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Research Article

Prevalence of Cystic Echinococcosis in Selected Pastoral and Agro-Pastoral Districts of Uganda

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Abstract

A cross sectional ultrasound screening survey for human cystic echinococcosis (CE) was undertaken in the pastoral dist ricts of Moroto, Napak, Nakapiripirit and Amudat in Karamoja sub-region; and agro-pastoral communities of Teso region, in the districts of Kumi and Bukedea. Other areas of the survey included: Nakasongola in Central region and Kasese district in the Western region of Uganda. A total of 3,636 participants were screened and 67 cases (1.84%) had CE. The organ most affected was the liver 61.2% (41/67), followed by the kidney 17.9% (12/67), spleen 10.44% (7/67), omentum 8.95% 6/67) and lungs 1.5% (1/67). All districts screened had positive cases with the highest prevalence occurring in Napak (3.9%) and the lowest in Nakapiripirit (0.45%). The prevalence of CE (0.5±0.3%) in South Karamoja (Amudat and Nakapiripirit) was significantly lower ($P<0.001$, $\chi^2= 18.98$) than in Central Karamoja (Napak and Moroto, 3.32±1.3%). The prevalence of CE in Teso region was found to be 1.21±0.8%, Kasese 2.15±1.2 % and Nakasongola 2.7±1.3%. The prevalence in south Karamoja was lowest. Overall, there was no significant difference ($P<0.05$; $\chi^2= 0.12$) in prevalence between males (1.7%) and females (1.9%). However, in Karamoja females (2.2% CI: 0.8-3.6) were more likely to be infected ($\chi^2= 16$; $P<0.05$) with CE than males (0.9% CI: 0.1-1.7). Sixty four percent (n=43) of the cysts detected were viable.

INTRODUCTION

Echinococcosis is an endemic zoonotic infection found throughout the developing world [1]. It is a neglected emerging and remerging disease, that has not attracted the attention of the developed world [2]. Cystic echinococcosis (CE) is caused by infection with the larval stage of the cestode *Echinococcus granulosus*. The definitive hosts of this parasite are carnivores which are infected when they ingest the organs of herbivores that contain hydatid cysts developing into adult tapeworms within the carnivores. Infected carnivores shed tapeworm eggs in their faeces which contaminate the ground predisposing cattle, sheep, goats, donkeys, pigs and wild ruminants to infection through contaminated soil, pasture and water. Once ingested, the eggs hatch and develop into cysts called hydatid cysts in their internal organs. The most common mode of transmission to humans is also by the accidental consumption of soil, water or food that has been contaminated by the faecal matter of an infected carnivore especially dogs. The disease caused in humans is called hydatidosis. Hydatidosis is most commonly found in people involved in herding of herbivores together with carnivores (dogs) [3]. Therefore, CE has been associated with high morbidity and mortality among the pastoral communities like Turkana, Toposa, Nyangatom, Hamar and Boran [4,5].

Pastoral and agro-pastoral (PAP) areas in Uganda present conditions likely to favor occurrence of CE among humans. The WHO lists Uganda as one of the countries with high endemicity of cystic echinococcosis [6], however, there is scanty information on the Ugandan situation on echinococcosis. In Uganda, a Mulago hospital based retrospective study from 1967-1972 [7] found 23 cases of echinococcosis among the Karimojong, Lango and Acholi people. In another study [8] an average of 20 surgical cases per year has been seen in hospitals in Karamoja and Mbarara in Uganda. An earlier preliminary survey using ultra sound [8] found a prevalence of hydatidosis to be 0.4% (n=7) among 1605 people screened. Among the wild animals hydatidosis has been reported in 33.3% of lions, 10.4 % of warthogs and 17.2% of buffaloes examined on postmortem in Queen Elizabeth National Park [9]. Also hydatidosis had been reported in impala and goats in Lake Mburo National park [10]. In dogs, prevalence of *E. granulosus* of 66.3% (n=217) had been reported on autopsy in Moroto district in Uganda [11]. Also a cross sectional survey done in Kasese district in western Uganda revealed that lack of knowledge about transmission of echinococcosis was the major risk factor for transmission of echinococcosis among humans [12]. It was against this background that a study was carried out to determine the prevalence of echinococcosis infection in PAP communities of Uganda.

MATERIALS AND METHODS

A cross sectional screening survey was undertaken from 2012 to 2014. The screening was done in the pastoral districts of: Moroto, Napak, Nakapiripirit and Amudat in Karamoja region. In agro-pastoral communities of Teso region, the districts of Kumi and Bukedea; Nakasongola in central region and in Kasese district in the Western region. These districts were purposively selected to represent PAP areas. A total number of 3,636 volunteers were screened as shown in Table 1. Permission was sought from Uganda's Ministry of Health Research Council and Makerere University College of Health Sciences Ethical and Institutional Review Board. Field activities clearance was sought from the District Health Officer, the Chief Administrative Officer and village Local Councils. Volunteers (study participants) were mobilized among communities in selected sub-counties parishes and villages respectively; screening was conducted at various health centers.

On the day of screening, discussions were first held with participants, explaining the purpose for screening. A portable ultrasound machine powered by a portable generator was used for screening. Questionnaires were used to capture data from consented adults or assented guardians for the minors who participated in the study. Volunteers were assured of free diagnosis with those found positive being referred to regional hospitals for further management.

In a well lit room, volunteers were asked to lie on the examination couch and expose the entire chest and abdomen. Examination gel was applied to the abdominal skin and an ultra-sound examination probe (Sonosite Sitelink 2.2) was used for examination. The entire abdominal cavity was scanned in four known basic planes [13] that is: transverse, sagittal, oblique subcostal and coronal supracostal planes. All pathological conditions identified were recorded, measured and photographic images taken by ultra-sound machine. Photographic images were printed for further evaluation. Suspected hydatid cysts were all studied and classified according to WHO classification [14] as shown in Table 2 as simple cysts (CL), first clinical cyst types CE 1 and 2; second clinical type CE 3 transitional stage cysts and third clinical group CE types 4 and 5. Cysts with the size below 5 cm were taken as small, 5–10 cm were taken as medium and beyond taken 10 cm as large. Those found positive were further interviewed using a special designed form to find the probable predisposing factors for acquisition of CE infection.

Data analysis

Data was entered into MS Excel 16.0 spreadsheet. Statistical analysis was done using R Statistical software program *Version 3.2.0*). Chi-square at 95% confidence level were used to examine differences in CE prevalence by region, districts, location within districts, gender and organs affected.

RESULTS

Participants' ages ranged from 2 years to 85 years. The prevalence of cystic echinococcosis on ultra sound screening by district were as shown in Table 3. All districts screened had positive cases. The overall prevalence was 1.84%, with the highest prevalence being in Napak at 3.9% and the lowest in Nakapiripirit at 0.45% (Table 3). The prevalence of CE (0.5±0.3%) in South Karamoja (Amudat and Nakapiripirit) was significantly lower ($\chi2=18.98, P<0.001$) than in Central Karamoja (Napak and Moroto, 3.32±1.3%). The prevalence in South Karamoja was found to be the lowest. The variation of prevalence of CE according to regions other than south Karamoja was as shown in Figure 1. The Central Karamoja had percentage prevalence of 3.32±1.3%, Teso region 1.21±0.8%, Kasese 2.15±1.2% and Nakasongola 2.7±1.3%. The level of significance differences of CE prevalences according to regions were as shown in Figure 1. Prevalence of CE according to sex and their level of significant difference according to regions were as shown in Table 4.

There were 67 lesions identified in total. Some participants had multiple lesions in different organs. The liver had the highest number of lesions with 61.2% (41/67) followed by the kidney (17.9%), spleen (10.5%), omentum (9%) and the lungs (1.5%)

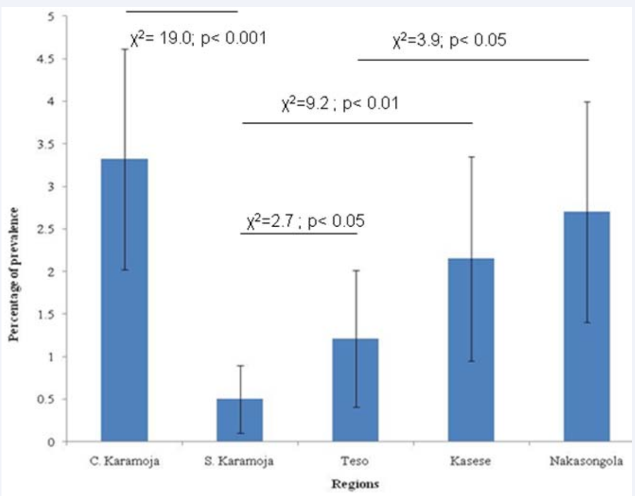

Figure 1 The prevelence of cystic ecchinosis among humans screened using ultrasound in postoral and agro- postoral regions in Uganda.

Table 1: Distribution of persons screened by gender and region.

Regions	Males	Females	Total	M:F ratio
Karamoja (North East)	580	1113	1693	1:1.9
Teso (East)	134	610	744	1:4.6
Nakasongola (Central)	166	429	595	1:2.6
Kasese (West)	437	167	604	1:0.4
Total	1317	2319	3636	1:1.8

Table 2: Details of classification of cysts detected using Ultrasound and their viability status.

Cyst type	Cyst status			Remarks
	Active	Fertile	Cyst wall	
CL	Yes	No	Not visible	Cysts are in early stage of development
CE1	Yes	Yes	Visible	Unilocular, anechoic or "snowflake sign"
CE2	Yes	Yes	Visible	Multiseptate and multivesicular, daughter cysts present
CE3	Transitional	Yes	Visible	Anechoic content with detached laminated membrane ("*waterlily* sign"). Decreased intracystic pressure. Cyst starting to degenerate
CE4	Inactive	No	Not visible	Heterogeneous hyperechoic or hypoechoic contents. No visible daughter cysts. "Ball of wool" sign due to degenerate membranes. Usually no viable protoscolices
CE5	Inactive	No	Calcified	Thick, variably calcified wall producing a cone-shaped shadow. Usually no viable protoscolices

Table 3: Prevalence of cystic echinococcosis in all the districts surveyed.

District	Number examined	Number positive	Prevalence (%)	Prevalence (%) at 95% confidence. interval
Amudat	338	2	0.59	0 - 1.4
Nakapiripirit	657	3	0.45	0 - 1.0
Moroto	290	8	2.7	0.8 – 4.6
Napak	432	17	3.9	2.1 – 5.7
Kumi	214	4	1.8	0 – 3.6
Bukedea	531	5	0.9	0.1 – 1.7
Nakasongola	595	16	2.7	1.4 – 4.0
Kasese	604	13	2.15	1.0 – 3.3
Overall	3636	67	1.84	1.4 – 2.3

Table 4: Prevalence of CE by sex and region.

Region	Percentage prevalence (95% confidence interval)			Level of significance difference at 95% confidence limit
	Males	Females	χ^2 Statistic	
Kasese	1.8 (0 - 3.8)	2.3 (0.9 -3.7)	0.14	P> 0.05 ns
Karamoja	0.9 (0.1 – 1.7)	2.2 (0.8 -3.6)	4.16	P<0.05*
Teso	2.3 (0 - 4.7)	1.0 (0.2 – 1.8)	1.4	P>0.05 ns
Nakasongola	4.2 (1.1 – 7.3)	2.1 (0.7 -3.5)	2.05	P>0.05 ns
Overall	1.7 (0.9 – 2.5)	1.9 (1.4 – 2.4)	0.12	P>0.05 ns

Ns: not significant difference; * significant difference

respectively. Details were as shown in Table 6. The prevalence of liver lesions with CE among males (1.55%) was not significantly different ($\chi2=2.13$, $P>0.05$) from that of females (0.97%). Males did not have lesions in the omentum or the lungs. No cysts were found to have more than one cavity. Details of classification of viability status and sizes of the cysts were as shown in Table 7.

Fertile cysts (CE1, CE2 and CE3) constituted 34.3% (n=23), active cysts (CL, CE1 and CE2) 61.2% (n=41), cysts in early development (CL) 29.9% (n=20), inactive cysts (CE4 and CE5) 17.9% (n=12); Cysts with viable status which were capable of causing clinical disease were 64% (n=43). Other disease conditions diagnosed with ultrasound screening were as shown in Table 7. Overall prevalence of leishmaniasis was found to be 2.85%.

Table 5: Number and percentage distribution of CE lesions according to different organs.

Organs	Number and percentage sex distribution		
	Males	Females	Overall
Liver	16 (64)	25 (59.50)	41 (61.20)
Spleen	4 (16)	3 (7.14)	7 (10.44)
Kidney	4 (16)	8 (19.00)	12 (17.91)
Omentum	1 (4)	5 (11.90)	6 (8.95)
Lungs	0 (0)	1 (2.40)	1 (1.50)
Total	25 (37.3)	42 (62.7)	67 (100)

Table 6: Viability status of the cysts found on ultra sound screening based on WHO Informal Working Group (2003) classification.

Type of cyst	Females	Males	Total	Percentage of total CE lesions
CL	13	7	20	29.9
CE1	12	5	17	25.4
CE2	1	3	4	6.0
CE3	2	0	2	3.0
CE4	2	0	2	3.0
CE5	5	5	10	14.9
Unclassified	7	5	12	17.9
Total	42	25	67	100

Table 7: *Echinococcus* infection and other disease conditions diagnosed by ultrasound in PAP communities of Karamoja, Teso and central regions of Uganda.

Condition	Region/district							
	Karamoja region				Teso region		Central region	Total
	Amudat	Moroto	Nakapiripirit	Napak	Kumi	Bukedea	Nakasongola	
Leishmaniasis	15 (4.4%)	3 (1%)	4 (0.6%)	15 (3.5%)	12 (5.6%)	6 (1.1%)	0	55
Congenital kidney disease	0	2	0	1	1	2	5	11
Splenomegaly	4	1	4	7	6	7	3	32
Other pelvic diseases	5	10	8	20	15	8	7	73
Total	24	16	16	43	34	23	15	171

DISCUSSION

This study showed that CE occurred in all pastoral and agro-pastoral study areas of Uganda with the overall prevalence of 1.84%. However, CE prevalence varied within regions and even intra-regionally. For example there were differences in Karamoja region where Napak district had a highest prevalence and Nakapiripirit lowest. Central Karamoja (Moroto and Napak) had the highest prevalence than South Karamoja (Amudat and Nakapiripirit districts). The differences could be due to differences in cultures and economic activities practiced by different ethnic groups referred to as the Karimojong. The people in the districts of Central Karamoja lived a pastoral life while those in the South were more sedentary. The largest Karimojong group in Central and southern Karamoja was subdivided into: Bokora occupying Napak district, the Matheniko in Moroto and Pian in Nakapiripirit North Karamoja were occupied by Jie in Kotido and Dodoth in Kaabong District. Meanwhile Amudat district was inhabited by the Pokot ethnic group [15]. The low CE prevalence observed in South Karamoja confirms a low prevalence (0.1%) earlier reported [4] among their neighboring Pokot pastoral communities in western Kenya. A similar prevalence of 0.2% was reported in Southern Turkana bordering the Pokot [4]. On the contrary, the high prevalence observed in Central Karamoja was in agreement with the high prevalence earlier observed [4] in Northwest Turkana (5.6%) and Northeast Turkana (2.7%) in Kenya; among the Toposa (3.1%) and Bouya (2%) people in South Sudan; and Nyangatom (2.9%) in Ethiopia. In a nutshell, this study confirmed the earlier postulations of spatial distribution of CE [4,16] that a high prevalence of CE occurred in circumscribed area covering North Western Kenya, North Eastern Uganda, South Eastern Ethiopia and Equatorial province of Sudan. This study has also confirmed that there was a high prevalence of CE in Napak and Moroto districts and the same may be true in neighboring districts where they shared grazing and watering areas during dry season.

Elsewhere, there was a low prevalence of CE in South Teso and South Karamoja (Figure 1). This could be attributed to differences in life styles, sources and level of water availability.

Central Karamoja, Nakasongola and Kasese practiced more of pastoralism. They experienced severe water shortages during dry season leading to high concentration of livestock with the accompanying dog population at watering points thereby increasing potential of transmission of CE between livestock, humans and carnivores [17]. Similar observations had earlier been made [4,18] whereby the Masai with similar life style to that of the Turkana had low prevalence of CE because they received more rainfall. Nakasongola was occupied by Bahima pastoralists who practiced nomadism and transhumance. While, Kasese was inhabited by Basongora who were pastoralists who often invaded Queen Elizabeth National Park for water and grazing [19]. In such areas, sources of water were open unprotected dirty water sources like: pools, dams, springs, lakes, rivers and swamps being shared between humans and animals including dogs. In such a situation, dogs' fecal matter can easily contaminate water sources with *E. granulosus* eggs. On contrary, in Teso where people live a sedentary life style, access to clean water was relatively better, water sources were often bore holes and protected wells where animal access was limited or completely denied.

Overall, there was no significant difference in prevalence between males and females with exception of Karamoja (Table 4). In this region, there was significantly a higher prevalence of CE in females than in males. This finding in Karamoja was in agreement with retrospective studies done in Uganda [7] and Tanzania in Ngoro Ngoro district [20]. Elsewhere, previous studies have reported a higher prevalence of CE in females than males [21-24]. There has been no clear explanation as to why sometimes there was sex difference in CE prevalence. Many suggestions can be put forward for this variation, however, gender roles in daily chores could promote increased interaction with the dog faeces, activities like fetching water, fruit and vegetable picking, tilling of land to grow crops, weeding of crops, eating of raw tubers like cassava and potatoes without washing and plastering of houses with mud could be playing a role. It has been observed that the dogs living in cattle camps defecate on the camp floor alongside cattle dung [25]. These dung mixtures when used for construction of huts and without adequate hand-washing may expose pastoral communities to a risk of infections with CE. This could be one of the most probable factors making women more prone to CE in Karamoja because it's the role of women to build and plaster the houses.

Our findings on visceral organ infectivity showed that the liver had the highest number of lesions followed by the kidney, spleen, omentum and lungs. This observation did not differ from earlier clinical surveys done elsewhere [22-24,26,27]. Also another epidemiological study done [4] based on surgical records, among the Masai people in Tanzania showed that the liver was the most affected organ. This was contrary to what was reported in Khartoum in Sudan where lung form of human CE was the most common [28-30]. This difference could be attributed to the differences of pathogenicity of strains of *E. granulosus* involved these areas. In Sudan, genotype G6 [31] was the most predominant as opposed to the G1 strain in Uganda [32,33].

In this study, cyst fertility rate was lower than expected. This finding agreed with what had earlier been observed [34] that cyst fertility detected by ultrasound was usually much lower than fertility detected by microscopic examination of harvested cysts. It has been found that the fertility rate of human hydatid cysts harvested and on direct microscopy was 75% [34]. Viability of 64% of the cysts detected in this study was higher than what was earlier reported of 57.1% [35]. This means that these cysts were capable of expanding or releasing antigens hence causing clinical disease or hypersensitivity reactions respectively. Viability of hydatid cysts have also a bearing on the type and success of treatment.

There were co-infections of CE encountered during the survey (Table 7). There was prevalence of 2.85% of suspected leishmaniasis infections. This condition was found in all the districts of Karamoja and Teso and not in Nakasongola. Amudat, Napak and Kumi registered the highest cases of leishmaniasis. Earlier study [36] had reported the occurrence of this disease in Amudat. This study was the first study to report the presence of the disease in Teso. The study reaffirms the earlier postulation [36] that this disease occurred in Karamoja and neighboring districts of Teso. Proper epidemiological surveys need to be carried out using appropriate gold standard diagnostic test to detect real magnitude of this disease so that control and preventive measures can be put in place. Elsewhere in Beira, Mozambique CE patients have been reported to have co-infections with cysticercosis, schistosomiasis and toxocariasis [37]. Also in China in Hui Autonomous Region CE patients had simultaneous infection with tuberculosis [38] which was not the case with our study.

It was concluded that CE was endemic and wide spread in PAP areas in Uganda, hence, challenging the original perception that the disease was absent in some regions of the country. Therefore, there was a need to create awareness of the hazards of CE to the local community and health workers in these areas. In addition, these frontline groups need to be educated about the epidemiology of CE as well as mitigation measures. There was also need for establishing screening and treatment units. Best model(s) of carrying out CE screening and treatment needs to be identified and adopted. Usually WHO assists countries to develop and implement pilot projects leading to validation of effective methods of controlling CE [3]. A policy and strategic plan for management of CE in Uganda needs to be created. In Uganda, there was no policy and a plan for management of CE. CE was not even listed in 2008-2015 health strategic plan as one of the neglected diseases in the country. This study therefore serves as an entry point for this disease to be considered as a serious medical issue in Uganda.

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