

# Cystic echinococcosis in Mundari tribe-members of South Sudan

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Many neglected tropical diseases, including the zoonotic disease cystic echinococcosis (hydatidosis), are endemic to East Africa. However, their geographical distribution is heterogenous and incompletely characterized. The aim of this study was to determine if Mundari pastoralists harbor endemic human hydatidosis. The survey was conducted in cattle camps randomly selected from accessible sites provided by officials in Terekeka, South Sudan. Following informed consent, a questionnaire collected demographic data and hydatid exposure risk. A systematic sonographic abdominal exam was performed using General Electric's LOGIQ Book XP with a 3C-RS 2–5 MHz curvilinear transducer. Six hundred and ten individuals were screened from 13 camps. Four infections were identified, all in women. The prevalence of abdominal hydatid disease in the Mundari tribe-members in cattle camps was 0.7% and all individuals reporting at least one high-risk exposure to hydatid disease. Cystic echinococcosis is endemic among Mundari pastoralists; however, it would appear to be less endemic than in neighboring tribes.

**Keywords:** Echinococcus/echinococcosis, Hydatid/hydatidosis, East Africa, Pastoral, Ultrasound/sonographic, Zoonotic/zoonosis

## Introduction

Mapping of neglected tropical diseases to target public health interventions is ongoing in South Sudan.<sup>1,2</sup> Determining disease prevalence among various tribal groups and the geographic extent of these infections is important for planning effective and affordable disease control strategies.

Cystic echinococcosis (hydatid disease or hydatidosis) was recently added to the World Health Organisation (WHO) list of zoonotic neglected tropical diseases.<sup>3</sup> In addition to livestock intermediate hosts, hydatid disease is endemic among Toposa and Bouya pastoralists in South Sudan and multiple Kenyan, Somali, and Ethiopian pastoral tribes.<sup>4–6</sup> However, the distribution and prevalence of the disease elsewhere in South Sudan is unknown. Many of the approximately 600 tribes in Sudan are pastoral and potentially at risk of cystic echinococcosis. Given the difficulties associated with the diagnosis and treatment of this disease in resource-limited settings, defining groups in need of intensive disease control is vital for targeted, cost-effective interventions.

In addition to the clinical and public health concerns raised by this disease in low-resource

settings, cystic echinococcosis exacts a toll on the economies of pastoral peoples. Studies from Tunisia and Ethiopia have independently shown that in countries with large pastoral economies, cystic echinococcosis can have a substantial negative impact on local and national livelihoods.<sup>7,8</sup> Consequent to a reliance on scarce natural resources and cattle, pastoralists and their livestock are particularly vulnerable to drought, food shortages, lack of access to grazing lands, conflict, and disease.<sup>9</sup> Therefore, cystic echinococcosis can have a disproportionate impact when superimposed on these other factors and jeopardize an already precarious way of life for a substantial proportion of East Africans.

We sought to continue mapping the distribution of this disease among pastoralists by determining the prevalence of abdominal hydatidosis in the Mundari tribe of South Sudan using sonographic examination. In addition, we attempted to characterize those particular behaviors that might place the tribe at risk of human cystic echinococcosis.

## Methods

### *Ethical considerations*

Ethical approval was granted by institutional review boards of the London School of Hygiene and Tropical Medicine, the Ministry of Health of the Government of South Sudan and Terekeka State.

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**Figure 1** Schematic maps of South Sudan and Terekeka.

Before screening an area, the local Government of South Sudan Primary Health Care Center was made aware of the study and taught about the necessary monitoring of any participants that were started on anti-helminthic chemotherapy. Study staff and a Mundari representative met with chiefs and elders of each cattle camp (nomadic pastoral temporary settlement) to explain the survey procedures in detail. All communication was exchanged in local dialect and informed consent was taken from the chief, who served as the consent of the entire camp. Each participant was then asked to provide informed assent before enrollment.

After examination, non-pregnant participants with active hydatidosis and without sonographic evidence or a history of hepatitis, were counseled and offered treatment according to standard-of-care.<sup>23–28</sup> Participants started on chemotherapy were either taken to the nearest Primary Health Care Center to introduce them to the facility handling their care or were asked to attend there within two weeks and monthly thereafter for follow-up. After being screened, the camp chiefs organized the inhabitants into groups that discussed prevention strategies for hydatidosis and other fecal–oral transmitted diseases in a local language with study staff members.

#### *Study area and site selection*

The survey was conducted in Terekeka, South Sudan (Fig. 1). Terekeka is bounded to the east by the White Nile and the west by Lake Madi, covering approximately 18 400 km<sup>2</sup> and carrying 144 300 people. This area is dominated by the Mundari Tribe. In the dry season, cattle camps converge on the Nile River for reliable water supply (Fig. 2). During South Sudan's rainy season, from June through October, the camps disperse to less competitive grazing areas. The present study took place during August and September, which

made access to many of the camps difficult due to flooding and washouts.

To simply determine the presence or absence of hydatid disease in the Mundari Tribe, the target sample size was 500 individuals. With the help of county administrators and local pastoralists, a list of cattle camps was created. Camps were separated into two categories (non-accessible and possibly-accessible/accessible) and 10 were randomly selected from the latter category. Camps were individually mobilized the day before the study and all adults, defined as older than 13 years of age or able to participate in scarification ceremonies, that provided informed consent were interviewed using a structured questionnaire and examined using ultrasound.

#### *Study procedures*

##### **Verbal questionnaire**

After informed consent, chiefs responded to a structured questionnaire about the number of livestock in camp, prevalence of hydatid cysts in slaughtered animals, role of dogs in their way of life, uses of animal excreta and products, and general knowledge of cystic echinococcosis in humans. Individual participants who gave informed assent answered questions related to hydatid exposure including: age, sex, level of education, type of livestock cared for, whether or not they handled offal and access of dogs to offal, cooking vessels and living areas.

##### **Ultrasonographic examination**

Systematic abdominal and pelvic ultrasonographic exams were performed by a single examiner using a General Electric LOGIQ Book XP (General Electric Healthcare, Barrington, IL, USA) with a 3C-RS 2–5 MHz curvilinear transducer. Examinations of the liver, pancreas, kidneys, spleen, bladder, and pelvic organs in two perpendicular planes and with multiple sweeps were performed.



**Figure 2** A man from the Mundari tribe stands among cattle. Photo by Kyodo/Landov.

Cystic structures resembling hydatid disease were characterized using WHO staging criteria for human hydatid disease:<sup>10</sup>

- CL: unilocular cystic lesion with uniform anechoic content without a well-defined hyperechoic rim;
- CE 1: unilocular simple cyst with visible wall and uniform anechoic content or fine intracystic echoes due to shifting of brood capsules ('hydatid sand');
- CE 2: multivesicular, multiseptated cysts with a visible wall which may form a 'wheel-like', 'rosette-like', or 'honeycomb-like' appearance due to daughter cysts;
- CE 3: unilocular cyst that may be less rounded due to loss of intracystic pressure which may contain daughter cyst(s) filled with anechoic content and the detached laminated membrane from the cyst wall which appears as a floating membrane, the 'waterlily sign';
- CE 4: heterogenous, hypoechoic, or hyperechoic degenerative contents without daughter cysts, may resemble a 'ball of wool';
- CE 5: cystic structure characterized by a thick calcified wall, often arch shaped producing a cone of shadow.

**Table 1** Characteristics and response of individuals sonographically screened for hydatid disease

Characteristic	N=610
Age, median (IQR)	25 (19–32)
Male, <i>n</i> (%)	481 (79)
Occupation, <i>n</i> (%)	
Pastoralist	570 (93)
Farmer	11 (2)
Other	29 (5)
Livestock care*, <i>n</i> (%)	
Cows	587 (96)
Goats	592 (97)
Sheep	584 (96)
Other	0 (0)
Dog behavior†, <i>n</i> (%)	
Free to roam	544 (89)
Scavenge from pots	580 (95)
Important to way of life	581 (95)
Educational level, <i>n</i> (%)	
None	580 (95)
Primary	15 (3)
Secondary	7 (1)
>Secondary	8 (1)

Note: \*Participants who responded that they care for a particular species of livestock, not mutually exclusive.

†Participants who responded that dogs are free to roam around the camp, scavenge from pots and are an important to their way of life.

### Data analysis

Data analyses were conducted in STATA 10 (College Station, TX, USA). Descriptive statistics were calculated for the study population. Proportions and 95% confidence intervals (CIs) were calculated using the exact binomial distribution.

### Results

In total, 610 individuals and 13 chiefs from 13 cattle camps were screened. The median age of participants was 25 years (inter-quartile range: 19–32). Seventy-nine percent of participants were male. Only 5% of participants reported attending any school. The majority of individuals were solely pastoralists (93%) (Table 1).

On average, chiefs and butchers reported that around 30% of slaughtered animals to have cystic structures in abdominal viscera (range: 0–6 out of 10).



All chiefs reported knowing about cystic echinococcosis in at least one of their slaughtered livestock since the last rainy season. They also reported that individuals living in their cattle camp ate animals with cystic structures seen at slaughter after cysts were cut out and all products of the dead animal either well cooked or boiled before consumption. No chief endorsed individuals consuming raw or purposefully undercooked livestock products, a known proxy for risk of hydatidosis. Similarly, all chiefs were aware of the potential risk of human infection with hydatid disease. Every chief reported that the camp did not use fecal material from the camp's floor for hut construction or bedding material.

Livestock, though owned by families, are cared for communally. Therefore, every participant reported caring for or sleeping in proximity with goats, sheep, and cows. No one reported caring for camels or swine, nor were any observed in the camps during the study period. Most individuals (89%) stated that dogs are free to roam around the compound. Almost 95% of individuals screened reported sharing familial cooking vessels with dogs. Nearly 97% of screened individuals reported leaving offal for dogs to eat or giving offal directly to dogs. No one practiced offal burial or burning.

The abdominal sonographic prevalence of hydatidosis was 0.7% (4/610, 95% CI: 0.2–1.7%). All infections were seen in women (female 4/129, 3.1%, 95% CI: 0.9–8.0%). The average age of these individuals was 46 years. No recorded factors differed between these women and the rest of the screened sample on multivariate analysis. All four lesions were intrahepatic. Two of the four complex cysts were consistent with WHO *E. granulosus* Cyst Type 4 or 5 by being amorphous cysts with mixed echoic contents, calcified rim and a having a 'ball of wool' appearance. These stages are presumed not to produce daughter cysts and are considered transitional/inactive. The risks of disease progression and benefits of treatment were discussed through translators. Both participants agreed to forego any treatment and undergo clinical surveillance with PHCC staff. Of the other two women, the first had WHO Type CL, which was demonstrated by a unilocular cyst with a uniformly anechoic appearance without a well-formed cystic rim. This lesion may have been a simple, benign cyst. However, given her risk of hydatid infection, she was offered and accepted treatment. The last woman had Type CE1, a unilocular cyst with a general anechoic appearance, though notably having fine intracystic echoes consistent with hydatid sand. She too accepted treatment.

## Discussion

The present survey aimed to continue the estimation of hydatid disease among pastoralists in South

Sudan, specifically the Mundari Tribe of Terekeka. In addition, we attempted to document behaviors that might place the tribe at risk of cystic echinococcosis. The study found that hydatid disease is endemic in the Mundari tribe and women are at higher risk of exposure and subsequent infection than men.

The 0.7% prevalence described here is consistent with other estimates from sonographic screening studies of pastoral East African tribes. The most geographically proximate study involved the Toposa tribe in Eastern Equatoria, South Sudan, which found that 3% of sonographically screened persons had evidence of hydatid disease.<sup>11</sup> Other East African pastoral tribes were documented to have sonographic prevalence between 0.1% and 5.6%.<sup>12</sup>

Although we found no men with hydatid disease, 3% of women had evidence of infection. Livestock dung and dog excreta are both present on the camp floor. Women sweep debris from the camp floor into piles using bare hands and arms daily for burning. Subsequently, female Mundari tribe-members are at risk of acquiring infection through direct contact with dog feces on the camp floor. Mundari men oversee the livestock grazing and do not participate in this practice. This may substantially reduce the risk of cystic echinococcosis in male pastoralists by limiting their exposure from this gender-specific task. Higher burdens of hydatidosis in females has been repeatedly described.<sup>13,14</sup>

In accordance with the high prevalence of hydatidosis in veterinary studies across Sudan, the studied camps reported that nearly half of all dying animals had cystic disease in their viscera. However, relatively few people were found to have sonographic evidence of cystic echinococcosis. This has not been the case for other pastoral groups surveyed in East Africa, which may be partially explained by several ethno-cultural and ecological difference between tribes.

The Mundari avoid slaughtering healthy animals for consumption. Instead, only dying or recently dead animals are butchered for consumption. Cystic structures are conventionally excised before ingestion. Further, the Mundari do not eat raw meat nor drink unboiled blood or urine. This contrasts with other tribes from the same region, including the Toposa, who do consume raw blood and eat raw meat during slaughter or celebration and have been reported to have a prevalence of cystic echinococcosis of 3.5%.<sup>4,5</sup> Humans do not contract hydatidosis from protoscoleces within ingested cysts of uncooked meat. Instead, humans are accidentally infected from embryonated eggs in definitive host feces. However, numerous surveys have documented such behavior as a proxy for human infection, as it gives domesticated dogs access to infected viscera, perpetuating the transmission cycle.

Mundari pastoralists living in cattle camps sleep on cow hides, not on huts or on raised resting places made from dung. *Echinococcus granulosus*, the dominant species in eastern and northern Africa,<sup>15</sup> replicates in carnivorous intestine, not ruminant gut. Therefore, contact with cow dung does not transmit the infection. However, unintentional ingestion of dog excreta does transmit hydatid disease. Dogs that live in the cattle camp defecate on the camp floor alongside cattle dung and while contracting infection from simply living within dung structures is highly unlikely, the construction of huts with mixed excreta from the camp's ground without adequate hand-washing may provide exposure for transmission.<sup>16</sup> Since the Mundari do not sleep on or live within dung structures, their exposure risk may be less than other pastoral groups in the region.

The majority of ruminant infections occur as a result of ingestion of grass contaminated with carnivore feces. Eastern Equatoria and Turkana, geographically adjacent homelands of the Toposa, Bouya, and Turkana tribes, are inhabited by natural carnivores (lions, hyenas, and wild dogs) and *E. granulosus* reservoirs, such as antelope species.<sup>12</sup> Terekeka does not have natural carnivores or reservoirs, as these were killed or displaced during years of conflict. The lack of natural reservoirs is likely to impact the transmission cycle, reducing the amount of fecal contamination within grazing areas in Terekeka. In addition, Mundari dogs are trained to not venture outside of the camp to watch over cattle during grazing since there are no natural predators to protect them from. Instead, the canines remain around the camp with the women and elders who do not tend to livestock. This contrasts to the behavior of dogs in the Toposa, Bouya, and Turkana tribes, which must defend the livestock from natural predators while grazing. Since Mundari dogs generally do not defecate on grasses that are consumed by cattle, there may be limited *Echinococcus* transmission via this route.

Terekeka receives more than 900 mm of rainfall annually and lies in the flood plain of the Nile River.<sup>17</sup> Grazable grasses are plenty and competition for grassland is much less than in the drier areas inhabited by the Toposa, Bouya, and Turkana tribes, which often receive less than 300 mm of rain each year.<sup>18</sup> It is possible that the limited availability of grazable pastures in areas inhabited by the latter tribes leads to a higher density of contaminated grasses, in turn increasing livestock exposure to contaminated fecal matter, probability of infection and potential transmission to carnivores and humans.

This study had several limitations. Ultrasonography has a high inter-rater variability. To minimize this, comprehensive training was provided for the

ultrasonographer to create a systematic exam for capturing reliable images and sweeps to reduce potential misinterpretation. This study attempted to exhaustively survey all individuals in each cattle camp. However, some camps did not allow us permission to examine women in equal proportion to men. Subsequently, we may have missed sonographic evidence of cystic echinococcosis in those at greatest risk due to their intense exposure to camp floor excreta. In addition, we were not able to reach a number of cattle camps due to inaccessibility or insecurity. Theoretically, these camps may be at higher risk of hydatid disease than the camps we sampled; however, given their relative ethnocultural and ecological equivalence, it is unlikely that the prevalence in those camps differed significantly from those that were examined. Cystic echinococcosis infects the liver and other abdominal viscera much more frequently than extra-abdominal organs. However, since we only examined intra-abdominal sites of potential infection, extra-abdominal disease may have been missed. Some groups have used a combination of ultrasonographic and serologic examinations to reduce the false-negative screens in those with small intra-abdominal or extra-abdominal disease.<sup>19–21</sup> Two Peruvian studies recorded prevalences of hydatidosis to be 5.7–9.3% with abdominal ultrasonography and up to 18.2% with immunoblot testing.<sup>20,22</sup> However, there is not a sensitive serological assay for East Africa. Given no plausible reason why the Mundari would have a less than normal ratio of abdominal to extra-abdominal disease, we do not believe that this led to an inaccurate portrayal of the actual prevalence.

## Conclusion

Cystic echinococcosis is endemic among the Mundari pastoralist tribe living in cattle camps in Terekeka, South Sudan. However, the prevalence is less than other published reports from neighboring tribes of Eastern Equatoria and western Ethiopia. The Mundari live in an area with less grazing competition and without natural intermediate or definitive hosts of *E. granulosus*. In addition, they do not consume raw meat or unboiled blood and do not use animal excreta to build homes. Together, they may have a substantially lower exposure than their pastoral neighbours. As in many preceding studies, women harbour a disproportionate burden of hydatidosis compared to men likely related to gender-specific, high-risk tasks. Understanding the relatively low, though endemic, prevalence of this group may lead to useful health education targeted at limiting the exposure of other pastoral groups to this difficult-to-treat infection. To determine whether the present findings are more widely applicable, other pastoral

tribes should be screened for cystic echinococcosis and data on potential risk factors for *echinococcus* infection should be collected. If future studies corroborate the present findings, then targeted behavior-change education, such as cooking or boiling products of slaughter and avoidance of direct contact with camp floor excreta, may be an effective means to decrease transmission of hydatid disease.

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