seasonal streams which provide conducive environments for blackfly breeding. As the main river in the Raga onchocerciasis focus is the Jur River, onchocerciasis is also known there as Jur River Blindness.

Study design and sample size

We conducted a cross-sectional community-based survey following the Knowledge, Attitude and Practices (KAP) approach. The sample size was calculated using the Cochran's formula for studying proportions: $n = z^2 pq/d^2$ [21]. Since there was no prior data on onchocerciasis in the study site, a conservative prevalence of $p=50\%$ was assumed. Using this formula and considering $z=1.96$ and $d=0.05$, $n=385$ participants had to be recruited into the study. Considering a 20% non-response rate, the final minimal sample size was 462 per study group (eye patients and carers). Therefore, the overall required minimal sample size was calculated as: $N=462 \times 2=924$. Hence 462 participants from each group.

Selection of study participants

Within the parishes of Raga County, we selected villages that have a prayer centre/church and a catechist in charge of the congregation. This facilitated the easy mobilization of community members. Selected villages included: Abulu, Kuru, Deim Zubeir, Sopo, Mangayat, Raga, Kata, Boromedina, Ferika, Sira Malaga and Timsah.

As this KAP study was conducted concomitantly with an ophthalmological care mission, mostly persons with eye problems and their carers were mobilized in the study villages. Systematic random sampling was used to select participants among the patients with eye disease and their carers who showed up at the recruitment site (church/prayer centre). Participants with eye disease were seated on benches and every fifth patient was selected to be recruited in the study. Carers were lined up behind the participants with eye disease, and the carer of every third eye diseased patient was selected for recruitment. This method was used to avoid the selection of carers from the same household as the recruited eye patient.

Study procedure: Study participants were interviewed using a standardized questionnaire adapted from a study by Weld-

egebreal et al. [23]. The questionnaire included questions about socio-demographic characteristics, and structured close-ended questions relating to knowledge, attitudes, and practices about onchocerciasis and CDTI.

Data analysis: Results were entered into an Excel spreadsheet and later transferred onto SPSS version 21 for analysis. Descriptive statistics were performed by summarizing continuous variables as medians with Interquartile Ranges (IQR), and categorical variables as percentages.

Ethics statement: Ethical approval was obtained from the ethics committee of the Ministry of Health of South Sudan (June 2023, MOH/ERB/P/35/15/05/2023) and from the ethics committee of the Antwerp University Hospital, Belgium (April 2019, B300201940004). The study was conducted following the principles outlined in the Declaration of Helsinki. Confidentiality and free-will participation were ensured. Oral informed consent in the presence of a witness was obtained from all potential participants and from the parent/guardian of each participant under 18 years of age.

Results

Of the 963 persons asked to participate, 932 (96.8%) participated in the study; 56.4% (526) were males and 43.6% (406) females (Table 1). The median age of participants was 48 years (IQR: 37-60). Most participants (88.1%) were married, and more than half (53.1%) had not received any formal education (Table 1). Almost one third (30.6%) of participants were from Raja town, and another 13.9% from Deim Zubeir town; the rest from smaller villages.

Participants' knowledge about onchocerciasis

The majority, 93.8% (874) of participants had heard of onchocerciasis and 83.4% (777) knew that onchocerciasis is transmitted by blackfly bites. One third of participants, 33.5% (312), agreed that blindness is the main sign of onchocerciasis, and 77.8% (725) upheld that onchocerciasis is a preventable disease (Table 2).

Table 2: Knowledge of 932 community respondents about onchocerciasis in Raga County, March 2023.

Variable	Frequency	Percentage (%)
Have you ever heard about the disease called onchocerciasis?		
Yes	874	93.8
What causes onchocerciasis?		
Filarial worm	13	1.4
Black fly	796	85.4
Mosquito	24	2.6
Living in poor environmental sanitation	5	0.5
Eating contaminated food	1	0.1
Being not vaccinated	1	0.1
I do not know	92	9.9
How is onchocerciasis transmitted?		
Blackfly bite	777	83.4
Contact with infected person	5	0.5
Mosquito bite	34	3.6
Through breath	17	1.8
I do not know	99	10.6

Table 1: Socio-demographic characteristics of 932 participants in Raga County, March 2023.

Variable	Frequency	Percentage (%)
Gender		
Male	526	56.4
Female	406	43.6
Age group (years)		
10-19	9	1.0
20-29	93	10.0
30-39	172	18.5
40-49	221	23.7
50-59	175	18.8
60-69	138	14.8
70 or more	124	13.8
Marital status		
Married	825	88.5
Unmarried	84	9.0
Others	23	2.5
Educational Status		
Unschooling	495	53.1
Primary level	318	34.1
Secondary and above	119	12.8
Occupation		
Farmer	508	54.5
Civil Servant	136	14.6
House wife	56	6.0
Business person	55	5.6
Others	177	19.0
Family Size		
1-4	226	24.2
5-10	476	51.1
>10	230	24.7
Place of Residence		
Abulu	49	5.3
Kuru	49	5.3
Deim Zubeir	130	13.9
Sopo	17	1.8
Mangayat	80	8.6
Raga	285	30.6
Katta	33	3.5
Boro Medina	73	7.8
Firka	73	7.8
Sir Malaga	74	7.9
Timsah	69	7.4

Up to 3.2% of participants reported having epilepsy, and in 16.8% of all participants, there was a family history of epilepsy.

What are the signs and symptoms of the disease?		
Itching	190	20.4
Edema	11	1.2
Skin change	419	45.0
Blindness	312	33.5
Do you think onchocerciasis is a preventable disease?		
Yes	725	77.8
No	41	4.4
I do not know	166	17.8

Participants' attitudes towards onchocerciasis

Among the interviewed individuals, 375 (40.2%) reported that they/their family member had already been sick from onchocerciasis. Most participants (856/932, 91.8%) considered onchocerciasis as a serious disease, with 93.2% affirming that onchocerciasis need to be treated (Table 3). Ivermectin (Mectizan) was approved as the appropriate treatment by 98.2% of participants. Only 4 (4.0%) participants viewed CDTI as not useful in preventing onchocerciasis infection (Table 3).

Table 3: Attitude of the 932 participants towards onchocerciasis in Raga County, March 2023.

Variable	Frequency	Percentage (%)
Have you/your families ever been sick from onchocerciasis?		
Yes	375	40.2
No	446	47.9
I don't know	111	11.9
Is onchocerciasis a serious disease?		
Yes	856	91.8
No	13	1.4
I do not know	63	6.8
Do you think onchocerciasis needs treatment?		
Yes	869	93.2
No	6	0.6
I do not know	57	6.1
Do not know		
What is the type of treatment used for onchocerciasis?		
Modern	923	99
Traditional	9	1
If modern, which drug is needed to treat the disease		
Ivermectin/Mectizan	915	98.2
Albendazole	17	1.8
How do you/your family perceive ivermectin distribution (CDTI)?		
Very useful	783	84.0
Partially useful	30	3.2
Not useful	4	0.4
I do not know	115	12.3
Do you think the CDTI program is effective in controlling onchocerciasis?		
Yes	760	81.5
No	22	2.4
I do not know	150	16.1

Participants' practices towards onchocerciasis

The main practice for onchocerciasis prevention was to take the drug (ivermectin), as reported by 605 (64.9%) of participants. This was closely followed by the practice of wearing protective clothing as mentioned by 218 (23.4%) participants (Table 4). A minority also adopted that personal and environmental hygiene as preventive measures against onchocerciasis. Most participants (83.0%) reported taking ivermectin every distribution round as their contribution to CDTI; from a community perspective, 80.3% of participants agreed that all eligible members of their respective households had received ivermectin. Frequent reasons for missing ivermectin treatment included: Absenteeism during CDTI (89.4%), health problem at the time of distribution (7.0%), and pregnancy (3.6%). Most participants (83.3%) received their last treatment during the previous year, and 55.6% experienced side effects.

Table 4: Participants' practices regarding onchocerciasis prevention including Community Directed Treatment with Ivermectin (CDTI) in Raga County, March 2023.

Variable	Frequency	Percentage
What do you do to prevent onchocerciasis?		
Avoiding river bathing	69	7.4
Wearing protective clothes	218	23.4
Taking the drug (ivermectin)	605	64.9
Using bed net	24	2.6
Environmental sanitation	4	0.4
Personal hygiene	12	1.3
Concerning the wearing of protective clothes, in what way are they used?		
To cover the lower extremities (below the knees)	125	13.4
Around head & shoulders	137	14.7
I don't know	670	71.9
How frequently do you receive the drug ivermectin in a year?		
Once	684	73.4
Twice	40	4.3
Three times or more	23	2.5
I do not know	185	19.8
When did you received your last ivermectin treatment?		
Last year	776	83.3
Two or more years ago	156	16.7
Did you have any side effects of the drug?		
Yes	518	55.6
No	307	32.9
I don't know	107	11.5
What is your contribution in the CDTI?		
Taking the drug each round	774	83.0
I do not know	158	17.0
Have all eligible people in your household received ivermectin during the last CDTI?		
Yes	748	80.3
No	184	19.7
If your answer above is no, who missed the treatment? (n=184)		
My wife	16	8.7

My husband	11	6.0
Another family relative	115	62.5
My children	42	22.8
What are the reasons for you to have missed ivermectin treatment in the past?		
Pregnancy	34	3.6
Health problem	65	7.0
Not present during drug distribution	833	89.4
Do you know anyone in the village who has interrupted his/her ivermectin treatment?		
Yes	199	21.4
I do not know	733	78.6
If your answer is yes, what was the reason to interrupt the treatment? (n=199)*		
Side effect of drug	144	72.4
Lack of good case management of side effect	62	31.2
*Possibility to choose both options, hence a total of 206 responses from 199 respondents		

Discussion

Our study showed that the majority of study participants had a relatively good knowledge about onchocerciasis. The majority of them (83.4%) mentioned onchocerciasis is transmitted by blackfly bites. However, they also considered blackflies (85.4%) as the cause of onchocerciasis. Only 1.4% knew that a filarial worm is the cause of the disease. Similarly, in a survey conducted in 2022 in Southwest Ethiopia, 75% of the respondents considered blackflies as disease transmitter and cause of the disease while only 23.5% knew the causative agent of onchocerciasis [24]. In a survey conducted in 2015 in the Bandundu province in the Democratic Republic of Congo (DRC), only 49.8% considered blackflies as the cause of onchocerciasis and 7.8% a filarial worm [25].

In our study, skin changes, itching and blindness were reported as main signs and symptoms of onchocerciasis. A survey conducted in 2010 in Osun State in Nigeria, revealed that onchocerciasis was well known by its local name by 91.6% of the respondents but only 2.4% of them attributed the disease to blackfly bites and only 1.4% knew that it affects both the eyes and skin [26].

In our study most people (91.8%) considered onchocerciasis as a serious disease that requires treatment. Nearly all (98.2%) mentioned that onchocerciasis need to be treated with ivermectin (Mectizan). Most of them (77.8%) agreed that onchocerciasis is a preventable disease. This contrast with a study in 2021-22 conducted among Selamogo residents in the South Omo Zone, a hard to reach zone in Southern Ethiopia, where only 7.3% of the respondents considered onchocerciasis as a preventable disease [27]. However, other studies in Southwest Ethiopia and Northwest Ethiopia demonstrated better knowledge, with respectively 88.2% and 93.3% of the respondents admitting that onchocerciasis is preventable [28]. The fact that up to 84% of our participants considered CDTI to be useful to prevent onchocerciasis is encouraging as they would be likely to adhere to CDTI. In a study conducted in 2014-15 in Edo state in Nigeria, only 58.8% of the respondents knew onchocerciasis can be prevented [29]. In another survey in 2017-18 in the Southwest region of Cameroon 82.3% perceived CDTI as a very useful program and the majority (79.0%) responded that all

their eligible family members took the drug annually [30]. In the Bandundu province in the DRC, approximately 55.1% of respondents were considered to have good knowledge of onchocerciasis and CDTI but only 37% of them had a favorable attitude and 46% a positive perception about CDTI [25]. In a study conducted in an onchocerciasis endemic area in Osun State in Nigeria 89% of the respondents were unaware of any treatment for onchocerciasis [31].

The majority (83.3%) of our participants took ivermectin during the last CDTI round. This is in concordance with the fact that 12.3% reported that relatives missed the treatment. The main reason for not to take ivermectin was absenteeism during distribution. Raga and Diem Zubeir are areas where people are involved in agriculture, fishing, honey harvesting and wild animal hunting. For most of these activities, people need to go away from their houses. The CDTI programme should synchronize their activities with seasons where most, if not all, community members are present in their houses. Furthermore, community drug distributors should be properly motivated and/or incentivized to visit all household (possibly multiple times if the compound is initially found empty).

A large proportion (55.6%) reported to have experienced side effects after the intake of ivermectin. These are likely not severe and certainly not fatal, as this zone is not endemic for loiasis (can cause deadly encephalopathy upon taking ivermectin [32]). Side effects of ivermectin occur mainly in *O. volvulus* infected individuals and include pruritus, painful skin oedema, arthralgia, bone pain, thoracic pain, malaise, headache, and fever [33]. They are most pronounced in persons with a high microfilaria load, never been treated before with ivermectin, and generally disappear within a week [33]. Persons with severe reactions can be treated with acetylsalicylic acid and antihistamines [33].

Up to 3.2% of participants reported to have epilepsy, and 16.8% of them reported to have a family history of epilepsy. In onchocerciasis endemic areas with high ongoing *O. volvulus* transmission the highest epilepsy prevalence is observed in the 10-30 age group [13,14,34]. As this age group was underrepresented in our study sample it may be that the epilepsy prevalence in the total population maybe even higher than 3.2%. The prevalence of epilepsy in Raga County may be similar to the epilepsy prevalence in other onchocerciasis-endemic areas in South Sudan with high *O. volvulus* transmission such as in Maridi [13], Amadi [14] and Mvolo [34]. This epilepsy prevalence is much higher than the 1.4% median epilepsy prevalence in sub-Saharan Africa [35].

To the best of our knowledge, this is the first published KAP study about onchocerciasis and CDTI in South Sudan. While it finds strengths in its large sample size and collecting data from hard-to-reach and often forgotten areas, it also has some limitations. We only conducted a questionnaire survey among a convenient sample of the population who showed up for ophthalmological issues. As 56.4% of the study participants were males and few were younger than 30 years (11%), men and older people were over-represented in the sample. Therefore, the study participants were not representative of the general population. Moreover, as participants were persons with eye problems or their carers it is possible that they were more knowledgeable about onchocerciasis and more supportive of the CDTI programme than the general population. Data about who, responded to the questions (person with eye disease or carers) was not collected. Therefore, we were unable to compare the responses of those two categories of study participants.

Also, our quantitative survey made use of close-ended questions with suggested answers for participants to choose from, thereby missing out on deeper perceptions which could be assessed via a qualitative design. It would be very useful to complement our findings with focus group discussions among community members, healthcare providers and in-depth interviews of key persons involved in organizing the CDTI such as community distributors of ivermectin and public health officials. This would offer the opportunity to discuss more aspects and also obtain inputs from the community about measures to take to increase CDTI coverage.

Conclusion

Even though the community had a relatively adequate knowledge, attitude and practice about onchocerciasis and CDTI, there were still deficiencies in information about the causative agent, mode of transmission and prevention of onchocerciasis with misbeliefs. Coupled with absence during drug distribution, this could have a negative impact on the success of the onchocerciasis eradication programme. Hence, this study revealed the need for increasing the awareness about onchocerciasis in the area through community based campaigns during drug distribution. Therefore, the National Ministry of Health in collaboration with the State Ministry of Health and NGO partners need to increase awareness about onchocerciasis in these areas by conducting community based awareness campaigns especially during drug distribution. Such a campaign should stress on the importance of the CDTI programme to prevent onchocerciasis-associated morbidities including onchocerciasis-associated epilepsy. Moreover, it will be important to explain the potential side effects of ivermectin and to provide access to medication to counteract these side effects. Furthermore, there is a need to synchronize drug distribution with seasons of less “out of house” activities so that a maximum drug uptake is achieved. Additional qualitative research may be needed to plan and optimize the implementation of CDTI campaigns.

Declarations

Authors summary: Onchocerciasis (river blindness) is a neglected tropical disease associated with a high burden of disease in South Sudan. This study delves into the community’s awareness, beliefs, and practices regarding onchocerciasis and its prevention through Community-Directed Treatment with Ivermectin (CDTI). The community displayed a commendable understanding of onchocerciasis with 78% recognizing it as preventable. The majority (84%) appreciated the value of CDTI in preventing onchocerciasis, showcasing its potential impact. Despite positive sentiments, over half (56%) of the interviewed individuals experienced side effects from ivermectin, indicating a need for better information and support. About 12% of participants reported family members missing treatments, often due to being absent during distribution. These findings underscore the importance of enhancing community engagement via awareness campaigns, improved communication about treatment benefits and potential side effects, and collaborative decision-making particularly regarding the timing of CDTI to foster the fight against river blindness in South Sudan.

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Availability of data and materials: Data will be made available for additional analyses upon request.

Competing interests: The authors declare that they have no competing interests.

Authors’ contributions: KS and JL are responsible for the concept, designing and the research, and supervising the work. JL, SM, and JP conducted the interview and are responsible for filling up the questionnaire from the study participants. TG and JP coordinated research activities and designed the budget, plan and resource mobilization for the financial support. KS and JL carried out statistical analysis and data interpretation. RC, TF, JT, KS, KA, AC, JSF, SJ, and JL edited and modified the manuscript. All authors read and approve the manuscript.

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