Review of Cystic Echinococcosis in Nigeria: A Story of Neglect

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Received: 3 July 2019 / Accepted: 14 September 2019
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Abstract
Purpose Cystic echinococcosis (CE) caused by Echinococcus granulosus sensu lato is a widespread zoonotic disease of global concern. In Nigeria, the exact picture/status of CE is unclear, as most of the states are largely uninvestigated. Yet, as with every parasitic zoonosis, the first step towards planning a comprehensive management and control programme involves assessment of available national/regional prevalence data, host range, and risk factors at play in the transmission dynamics.
Methods Published articles on echinococcosis were searched on PubMed and Africa Journal Online (AJOL) databases. Inclusion criteria were based on studies reporting prevalence of echinococcosis in animals and humans (including case reports) from 1970 to 2018.
Results In this study, we evaluated and summarized cystic echinococcosis reports in Nigeria and found that post 1970–80s, studies on cystic echinococcosis have remained sparse regardless of the high prevalence recorded in the early years of CE investigation. In addition, information on the genetic population structure and the role of wildlife in CE transmission is still lacking.
Conclusions This study appraises the prevalence and distribution of CE in Nigeria and identified areas where surveillance and control efforts should be focused and intensified.

Keywords Cystic echinococcosis · Prevalence · Risk factors · Nigeria

Introduction
In spite of the growing knowledge on the genetic population structure, host range and the complex taxonomic challenges, cystic echinococcosis (CE) remains a major neglected topical zoonotic disease across the world and more so in Nigeria. Caused by species of the genus Echinococcus, E. granulosus sensu stricto is responsible for the majority of CE cases, resulting in serious public health concerns [30] and losses reaching over $2 billion annually [83, 84]. CE is a chronic infection both in animals and humans and can take years for the infection to be noticed. Detection of CE infection is common during postmortem examination of animals and often incidental in humans. CE is sometimes asymptomatic due to its progressive nature except when cysts rupture to release antigenic material that causes reaction or active cysts located in certain anatomical regions (e.g., eyes and joints) exert pressure on surrounding tissues resulting in pain or discomfort (for further details on clinical manifestations, see [18, 43, 64]. Within the E. granulosus sensu lato group, mitochondrial genome studies showed considerable levels of variation [16, 17] which led to the adoption of the G1–10 nomenclature to accommodate the diversity. The grouping broadly consists of E. granulosus s.s. (G1 and G3), E. equinus (G4), E. ortleppi (G5), E. canadensis (G6–10), and E. felidis [47]. This variation has been found to be relevant for host specificity and infectivity, biochemical and morphological differences, development of diagnostic tools, drug production, vaccine development, and ultimately, management and control schemes [20, 78]. Meanwhile, there are pending controversies regarding the taxonomy of E. canadensis group and some authors have suggested categorization of genotypes G6/7 as E. intermedius, and genotypes G8 and G10 as E. canadensis [45, 47, 52].
Cystic echinococcosis is a common presentation in Africa as it is a major public health concern particularly in Northern and Eastern African countries. CE also results in huge economic losses due to the condemnation of infected animal organs slaughtered for food, and in severe cases could cause growth retardation and poor meat and milk outputs impacting negatively on the overall livestock value chain [39] with a potential annual financial loss of up to $100,000 [13, 34, 39, 65].

Nigeria is a West African country with a population of over 180 million people. It is divided into 36 states and a Federal Capital Territory (Abuja) and further grouped into six geopolitical zones (North-East, North-Central, North-West, South-East, South-South, and South-West) based on ethnicity and common history/ancestry. Owing to its favorable climate, it supports a large biodiversity and thus endemic for a number of parasitic zoonoses including echinococcosis. Historically, the earliest record of echinococcosis dates back to investigation in 1958 by the Nigerian Ministry of Health of 82 animals from the then Eastern region [31]. Thereafter, hydatid cysts were supposedly recovered from slaughtered cattle at the municipal abattoir in present day Kano state in 1961 with incomplete description of the encountered cysts as well as the number of infected animals and species responsible for the infection [32].

Between 1970 and 1990, CE investigations were frequently carried out in Nigeria (mainly in northern zones). Hitherto, epidemiological surveys have remained sparse since the 1990s, making the status of CE in Nigeria difficult to describe [5, 29]. So far, the information on CE for Nigeria has largely relied on data from 1970 to 2000 which could underestimate the disease’s current impact and importance considering likely changes that might have occurred overtime on possible sundry risk factors as well as the absence of active control programs. Therefore, to plan toward management and control, we presented a systematic review of CE studies in Nigeria from inception to date, identified research gaps, and highlighted the need to address potential bottlenecks to disease eradication.

Data Collection and Selection

Published articles on echinococcosis were searched on PubMed and Africa Journal Online (AJOL) databases in April/May 2018, and repeated in June/July 2018, using “echinococcus* OR hydatid* AND Nigeria” alone or in combination as search terms. In addition, specific searches with the name of each state were also performed. Inclusion criteria were based on the studies reporting prevalence of echinococcosis in animals and humans (including case reports) from 1970 to 2018.

Prevalence of Cystic Echinococcosis in Nigeria

Prevalence in Animals

In Nigeria, dogs are the only definitive host known to be involved in maintaining the transmission of echinococcosis. The role of wild canids is currently unknown. The first record of echinococcosis in dogs was reported in 1979 from a population of stray dogs in northern Nigeria [24, 25]. Since then, infection rates in dogs have been found to vary across the country with certain factors contributing to the observed variation (Table 1). In another study, where prevalence among dogs from various backgrounds was investigated, significantly higher infection rates were found in hunting dogs than in companion dogs. Factors contributing to this disparity reportedly include feeding and management practices by dog owners as well as age of the dogs as infection was higher in the older than younger dogs [5].

On prevalence across the country, lower prevalence was commonly reported in the North [23–25] than in the South (Table 1) such that in a study, 85% prevalence was found in a community in Niger Delta [9]. In this region, dogs are slaughtered for food [33, 40, 76]; therefore, there is high demand for dog meat and consequently increase in dog rearing activities constituting a risk to an increase in disease prevalence and distribution. Also, disparity in dog density between the North and South could provide a possible explanation to the differences in prevalence between both regions [40, 56, 76]. Nonetheless, more studies are required to understand the distribution of CE across zones.

In Nigeria, besides meeting human needs, livestock rearing is occupational especially in the North and partly in the South where pastoralists often migrate in search for greener pasture and water for their herds. Over the years, while livestock population has risen significantly with current estimates of 19 million cattle, 37 million sheep, 65 million goats, 6 million pigs, 1 million donkeys, and 277,000 camels [53], the impact of echinococcosis on livestock production has remained unevaluated. For instance, between 1960 and 1980, when cattle population was around 4–12 million [15, 36], cystic echinococcosis prevalence was estimated at 1.5–14% in cattle [24, 29]. With the current estimates of cattle population and that of CE prevalence in cattle (0.07–24.3%) [48, 58, 59, 73] coupled with lack of control measures, the impact on cattle production may have risen.

In the 1980s, CE prevalence in sheep and goats was reported to be highest in the Niger Delta region as they were 24.4% and 42.2%, respectively [10], while in Kano state, prevalence was 7.1–11.4% and 18.4–26.4%.
Table 1  Cystic echinococcosis prevalence in animals in Nigeria (1970–2018)

| Zone          | State          | Host    | Sample size | No infected | Prevalence (%) | Host status   | Detection method   | References |
|---------------|----------------|---------|-------------|-------------|----------------|---------------|-------------------|------------|
| North-Central| Plateau        | Pigs    | 170         | 0           | 0              | Slaughtered   | PM/microscopy     | [26]       |
|               | Plateau        | Sheep   | 293         | 4           | 1.4            | Slaughtered   | PM/microscopy     | [26]       |
|               | Plateau        | Goats   | 360         | 0           | 0              | Slaughtered   | PM/microscopy     | [26]       |
|               | Plateau        | Cattle  | 811         | 0           | 0              | Slaughtered   | PM/microscopy     | [26]       |
|               | Plateau        | Dogs    | 74          | 0           | 0              | Stray        | PM/microscopy     | [23]       |
| North-East    | Yobe           | Cattle  | NA          | NA          | 0.4            | Slaughtered   | PM/microscopy     | [73]       |
|               | Yobe           | Sheep   | 29,120      | 20          | 0.07           | Slaughtered   | PM/microscopy     | [74]       |
|               | Yobe           | Goats   | 87,253      | 9           | 0.01           | Slaughtered   | PM/microscopy     | [74]       |
|               | Yobe           | Camel   | 404         | 7           | 1.73           | Slaughtered   | PM/microscopy     | [42]       |
| North-West    | Kaduna         | Dogs    | 330         | 4           | 1.2            | Sub urban    | NA                | [24]       |
|               | Kaduna         | Sheep   | NA          | NA          | 7.1            | Slaughtered   | PM/microscopy     | [24]       |
|               | Kaduna         | Goats   | NA          | NA          | 18.4           | Slaughtered   | PM/microscopy     | [24]       |
|               | Kaduna         | Cattle  | NA          | NA          | 1.5            | Slaughtered   | PM/microscopy     | [24]       |
|               | Kaduna         | Camel   | NA          | NA          | 70.9           | Slaughtered   | PM/microscopy     | [24]       |
|               | Kaduna         | Pigs    | NA          | NA          | 5              | Slaughtered   | PM/microscopy     | [24]       |
|               | Kaduna         | Dogs    | 180         | 1           | 0.6            | Stray        | PM/microscopy     | [25]       |
|               | Kano           | Dogs    | 145         | 9           | 6.21           | Stray        | PM/microscopy     | [29]       |
|               | Kano           | Goats   | 1260        | 334         | 26.5           | Slaughtered   | PM/microscopy     | [29]       |
|               | Kano           | Sheep   | 1800        | 205         | 11.4           | Slaughtered   | PM/microscopy     | [29]       |
|               | Kano           | Camels  | 3580        | 1987        | 55.5           | Slaughtered   | PM/microscopy     | [29]       |
|               | Kano           | Cattle  | 4844        | 712         | 14.7           | Slaughtered   | PM/microscopy     | [29]       |
|               | Kaduna         | Camels  | 18          | 9           | 50             | Slaughtered   | PM/microscopy     | [26]       |
|               | Kaduna         | Pigs    | 147         | 0           | 0              | Slaughtered   | PM/microscopy     | [26]       |
|               | Kaduna         | Goats   | 885         | 7           | 0.79           | Slaughtered   | PM/microscopy     | [26]       |
|               | Kaduna         | Sheep   | 910         | 2           | 0.21           | Slaughtered   | PM/microscopy     | [26]       |
|               | Kano           | Goats   | 1260        | 334         | 26.5           | Slaughtered   | PM/microscopy     | [26]       |
|               | Kano           | Cattle  | 1515        | 0           | 0              | Slaughtered   | PM/microscopy     | [26]       |
|               | Kano           | Sheep   | 1800        | 206         | 11.4           | Slaughtered   | PM/microscopy     | [26]       |
|               | Kano           | Camels  | 3580        | 1987        | 55.5           | Slaughtered   | PM/microscopy     | [26]       |
|               | Kano           | Cattle  | 4844        | 713         | 14.7           | Slaughtered   | PM/microscopy     | [26]       |
|               | Kano           | Dogs    | 145         | 9           | 6.2            | Stray        | PM/microscopy     | [23]       |
|               | Kaduna         | Dogs    | 330         | 4           | 1.2            | Stray        | PM/microscopy     | [23]       |
|               | Kano           | Goats   | 130         | 31          | 23.8           | Slaughtered   | ELISA/WB          | [46]       |
|               | Kano           | Sheep   | 138         | 50          | 36.2           | Slaughtered   | ELISA/WB          | [46]       |
|               | Sokoto         | Camels  | 3545        | 318         | 8.97           | Slaughtered   | PM/microscopy     | [48]       |
|               | Sokoto         | Goats   | 14,134      | 4           | 0.03           | Slaughtered   | PM/microscopy     | [48]       |
|               | Sokoto         | Sheep   | 16,345      | 23          | 0.14           | Slaughtered   | PM/microscopy     | [48]       |
|               | Sokoto         | Cattle  | 46,223      | 34          | 0.07           | Slaughtered   | PM/microscopy     | [48]       |
|               | Sokoto         | Camels  | 200         | 84          | 42             | Slaughtered   | PM/microscopy     | [49]       |
|               | Sokoto         | Camels  | 189         | 112         | 59.3           | Slaughtered   | ELISA             | [58]       |
|               | Sokoto         | Cattle  | 285         | 69          | 24.3           | Slaughtered   | ELISA             | [58]       |
|               | Sokoto         | Cattle  | 285         | 5           | 1.8            | Slaughtered   | PM/microscopy     | [59]       |
|               | Sokoto         | Camel   | 189         | 84          | 44.4           | Slaughtered   | PM/microscopy     | [59]       |
|               | Sokoto         | Sheep   | 186         | 0           | 0              | Slaughtered   | ELISA, PM/ microsc | [67]       |
| Former Northern region | Northern region | Sheep | 458,603 | 422 | 0.09 | Slaughtered | PM/microscopy | [27] |
|               | Northern region | Goats   | 1,417,096  | 614         | 0.04           | Slaughtered   | PM/microscopy     | [27]       |
respectively [20, 36]. Recent serological reports have also confirmed high levels of antibodies against *Echinococcus* spp. in sheep and goats slaughtered in Kano (Table 1). Conversely, other states like Kaduna, Sokoto, Yobe, Borno, and Plateau experience lower prevalence (Table 1), while a number of states are yet to be investigated (Fig. 1).

Among all livestock forms, higher prevalence is frequently encountered in camels than in other hosts. In the North, between 55.5 and 70.9% have been reported in some states during the 1970–1980s [25, 29], while recent surveys within the last decade put CE prevalence between 8.97 and 59.3% in camels being higher than other concurrently examined livestock [48, 49, 58, 59]. In few northern states, infection in camels could be very low as reported in Maiduguri, Borno state where a prevalence of 1.73% (mainly fertile cysts) was found among 404 camels examined postmortem [42]. To date, this variation in infection rates among livestock across the country specifically in states where surveys have been conducted (Fig. 1, Table 1) has been reported to be largely influenced by different animal husbandry practices, conditions of slaughter slabs, and distribution or abundance of free range hosts among other factors [5, 10, 61]. Another uninvestigated factor in Nigeria probably accounting for the observed prevalence variation could be host susceptibility and/or specificity to species within *E. granulosus* sensu lato complex.

### Prevalence in Humans

In humans, information on cystic echinococcosis is rather scarce. Although where available, low prevalence is commonly reported and has been attributed to the use of less sensitive diagnostic tools [28] or the fact that CE is not considered during routine medical examination. Till now, human CE has been investigated in few states and are mostly retrospective studies involving assessment of hospital records [28, 61] (Table 2). One of such earliest surveys conducted in three northern states (Plateau, Kano, and Kaduna) identified only one case out of 620,695 examined records [28]. Nonetheless, *Echinococcus* cyst develops progressively and detection in humans is most times incidental with infection being rather asymptomatic except during unusual presentation involving certain organs, in which case the resultant discomfort may warrant diagnosis. Despite that, misdiagnosis and consequently mistreatment are challenges often faced as a result of poor knowledge of the nature of the disease [35]. While human cases of CE remain a rarity, another report was a case of an 18-year-old female from southeastern

| Zone          | State          | Host       | Sample size | No infected | Prevalence (%) | Host status | Detection method | References |
|---------------|----------------|------------|-------------|-------------|----------------|-------------|------------------|------------|
| Northern region | Pigs          | 23,830     | 39          | 0.16        | Slaughtered    | PM/microscopy | [27]             |
| Northern region | Cattle        | 1,815,792  | 1767        | 0.1         | Slaughtered    | PM/microscopy | [27]             |
| Northern region | Camel         | 49,220     | 659         | 1.34        | Slaughtered    | PM/microscopy | [27]             |
| Anambra       | Dogs           | 182        | 8           | 4.4         | Rural dogs     | PM/microscopy | [57]             |
| Anambra       | Cattle         | 551        | 0           | 0           | Slaughtered    | PM/microscopy | [61]             |
| Anambra       | Pigs           | 2126       | 0           | 0           | Slaughtered    | PM/microscopy | [61]             |
| Anambra       | Goats          | 3830       | 0           | 0           | Slaughtered    | PM/microscopy | [61]             |
| Anambra       | Pigs           | 31,005     | 1           | 0.003       | Slaughtered    | PM/microscopy | [61]             |
| Anambra       | Camel          | 373,242    | 7           | 0.002       | Slaughtered    | PM/microscopy | [61]             |
| Anambra       | Goats          | 476,249    | 249         | 0.05        | Slaughtered    | PM/microscopy | [61]             |
| Rivers        | Dogs           | 60         | 51          | 85          | Urban dogs     | Microscopy    | [9]              |
| Rivers        | Cattle         | 320        | 101         | 31.6        | Slaughtered    | PM/microscopy | [10]             |
| Rivers        | Goats          | 320        | 135         | 42.2        | Slaughtered    | PM/microscopy | [10]             |
| Rivers        | Pigs           | 320        | 179         | 55.9        | Slaughtered    | PM/microscopy | [10]             |
| Rivers        | Sheep          | 320        | 78          | 24.4        | Slaughtered    | PM/microscopy | [10]             |
| Cross River   | Dogs           | 254        | 2           | 0.78        | Urban dogs     | Microscopy    | [77]             |
| Oyo           | Dogs           | 155        | 15          | 9.68        | Hunting/Companion | ELISA   | [4]              |
| South-West    | Dogs           | 273        | 34          | 12.45       | Hunting/Companion | ELISA   | [5]              |
| Oyo           | Dogs           | 102        | 49          | 48          | Owned dogs     | Microscopy    | [8]              |
| Oyo           | Sheep/goats    | 215        | 60          | 28          | Slaughtered    | PM/microscopy | [8]              |

 PM postmortem examination, WB Western Blot, ELISA enzyme-linked immunosorbent assay
Nigeria diagnosed of a fertile unilocular pulmonary cyst, presumed to be *E. granulosus* [6, 7]. This report prompted subsequent examination of hospital records of the University of Nigeria Teaching Hospital, in Enugu State (which also served Anambra State at the time) with preference to surgery cases from January 1977 to December 1986; however,
findings from this investigation revealed no documented case. On further examination of rural health centers and some rural- and urban-located hospitals, it was observed that medical personnel in these institutions lacked requisite knowledge of the disease which might have resulted in misdiagnoses [61]. However, in recent times, two cases have been reported and include a musculoskeletal involvement with HIV coinfection in a patient admitted to The University of Jos Teaching Hospital [63], and an orbital echinococcosis manifested in protrusion of the eye and poor vision in The University College Hospital, Ibadan [35]. In the latter case, the patient had an existing record of being managed for 22 years in the teaching hospital with clinical history of painless protrusion of an eye and was treated for non-specific orbital inflammatory disease until further histopathological examination confirmed the presence of a cyst with a laminated and a germinal layer.

Therefore, it is plausible that most human cases of CE presented in a number of hospitals could similarly be misdiagnosed and consequently mistreated. This problem of misdiagnosis remains a serious challenge in identifying CE cases in Nigeria. So far, the only population-based survey was carried out using 176 blood sera from hospital patients in Minna and Abeokuta in Niger and Ogun states, respectively, and mean result showed a 0.53% CE seroprevalence [70]. Although human CE in Nigeria is usually rare, a similar situation exists in many eastern and northern African countries where human prevalence usually ranged from 0 to 2% and is yet considered a disease of major public health concern [12, 44, 50, 68]. Regardless, the need for differential diagnosis of suspected cases remains invaluable in appraising the level of human CE infection across zones in Nigeria.

**Risk Factors**

In endemic regions across the world, infection is associated with resource poor settings, pastoral lifestyle, certain pasture types as well as socio-economic and behavioral practices [19, 55, 79, 80]. Other factors include sources of drinking water, changes in environmental conditions favouring egg survival, transboundary animal movement and livestock trade, high proportion/presence of stray dogs, frequency of dog-man contacts, poor abattoir conditions and poor disposal of waste from slaughterhouses [2, 11, 19, 55, 60, 62]. In Nigeria, besides these factors [5, 35], others such as age of livestock have been labeled to be largely responsible for higher infection rate in camels than in other livestock. For example, camels are often slaughtered after 8–10 years of age, leading to increased exposure and subsequent risk of acquiring the infection [59]. Recently, differences in ambient temperatures across the country were suggested as a possible factor that could cause prevalence to vary as some states experience temperatures as high as 45 °C resulting in desiccation of eggs [42]. Furthermore, since no study has been dedicated to identifying local factors responsible for human and livestock CE in Nigeria, the need for such studies to identify local transmission patterns cannot be overstated.

**Bottleneck of Cystic Echinococcosis Research in Nigeria**

Over the years, global echinococcosis research has recorded huge progress, including the discovery of new species and expanding knowledge on genotypes and haplotype variation, complete genome sequences, development of sensitive detection techniques, and the EG95 vaccine [1, 37, 51, 54, 75, 81, 85]. In spite of these feats, a lot of questions remain unanswered on the status of CE in Nigeria. For example, of the available studies on echinococcosis, over 60% were conducted between 1970 and 1990, and less than 10% in the last decade. Another challenge is in the choice of diagnosis/detection tools used, such as microscopy, postmortem examination, immunodiagnosis, and molecular techniques. While the challenge of microscopy is in its inappropriateness for investigating intermediate hosts, age of livestock, early infection, and variation in cyst development in intermediate hosts [38, 41, 71] may fraught the effort of postmortem approach (the gold standard). Only recently, surveys have sparingly employed ELISA [5, 46, 58, 67], sonography, and histopathology techniques [35, 63]. Following WHO publication of Standardized classification of ultrasound images of CE, the use of sonography in combination with differential diagnosis has been advocated as a useful tool in detecting different presentation of CE infection in humans especially among population at risk [82]. With serology, the inability of available immunodiagnostic tools (ELISA) to discriminate between strains/genotypes or the problem of specificity/sensitivity or cross reactivity remains a major reason why serology is still debatable [22, 66, 72]. Although some antigens and recombinant proteins have shown potential for diagnosis, standardization and understanding of their performance level is greatly needed (see [69] for details on diagnosis of CE).

On the other hand, PCR-based techniques are regarded to be highly useful for genotyping [17, 21], however, have not been applied in Nigeria till date; hence, the consequent lack of information regarding the genotypes and genetic population structure unlike in northern and eastern African countries where genotypes and their genetic variation have been largely investigated [3, 14]. Though no molecular data exists on the circulating E. granulosus genotypes, it will be apt to find the common E. granulosus s.s., and E. canaden sis (G6) considering the involvement of intermediate hosts like sheep, goats, cattle, and camel coupled with the high
species/haplotype diversity reported in the sub-Saharan African region [30]. Further, the interface between wildlife and domestic CE transmission remains largely uninvestigated.

Conclusion

The available data clearly emphasize the public health implication of CE, especially in regions where cases (both human and animals) have been reported and factors enhancing transmission are present. With the lack of information on two-third of the states and absence of data on the circulating genotypes, it is pertinent to state that CE in Nigeria is highly under-investigated and thus neglected regardless of its high zoonotic potential and the public health outcry raised in the early years of investigation. Thus, it is important to evolve and embark on a comprehensive animal and human survey across the country to make data available especially on the genetic population structure, local risk factors enhancing transmission, and the role of wildlife in CE transmission as a critical step to evaluating areas where control efforts should be prioritized. In addition, awareness of the nature of the disease and trainings on the diagnoses should be provided for medical personnel in hospitals to differentially diagnose and identify CE on presentation. Finally, we recommend participation in initiatives like the German–African consortium on cystic echinococcosis: Cystic Echinococcosis in sub-Saharan Africa Research Initiative, CESSARI (https://gdri-ehede.univ-fcomte.fr/spip.php?article23&lang=en), a collaborative platform and capacity building forum aimed at effectively and efficiently investigating CE epidemiology, economic impact and genetic diversity toward disease eradication.

Acknowledgements

This study was supported by Central Public-interest Scientific Institution Basal Research Fund (1610312017001; 1610312016012), National Key Basic Research Program (973 Program) of China (2015CB150300), National Key Research and Development Plan (2017YFD0501301), and NBCITS, MOA (CARS-38), while John Asekhaen Ohiolei’s Ph.D. program was funded by the Chinese Government Scholarship Programme of the Peoples Republic of China.

Compliance with ethical standards

Conflict of interest The authors declare that they have no competing interests.

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