Epilepsia, 53(12):2194–2202, 2012
doi: 10.1111/j.1528-1167.2012.03687.x

FULL-LENGTH ORIGINAL RESEARCH

Prevalence of neurocysticercosis among people with epilepsy in rural areas of Burkina Faso

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Summary

Purpose: To estimate the lifetime prevalence of neurocysticercosis (NCC)–associated epilepsy and the proportion of NCC among people with epilepsy in three Burkina Faso villages.

Methods: Three villages were selected to represent three types of pig-rearing methods: (1) Batondo, where pigs are left to roam; (2) Pabré, where pigs are mostly tethered or penned; and (3) Nyonyogo, where the majority of residents are Muslim and few pigs are raised. In Batondo and Nyonyogo, all concessions (a group of several households) were included. Half of the concessions in Pabré were randomly chosen. All households of selected concessions were included, and one person per household was randomly selected for epilepsy screening and serologic testing for cysticercosis. Self-reported cases of epilepsy were also examined and confirmed cases included in analyses other than the estimate of NCC-associated epilepsy prevalence. Epilepsy was defined as ever having had more than one episode of unprovoked seizures. Individuals with medically confirmed epilepsy had a computerized tomography (CT) scan of the brain before and after contrast medium injection. The diagnosis of NCC was made using a modification of the criteria of Del Brutto et al.

Key Findings: Thirty-nine (4%) of 888 randomly selected villagers and 33 (94%) of 35 self-reported seizures cases were confirmed to have epilepsy by medical examination. Among the 68 participants with epilepsy who had a CT scan, 20 patients were diagnosed with definitive or probable NCC for a proportion of 46.9% (95% confidence interval [CI] 30.2–64.1) in Batondo and 45.5% (95% CI 19.0–74.1) in Pabré. No cases of NCC were identified in Nyonyogo.

Significance: All the definitive and probable cases of NCC were from the two villages where pig breeding is common. Prevention policies intended to reduce the burden of epilepsy in this country should include measures designed to interrupt the life cycle of *Taenia solium*.

KEY WORDS: Epidemiology, Epilepsy, CT scan, Neurocysticercosis, Sub-Saharan Africa.

Many studies have reported a higher prevalence of epilepsy in developing countries than in the developed world (reviewed by Roman et al., 2000; Preux & Druet-Cabanac, 2005). Infections such as malaria, meningitis, viral or bacterial encephalitis, and perinatal factors are mentioned as possible reasons for the higher prevalence (Jallon, 1997; Preux & Druet-Cabanac, 2005). One infection that has received only limited attention in some parts of the world is neurocysticercosis (NCC), although it has been reported as the most frequent parasitic infection of the central nervous system (CNS) (Garg, 1998; Roman et al., 2000).

Neurocysticercosis results from the invasion of the CNS by the larval stage of *Taenia solium* after ingestion of the parasite eggs. The adult form of the parasite is hosted by humans, causing an intestinal parasitosis (taeniasis), occurring when humans consume poorly cooked pork infected with larvae (metacestode cysticerci) of the parasite. The eggs of the parasite are shed in human feces. Pigs, the

Accepted August 6, 2012; Early View publication November 13, 2012.
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intermediate hosts, become infected when consuming human feces or food or water contaminated by human feces. Poor pig management practices, hygiene, and sanitation all contribute to the transmission of *T. solium* infection (Pal et al., 2000; Preux & Druet-Cabanac, 2005).

Humans may become accidental hosts for the larvae leading to cysticercosis when they ingest the parasite's eggs in contaminated food or water (oral–fecal contamination) (Palacios et al., 1997; Pittella, 1997; Garg, 1998; Pal et al., 2000). Cysticercosis occurs when the larvae migrate from the intestine to any tissue, but the CNS is believed to be a site of predilection. Once established in the CNS, the larva evolves through four different stages: *cystic,* where it is a viable vesicle of 10–20 mm full of liquid containing a scolex; *colloidal,* where the vesicle starts degenerating, causing a thickening of the liquid and often causing an inflammatory reaction in surrounding brain tissue; *granular,* with deposition of mineral salts; and finally *calcified,* or nonviable cysts, which appear as hyperdense areas on computerized tomography (CT) (Palacios et al., 1997; Pittella, 1997; Garg, 1998; Nash et al., 2004). The larvae may migrate into any structure of the CNS, including the spinal cord, subarachnoid space, and ventricles, but the most common site is the parenchyma. The CNS symptoms reflect both the location and the inflammation caused by the larvae, with seizures being the most common presentation (Garg, 1998; White & Garcia, 1999; Pal et al., 2000; Riley & White, 2003; Carabin et al., 2011). Other CNS symptoms include severe progressive headache, focal neurologic deficit, hydrocephalus, or symptoms of intracranial hypertension (nausea, dizziness, vomiting, or visual symptoms) (Palacios et al., 1997; Garg, 1998; White & Garcia, 1999; Pal et al., 2000; Prabhakar & Singh, 2002a; Carabin et al., 2011).

Neuroimaging is a key tool in the diagnosis of NCC (Palacios et al., 1997; Garg, 1998; White & Garcia, 1999; Pal et al., 2000). Serologic tests detecting specific antibodies or antigens of *T. solium* determine past exposure or current infection status but are not by themselves diagnostic for NCC.

A meta-analysis by Quet et al. (2010) reported an association between epilepsy and seropositivity to cysticercosis from prevalence case–control and cross-sectional studies conducted in sub-Saharan Africa. Yet, when the present study was initiated, no information was available on the prevalence of either human cysticercosis or NCC and their association with epilepsy in Burkina Faso. Such information is key to the development of effective prevention programs and policies for epilepsy in the country. The objective of this study was to estimate the prevalence of NCC-associated epilepsy in three villages of Burkina Faso and the proportion of NCC among people with epilepsy (PWE). We also describe the stage of cyst evolution among people who had CT-identified lesions of NCC.

Methods

Study sites

Three villages where epilepsy was believed to be common were selected. In two villages (Batondo and Pabré), many villagers were raising pigs. In the third village (Nyonyogo), very few pigs were raised. Batondo is located 140 km northwest of Ouagadougou, which is the capital city of Burkina Faso. The village is inhabited by approximately 3,000 people. Nyonyogo, located 30 km northeast of Ouagadougou, is inhabited by approximately 1,500 people. Pabré is located 20 km northeast of Ouagadougou and includes approximately 4,000 inhabitants.

Sampling strategy

The field investigation team included a medical doctor, a veterinarian, and two interviewers. The team was aided by a translator in Batondo, where some participants only spoke Lélé, the local language. Before the start of the study, each village was visited and each concession identified (a concession is a group of households whose members share a common ascendant and live together in proximity). A clustered random sampling strategy was used to select participants. Concessions were sampled in the first stage with all concessions included in Batondo (N = 130) and Nyonyogo (N = 131) and 336 of 609 concessions randomly selected in Pabré. The difference in the number of sampled concessions can be explained by the social structure of each village. In Pabré and Nyonyogo, each concession generally includes only one household (father, mother, and children), whereas in Batondo, related families live together in the same concession (with an average of three households per concession). In the second stage, all households in each selected concession were invited to participate. This resulted in sampling 357, 343, and 187 households in Pabré, Batondo, and Nyonyogo, respectively. The smaller number of households in Nyonyogo was due to the smaller population size in this village. In the third stage, one individual was selected randomly from each household for a screening interview on epilepsy. To be included in this study, a participant had to be at least 7 years of age and resident in the village for at least 1 year.

In all three villages, several individuals who had not been randomly selected directly contacted the field physician because they believed they had epilepsy and wanted to be examined (self-reported cases of epilepsy).

Screening interviews for epilepsy

A screening questionnaire for epilepsy, adapted from the International League Against Epilepsy (ILAE) screening questionnaire (Preux et al., 2000), was administered to the randomly selected participants of the three villages and to the self-reported cases of epilepsy. Details on this questionnaire can be found elsewhere (Nitiéma et al., 2012). Those who screened positive for seizures were evaluated for epilepsy by the field physician.

Definition and confirmation of epilepsy

The field physician conducted a full neurologic examination and asked further questions about epilepsy of participants who had screened positive on the questionnaire (Nitiéma et al., 2012). Epilepsy was defined as ever having had at least two apparently unprovoked seizures separated by at least 24 h. Only patients who met this definition were considered as confirmed epilepsy cases and were used to calculate estimates of lifetime prevalence. Active epilepsy was defined as the occurrence of at least one epileptic seizure during the previous 3 years or the use of antiepileptic drugs for seizures within the three last years. Seizures types were classified according to the ILEA guidelines based on the participant's description of seizure manifestations (Engel, 2006).

Serologic test

The results of the serologic survey in this population and the details of the diagnostic test used are reported elsewhere (Carabin et al., 2009). The enzyme-linked immunosorbent assay for the detection of circulating antigens to the metacestodes of *T. solium* (AgELISA) was used (Brandt et al., 1992; Dorny et al., 2004a).

Cranial computerized tomography

All confirmed epilepsy cases were offered a CT scan of the brain, except pregnant women. The cranial CT was performed at the Centre Hospitalier Universitaire Yalgado Ouédraogo (CHU-YO) in Ouagadougou using a General Electric 4 Barrett Model Hi Speed QX/I apparatus (General Electric, Schenectady, NY, U.S.A.). Images were taken without and with injection of contrast product (50 ml of Telebrix® 35, Guerbet, Aulnay-sous-Bois, France) 5–10 min following the injection.

Identification and classification of lesions of neurocysticercosis

The CT films were read independently by two trained radiologists (RC and M-PB-M). Films for which there were disagreements were reviewed and discussed until an agreement was reached. Lesions were classified as ''active'' cystic lesions only, ''transitional'' ring-enhancing lesions only (colloidal and granular), ''calcified'' lesions only, or ''mixed'' when there were calcifications with active or transitional lesions as suggested by Carpio et al. (1994). These NCC lesions were further classified according to the recommendations of Del Brutto et al. (2001). Briefly, lesions showing a cyst with a scolex were considered as pathognomonic for NCC and constitute an absolute criterion for diagnosis. Lesions highly suggestive of NCC included ring or nodular enhancing lesions or parenchymal calcifications and constitute major criteria (each lesion counts for one major criterion). Lesions compatible with NCC included hydrocephalus or abnormal enhancement of the leptomeninges and myelograms showing filling defects of the contrast medium and are considered minor criteria. The latter criterion could not be used, since no myelogram was performed.

Classification of a single calcification was often the source of disagreement between the radiologists. Hence, eight cases with solitary calcified cysts were reviewed by a neurosurgeon with expertise in diagnosing solitary cysticercus granuloma and calcification (VR). His diagnosis for those cases was used. The number and location of all NCC lesions were noted.

Diagnosis of neurocysticercosis

The diagnosis of NCC was made according to modified criteria of Del Brutto et al. (2001), with a serologic test positive by AgELISA replacing the Enzyme-Linked Immunoelectrotransfer Blot (EITB) test positive as a major criterion for diagnosis. This modification was made based on a recent study, which showed that serologic AgELISA and EITB tests had very similar sensitivities for detecting current infection and the AgELISA showed a better specificity than EITB (Praet et al., 2010). All the participants confirmed with epilepsy had at least one minor criterion (epilepsy) and one epidemiologic criterion (living in a *T. solium* endemic area).

With the diagnostic tools available in our study and the fact that all participants had at least one minor and one epidemiologic criterion, NCC cases were considered definitive if they had at least one absolute criterion or at least two major criteria. NCC cases were considered probable if they had one major criterion or two minor criteria.

We define NCC-associated epilepsy cases as those cases of epilepsy that met the modified diagnostic criteria of definitive or probable NCC.

Statistical analysis

The level of agreement between the two radiologists was assessed by the kappa statistic (Fleiss, 1980) and interpreted according to Landis and Koch (1977). Confidence intervals (CIs) for proportions were calculated using the ''mid-p'' binomial-based method (Newcombe, 1998). The level of significance for statistical tests was set at 0.05. All statistical analyses were done with SAS version 9.2 (Cary, NC, U.S.A.) and Open-Epi version 2.3 (Dean et al., 2010). The village-specific prevalence of NCC-associated epilepsy was estimated by multiplying the prevalence of epilepsy among those selected at random by the proportion of NCC conditional on having epilepsy from all of those who had a CT scan. This prevalence was estimated using WinBugs 1.4.3 (Imperial College, London and Medical Research Council (MRC), Cambridge, United Kingdom) and represents the proportion of people in each village estimated to have both epilepsy and NCC. A model using the proportion of NCC only among those randomly selected individuals led to very similar estimates (results not shown). The cross-sectional association between age, gender, age at first seizure, times since last seizure and since seizures onset and the presence of NCC was

estimated using the prevalence proportion ratio (PPR), where the prevalence of NCC in one group was divided by the prevalence of NCC in the reference group for each variable. PPRs were estimated between the factors of interest and definitive, probable, and all NCC cases (one estimate for each variable and category of NCC). The 95% CI of the PPR was calculated using the method proposed by Katz et al. (1978) for computing the CIs of the ratio of two proportions.

Ethics

Randomly selected individuals and self-reporting epilepsy cases, or parents of children of <15 years old, were asked for their consent to participate in the study. The consent forms were written in either French or in the local language. If the participant could not read, the form was read and clearly explained to her/him or to the legal guardians of the children. There is no written form of Lélé, so the consent was read to people in Batondo who did not speak French or other written languages. The participants were asked to sign a consent form if they agreed to participate. The signature was an ''X'' when the respondent was unable to write their name. The study was approved by the ethical committee of the Center MURAZ (Ref. 02-2006/CE-CM) and by the University of Oklahoma Health Sciences Center Institutional Review Board (IRB# 12694).

Results

Study population and prevalence of epilepsy

Of the 888 randomly selected villagers interviewed, 70 (8%) screened positive and 39 of these were confirmed with epilepsy. As reported elsewhere, this corresponded to an estimated lifetime prevalence of 4.8% (95% CI 2.9–7.6), 3.1% (95% CI 1.7–5.4), and 6.5% (95% CI 3.5–10.7) in Batondo, Pabré, and Nyonyogo, respectively (Nitiéma et al. 2012). Overall, 29 (74%), 21 (54%), and 3 (8%) reported having experienced generalized seizures, simple partial seizures, and partial seizures secondarily generalized at least once, respectively. Thirty-six of the 39 had a cranial CT. Among the three confirmed cases that did not have a CT, two (from Pabré) did not show up the day of the examination and the other (from Batondo) was a pregnant woman. Among the 36 participants with a CT scan, the time between the self-reported onset of seizures and the time of the screening questionnaire was <12 months, 1–2, 3–4, and 5 years or more for 2 (6%), 8 (22%), 3 (8%), and 21 (58%), respectively. Two participants (6%) did not remember the time of onset of their seizures.

Among the 39 subjects self-identified as having epilepsy, four were younger than 7 years of age (brought by their parents) and thus ineligible. Two of the remaining 35 were not confirmed as having epilepsy by the study doctor. Among the 33 self-reported and confirmed epilepsy cases, 33 (100%) and 11 (33%) reported having experienced generalized seizures and partial seizures at least once, respectively. None of them reported an episode of partial seizures secondarily generalized. Thirty-two of the self-identified, confirmed epilepsy cases received a CT scan; the other one did not show up on the day of the CT scan examination. Among those participants, the time interval between self-reported onset of seizures and the day of the screening interview was <12 months, 1–2, 3–4, and 5 years or more in 3 (9%), 4 (12.5%), 4 (12.5%), and 21 (66%), respectively.

Agreement of the radiologists in reading the CT scans

Of the 68 initial readings of CT scans by the two radiologists, 54 were concordant (79% agreement). The kappa was 0.55 (95% CI 0.34–0.75) when ''uncertain lesions suggestive of NCC'' were grouped with ''other lesions, not NCC'' and 0.70 (95% CI 0.52–0.88) when those cases were considered as having evidence of NCC.

Lesions identified at the CT-scan examinations

Six cases (9%) had at least one lesion pathognomonic for NCC (cyst with scolex: four in combination with calcified lesions only, and two in combination with calcified and other types of lesions). A total of 13 (19%) had no pathognomonic lesions but did have at least one lesion highly suggestive of NCC including cysts without scolex (one case in combination with a calcification), colloidal cyst only (one case), and 12 had at least one calcification (11 with calcifications only). No evidence of hydrocephaly, lesions of the leptomeninges, or brain tumors was reported. In addition to the lesions suggestive of NCC, cerebral atrophy was observed in six participants (9%), one of whom also had calcified NCC lesions.

Diagnosis of neurocysticercosis

A total of 20 patients (29%) met the criteria of definitive (11) or probable (9) NCC. Among the 11 definitive NCC cases, 6 (55%) had one absolute criterion, 1 (5%) had three major criteria (two types of lesions highly suggestive of NCC and positive AgELISA), and 4 (36%) had two major criteria (one lesion highly suggestive of NCC and a positive AgELISA). Among the nine probable NCC cases, 8 (89%) had one type of lesion highly suggestive of NCC and 1 (11%) had a positive AgELISA. Two individuals in this group were missing AgELISA results.

Table 1 presents the distribution of patients according to the results of the AgELISA, presence of lesions of NCC, and presence of absolute and major criteria for NCC by village and by type of epilepsy case (randomly selected or self-reported). A total of 10 (17%) of 60 patients with available results were seropositive to the AgELISA (major criterion). None of the participants from Nyonyogo had either an absolute or a major criterion.

Table 1 also illustrates that there were no major differences in the proportion of lesions pathognomonic or suggestive of NCC or in the proportion of cases with at least one absolute or major criterion between cases selected at random and those

Table 1. Number (%) of randomly selected and self-reported confirmed epilepsy cases with a positive AgELISA test, absolute or highly suggestive lesions of NCC, and absolute and major criteria proposed by Del Brutto et al. (2001) in three villages of Burkina Faso, 2007

	Randomly selected participants (n = 36)				Self-reported participants (n = 32)			
Village	Positive serology (%)[a]	Absolute or highly suggestive lesions of NCC (%)	Cases with one absolute criterion (%)	Cases with at least one major criteria (%)[b]	Positive serology (%)[c]	Absolute or highly suggestive lesions of NCC (%)	Cases with one absolute criterion (%)	Cases with at least one major criteria (%)[b]
Batondo	3/13 (23)	6/15 (40)	1/15 (7)	6/15 (40)	5/12 (42)	8/17 (47)	4/17 (24)	4/17 (24)
Pabré	1/9 (11)	4/9 (44)	1/9 (11)	3/9 (33)	1/2 (50)	1/2 (50)	0/2 (0)	1/2 (50)
Nyonyogo	0/11 (0)	0/12 (0)	0/12 (0)	0/12 (0)	0/13 (0)	0/13 (0)	0/13 (0)	0/13 (0)
Total	4/33 (12)	10/36 (28)	2/36 (6)	9/36 (25)	6/27 (22)	9/32 (28)	4/32 (13)	5/32 (16)

[a]Three missing (two in Batondo, one in Nyonyogo).
[b]Excluding subjects with absolute criteria.
[c]Five missing in Batondo.

who self-reported as having epilepsy. The latter group showed a higher proportion of cases with one absolute criterion, but the difference was not statistically significant.

The demographic characteristics, types of NCC lesions and of seizures, and modified Del Brutto criteria (2001) of the 20 cases with definitive or probable NCC can be found in the Supporting Information. Table S1 shows that among NCC cases with seizure onset in the past 2 years (six cases), 3–4 years (two cases) and 5 years or more (11 cases) before the interview, 4 (67%), 1 (50%), and 3 (27%) had colloidal or cystic lesions, respectively.

Prevalence of NCC-associated epilepsy and proportion of NCC among PWE

Table 2 presents the proportion NCC among people with epilepsy in each village as well as the estimated prevalence of NCC-associated epilepsy. In Batondo and Pabré, nearly half of PWE (47% with 95% CI 32–61) were definitive or probable cases of NCC. The prevalence of NCC-associated epilepsy was 2.2% (95% Bayesian Credible Interval (BCI) 1.2–3.9) in Batondo; 1.5% (95% BCI 0.1–3.0) in Pabré and 0.2% (95% BCI 0.0–1.0) in Nyonyogo.

Table 2. Number (%) of definitive or probable NCC cases among confirmed cases of epilepsy and estimated prevalence of NCC-associated epilepsy (95% BCI) in three villages of Burkina Faso, 2007

	Randomly selected cases of epilepsy (n = 36) (%)	Self-reported cases of epilepsy (n = 32) (%)	Prevalence of NCC-associated epilepsy (95% BCI)[a]
Batondo	7/15 (47)	8/17 (47)	2.2% (1.2–3.9)
Pabré	4/9 (44)	1/2 (50)	1.5% (0.1–3.0)
Nyonyogo	0/12 (0)	0/13 (0)	0.2% (0.0–1.0)

[a]Based on randomly selected participants only; subjects screened positive for epilepsy but not examined by the physician and confirmed cases of epilepsy who did not have a CT were excluded from the analysis (seven in Batondo, nine in Pabré, and one in Nyonyogo).

Cross-sectional association between selected factors and neurocysticercosis

Prevalent cases of NCC-associated epilepsy had their first seizure at an older age than non-NCC cases (Table 3). NCC-associated cases of epilepsy also tended to be older than non-NCC cases.

Discussion

This is the first community-based study of NCC in Burkina Faso. Nearly half of the PWE living in two villages where pigs were raised had NCC, whereas we found no case of NCC among PWE in a village with very few pigs.

Our results are consistent with a recent meta-analysis, which reported that 29% (95% CI 23–36) of PWE have lesions of NCC on imaging of the brain (CT scans or MRI) in endemic areas (Ndimubanzi et al., 2010). Indeed, if we include patients with lesions suggestive of NCC based on the CT scan only (not serology), the proportion of PWE with NCC lesions is 28% (19 of 68) across the three villages.

Our results show a higher prevalence of NCC among PWE than in a clinic-based study from the Mbulu district of Tanzania where NCC was reported in 18% of 212 PWE diagnosed 2–4 years before the study and who were receiving care (Winkler et al., 2009). This difference could be explained by two main reasons. First, the Tanzanian study was limited to patients who were diagnosed at least 2 years before the start of the study, which contrasts with the fact that 25% of our cases had their first seizure in the last 2 years. Moreover, the percentage of active lesions was higher among cases with more recent seizure onset. This resulted in a larger proportion of cases in our study showing active lesions at the CT scan (37% of participants with lesions of NCC), in contrast to only 16% in the Tanzanian

Table 3. Distribution of subjects with epilepsy who had a cranial CT by NCC diagnosis and selected factors with their respective prevalence proportion ratios in three villages of Burkina Faso, 2007

	NCC			Prevalence proportion ratio (95% CI)		
	Definitive (row %)	Probable (row %)	Not (row %)	Definitive versus not	Probable versus not	All NCC versus Not
Gender						
Female (reference)	2 (7)	5 (19)	20 (74)	–	–	–
Male	9 (22)	4 (10)	28 (68)	2.68 (0.63–11.28)	0.62 (0.19–2.10)	1.22 (0.56–2.67)
Age groups						
7–17 years (reference)	1 (4)	3 (14)	18 (82)	–	–	–
18–39 years	6 (18)	4 (18)	23 (70)	3.93 (0.51–30.12)	1.04 (0.26–4.14)	1.67 (0.60–4.65)
40+ years	4 (31)	2 (15)	7 (54)	6.98 (0.88–54.30)	1.56 (0.31–7.78)	2.54 (0.88–7.35)
Age at the first seizures						
0–12 years (reference)	1 (6)	2 (3)	29 (91)	–	–	–
More than 12 years	10 (29)	7 (21)	17 (50)	11.11 (1.52–81.20)	4.52 (1.03–19.83)	5.33 (1.73–16.48)
Do not know[a]	0 (0)	0 (0)	2 (100)	–	–	–
Last seizures						
Within the last 3 years (active epilepsy)	9 (15)	8 (13)	43 (72)	0.60 (0.16–2.30)	0.94 (0.14–6.30)	0.76 (0.28–2.02)
More than 3 years (reference)	2 (25)	1 (13)	5 (62)	–	–	–
Time since onset[b]						
2 years or less	4 (23)	3 (18)	10 (59)	1.80 (0.62–5.24)	1.94 (0.53–7.04)	1.68 (0.79–3.56)
More than 2 years (reference)	7 (14)	5 (10)	37 (76)	–	–	–

[a]Not included in the computation of the PPR.
[b]Time since onset was unknown for two respondents.

study. In other words, our case group represented more recent epilepsy cases, which could explain why NCC lesions were more often observed. The alternative explanation (or additional one) may be that NCC is more common in the villages selected in our study.

In a study of volunteers in Menoua Division, Cameroon, a region where pig breeding is common, Nguekam et al. (2003) found a prevalence of 59% of brain CT lesions suggestive of NCC among those with a positive serologic reaction to *T. solium* larvae antigen identified with the AgELISA. In our investigation, 9 (90%) of 10 seropositive cases had absolute or highly suggestive lesions of NCC. Furthermore, in the investigation of Nguekam et al., only 22 (65%) of 34 seropositive cases agreed to the CT scan of the brain. The true proportion of subjects with CT-scan lesions suggestive of NCC could be higher or lower if the proportion was different in the participants who declined having the brain scan. In the Eastern Cape Province, South Africa, Foyaca-Sibat et al. (2009) reported a prevalence of 37% (95% CI 27–48) of CT-scan lesions suggestive of NCC in PWE receiving medical care. This high percentage of NCC-associated epilepsy may be due to the low proportion (<0.5%) of self-reported Muslims in the 2001 census in that province (Statistics South Africa, 2004).

No cases of NCC were found among PWE living in Nyonyogo, where most people do not consume pork meat and very few pigs are raised. Hence, it is unlikely that the environment is contaminated with *T. solium* in that village. Similar results were reported by Secka et al. (2010) in the Gambia in a case–control study that included 210 PWE (cases) and 420 controls matched by gender and age (±5 years), with 95% of the study population being Muslim. In that study, all the participants had a *T. solium* serologic screening with EITB and AgELISA, and the respondents with positive results (three cases and six controls with AgELISA; none with EITB) had a cranial CT scan. No significant association between epilepsy and cysticercosis was found (odds ratio 0.75, 95% CI 0.13–3.15]). None of the nine participants who had a CT scan had brain lesions suggestive of NCC.

The manner in which pigs are typically raised (e.g., confined, tethered, or roaming) may influence the prevalence of cysticercosis in both pigs and humans (Vázquez-Flores et al., 2001; Morales et al., 2006). The proportion of NCC among PWE was very similar in Pabré and Batondo, which is not surprising, since, even though pigs were raised differently during the rainy season, they were left to roam in both villages during the dry season (Ganaba et al., 2011). Indeed, the seroprevalence of pig infection was very similar in the two villages (Ganaba et al., 2011). What is more surprising, however, is that the seroprevalence to the antigens of *T. solium* in humans was much lower in Pabré than Batondo (Carabin et al., 2009). In a study in Cameroon, Shey-Njila et al. (2003) did not find any significant difference in the prevalence of cysticercosis between permanently confined and partially confined pigs. Rather, they identified factors associated with infection in pigs as the absence of latrines in the household and the defecation of the household members in the pigpens. A similar situation is likely to exist in the present study, where only 8% of the randomly selected

participants reported using a latrine in Batondo, 11% in Nyonyogo, and 37% in Pabré. The NCC-associated epilepsy cases were older in Pabré (median age 57 years) compared to the ones in Batondo (median age 25 years), but the difference was not statistically significant ($p = 0.09$ with the Mann-Whitney test). It is possible that the seroprevalence in Pabré was lower due to the recent improvements in sanitation, with the NCC being a reflection of past infections, but the proportion of NCC patients with active lesions was similar. The sample size in Pabré was very small, which limits our ability to explore the differences further.

People with NCC and epilepsy were older than PWE without NCC. In addition, the age at onset of seizures was higher among those with NCC-associated epilepsy as compared to those without NCC. This would support the often-mentioned fact that tapeworm infection and cysticercosis are rare in children and affect mostly adults (Prabhakar & Singh, 2002b). Moreover, epilepsy with hereditary or congenital etiology is more likely to manifest early, whereas epilepsy caused by environmental factors may have a later onset.

The present study had some limitations. First, the sample may not be representative of all PWE. Self-identified cases had more generalized seizures, which are easily identifiable by the subject, his or her family, and friends. Moreover, villagers younger than 7 years of age were not included in the study. Even if NCC is rare in young children, some cases have been diagnosed in this age group (Ruiz-García et al., 1997; Salazar & Cornejo, 1997; Ferreira et al., 2001; Scott et al., 2005; Saenz et al., 2006). Other plausible reasons for error in estimating the true prevalence of NCC-associated epilepsy are that PWE may conceal their disease because of fear of stigmatization, and the questionnaire used for screening might have failed to identify some cases with more unusual types of seizure manifestations that were not recognized as epilepsy. If the distribution of NCC varies depending on the type of seizures, it could introduce a bias in our estimate of the proportion of NCC among people with epilepsy. Moreover, imperfect sensitivity and specificity of the AgELISA test used to identify cysticercosis would have influenced the final serology results. The AgELISA serologic test is useful for the diagnosis of current cysticercosis cases, with active cyst antigens circulating in the sera. The AgELISA is not designed to detect past infections (Garcia et al. 2002; Dorny et al., 2004b), and has a poor sensitivity to do so (Praet et al., 2010). This means that the AgELISA will not perform well in older NCC cases (colloidal and calcified lesions) where no circulating antigens are expected to be present. In our study, 4 (40%) of the 10 cases with calcifications and colloidal lesions only with a serologic test had a positive result, whereas 5 (71%) of 7 cases with cystic lesions (with or without scolex) were tested positive. The positive AgELISA results in participants with calcified lesions is mostly likely due to cystic lesions located elsewhere (outside the brain) given the endemic nature of the study villages. Our choice of AgELISA instead of the EITB may have led to an underestimation of the proportion of PWE cases with NCC, or in classifying fewer NCC cases as definite instead of probable. In addition, serologic results for eight epilepsy cases were missing. Finally, magnetic resonance imaging (MRI) might have been more efficient in identifying some types of NCC lesions (White & Garcia, 1999). Indeed, MRI is superior to CT for detecting NCC lesions such as cysts present in the ventricules, whereas CT is more effective for the detection of calcifications. However, no MRI device was available in Burkina Faso at the time of the study. The fact that there were few PWE also limits our ability to run a multivariate analysis of factors associated with NCC. Finally, this was a pilot study including only three villages, which may not represent the true situation in the country as a whole.

Conclusion

This study found a high prevalence of NCC in people with epilepsy and of NCC-associated epilepsy in villages where pig breeding is common. Although it is not possible in this cross-sectional study to determine whether NCC was the cause of epilepsy or simply a comorbidity in people with epilepsy living in an endemic area, these data suggest that approximately 47% of epilepsy is *potentially* preventable in two of the three study villages. The next step in research should be to determine which strategies are most effective for prevention of NCC in areas where *T. solium* is endemic.

Acknowledgments

This study was funded by the National Institute for Neurological Disorders and Stroke (NINDS) and the Fogarty International Center (FIC) of the National Institutes of Health (“Epidemiology and Burden of NCC in Burkina Faso” (R21 NS055353) under the Brain Disorders in the Developing World: Research Across the Lifespan (BRAIN) program. We would like to thank Dr. Adèle Kam from the Health Sciences Research Institute (IRSS) for her substantial help in the analyses of the blood samples. We are also thankful to Henri Somé for all his help in the database development and management as well as to the field team including Ms Alida Da, Miss Chantal Millogo, Mr Leopold Bado and Dr. Adama Sow who were instrumental for the success of this work. Finally, we are grateful to all the inhabitants of the three villages for their wonderful welcome and participation.

Disclosure

None of the authors has any conflict of interest to disclose. We confirm that we have read the Journal’s position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

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Supporting Information

Additional Supporting Information may be found in the online version of this article:

Table S1. Demographic characteristics, neurocysticercosis diagnostic criteria, and type of seizures among 20 people with epilepsy and definitive or probable NCC in two Burkina Faso villages (2006).

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