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Systematic Review

The Prevalence and Determinants of *Taenia multiceps* Infection (Cerebral Coenurosis) in Small Ruminants in Africa: A Systematic Review

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Abstract: Cerebral coenurosis is a fatal parasitic neurological disease affecting the brain and spinal cord of goats and sheep. Despite the large population of sheep and goats in Africa, there is very little awareness about the scale or effects of the disease. A systematic review was conducted to bridge the gap in the current understanding of the epidemiology of *Taenia multiceps* coenurosis in Africa. A detailed literature search was conducted in EBSCOhost, Google Scholar, Research4life (AGORA), and PubMed for studies in *T. multiceps* coenurosis in goats and sheep in Africa for the period spanning January 2000 to December 2019. The search identified 574 publications, of which only 11 studies were eligible, highlighting major gaps in reporting of this disease in Africa. Data were extracted and evidence synthesized in relation to prevalence and determinants for *T. multiceps* coenurosis. The descriptive summary statistics revealed a median (IQR) prevalence of coenurosis of 22.3% (range: 18.3–26.4 percent) for community based and 14.8% (range: 3.8–45.6) for abattoir-based studies. This high prevalence indicates that *Taenia multiceps* coenurosis is an important disease for small ruminant productivity in Africa. Knowledge of the determinants of prevalence could be used to inform preventive measures, including behaviour change among livestock keepers and those involved in livestock slaughter to break cycles of transmission between small ruminants and dogs.

Keywords: Africa; *Taenia multiceps*; sheep; goats; prevalence; cerebral coenurosis

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1. Introduction

The *Taenia multiceps* coenurosis, also known as cerebral coenurosis, is a fatal parasitic neurological disease affecting the brain and spinal cord of goats and sheep. The disease is associated with a range of neurological syndromes due to the effect the disease has on the central nervous system. Cerebral coenurosis is caused by infection with larval stages (metacestodes) of the cestode parasite *T. multiceps.* Small ruminants (goats and sheep) are the common intermediate hosts, but wild herbivores may also be affected [1]. Definitive hosts are domestic dogs (*Canis lupus familiaris*) and wild canids such as wild dogs (*Lycaon pictus*), foxes (*Vulpes* spp.) and jackals (*Canis* spp.) [2]. While the parasite has little clinical effect on the definitive hosts, the intermediate stage *Coenurus cerebralis* (metacestodes) in small ruminants causes serious neurological disease. In small ruminants, cerebral coenurosis is characterized by dullness, circling, torticollis, loss of appetite, frequent vocalization, and separation from the flock, visual impairment, and muscle tremors. Other notable signs are pain response on pressure over the cystic area, and sometimes unilateral partial blindness, lateral tilting of the head either towards the right or left, head pressing, and feet stamping [3,4].

Small ruminants become infected when they ingest eggs from pastures and water contaminated with the feces of infected canids. After ingestion, in the small intestine the thick striated outer layer is digested and the egg hatches to release an oncosphere (hexancath embryo). The oncosphere then penetrates the intestinal wall and enters the bloodstream, travelling to the brain and spinal cord where *Coenurus cerebralis* cysts develop leading to cerebral coenurosis [2,5,6]. This metacestode larvae has a high affinity to cerebrospinal fluid (CSF) for their differentiation and development [7]. The incubation period varies from 15 to 33 days for acute onset and about six to eight months for the chronic form of the disease. Transmission to definitive hosts occurs when dogs or other canids ingest cysts in undercooked/raw infected brain from an intermediate host. The protoscoleces attach to the small intestinal wall and in about 21 days the worms begin to form proglottids. Each one of the segments (proglottid) of the *T. multiceps* contains both the male and female reproductive organs [8]. Each terminal proglottid of mature *T. multiceps* can contain more than 30,000 eggs [3–5]. Proglottids containing eggs detach from the end of the worm and pass out in the feces onto pasture where they can be ingested by goats and sheep [9–11].

Taenia multiceps has a worldwide distribution [2,4,6,12–17]. Coenurosis and *T. multiceps* have been well studied in some parts of the world, including epidemiological studies in Asia and Europe [1,4,5,8,9,18]. While the disease has been somewhat controlled in many industrialized countries, it continues to cause a burden of production losses in many extensive farming systems. Although the disease has been reported in a number of African countries including Ethiopia, Kenya, Egypt [6,16,19] and most recently in Tanzania [20–22], awareness of the magnitude and impact of the disease across Africa remains low.

Goats and sheep are important food animals in most rural settings in Africa and they contribute to the livelihoods of many in pastoral areas [23], with evidence that they are becoming increasingly important in some of the most precarious livestock-dependent communities [24]. This is likely to be partly a response to climate change, with small ruminants showing good adaptability to different climates [25]. In addition, goats and sheep are popular because they need little resource, low initial capital, and have shorter reproductive intervals than cattle. However, the productivity of small ruminants in Africa is limited by several factors, such as poor husbandry and diseases [25].

There is growing evidence that cerebral coenurosis is emerging as a major disease of concern for some small ruminant keepers in Africa [6,16,19]. In northern Tanzania, a recent survey found that neurological disease was reported as a leading animal health concern for pastoral livestock keepers [20,21,26]. A pilot study conducted in this area found that over 80% of neurological syndrome cases in small ruminants were due to *T. multiceps* coenurosis [26]. In Tanzania, the disease was mainly reported in pastoral areas [20,21,26] but little is known of prevalence and distribution of risk factors and determinants of prevalence of the disease in other small ruminant production systems. Overall, there remains a gap in awareness of the current epidemiology of cerebral coenurosis caused by *T. multiceps* in small ruminants, not just in Tanzania but also across the African continent.

To bridge this gap, we performed a systematic review on published, peer-reviewed literature. The search aimed to summarize and compare the epidemiological knowledge on the prevalence, geographic distribution and determinants for *T. multiceps* coenurosis prevalence in goats and sheep in African countries.

2. Results

2.1. Search Results

A total of 574 records were identified through database searches. Of these, 235 were duplicate records and were removed. Therefore, a total of 339 articles were considered eligible for the title and abstract screening. A further 307 were excluded after title and abstract review, and 21 were further excluded after full text review for the following reasons: full text not accessible (n = 2), full text duplicate (n = 5) and no primary data/summary statistics (n = 14); the list of the excluded full texts is provided in supplementary materials (Table S2). Finally, full text articles (n = 11) were eligible for inclusion in the study (Figure 1).

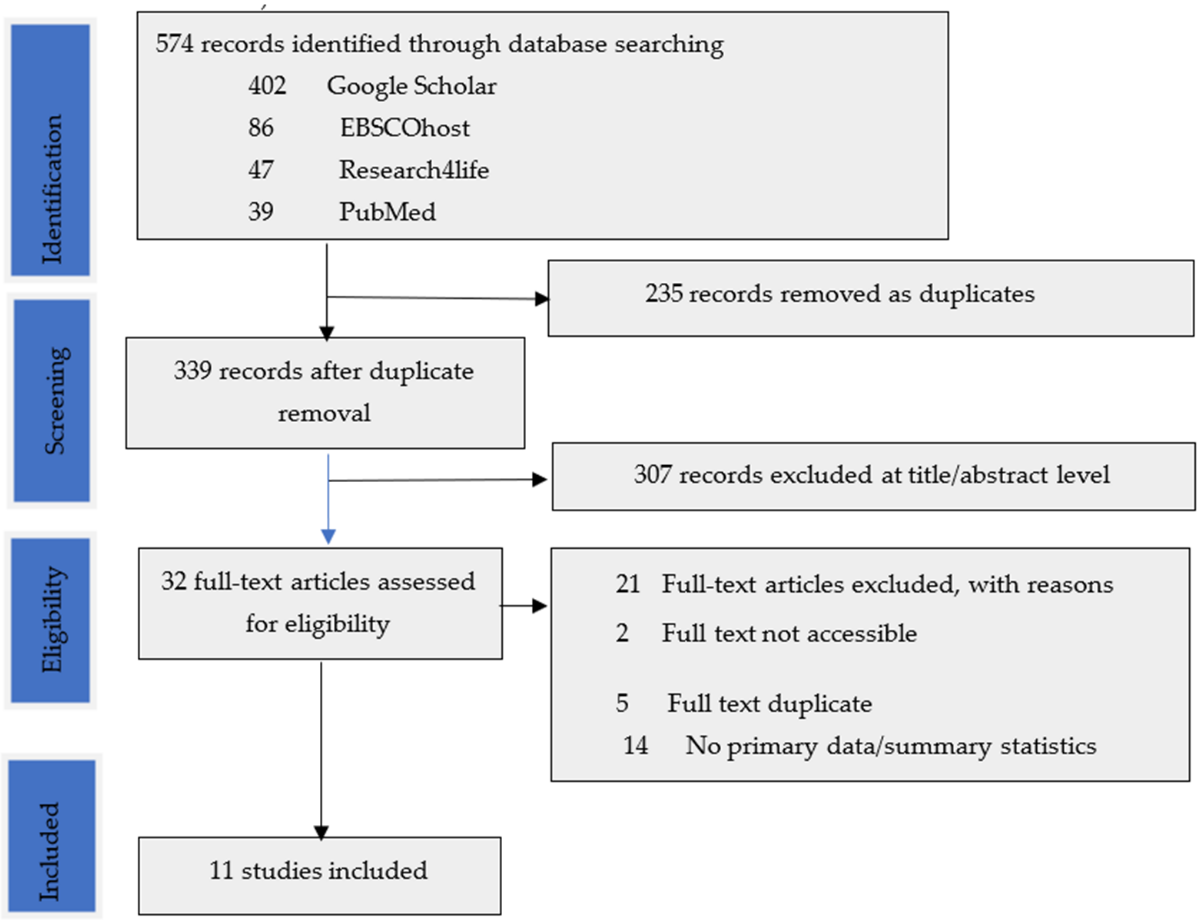

Figure 1. PRISMA flow chart showing selection of eligible articles for inclusion in the review 2000–2019.

2.2. Descriptive Summary of the Studies Included

Of the 11 studies included in this review, five (45.5%) articles were from Egypt, three (27.2%) from Ethiopia, two (18.2%) from Tanzania and one (9.1%) from Mozambique (the only country reporting coenurosis in small ruminants in southern Africa). Data on the location of each study by country, study type, settings under which the studies were carried out, prevalence are summarized in Table 1. Out of the eleven studies, four (36.4%) were community based cross-sectional studies and the remaining seven (63.6%) were abattoir-based cross-sectional studies.

Table 1. Prevalence of *T. multiceps* infection (cerebral coenurosis) for sheep and goats in Africa, 2000–2019.

Country	Study Settings	Diagnostic Test	Sheep *n*/N (%)	Goats *n*/N (%)	Comments	Citation
Ethiopia	Abattoir	PM	21/445 (4.7%)	-	N/A	[22]
Ethiopia	Abattoir	PM	23/111 (20.7%)	29/301 (9.6%)	N/A	[27]
Ethiopia	Abattoir	PM	19/384 (4.9%)	45/384 (11.7%)	N/A	[28]
Egypt	Abattoir	PM, PCR	3/80 (3.7%)	-	N/A	[29]
Mozambique	Abattoir	PM	-	12/142 (8.4%)	N/A	[30]
Egypt	Abattoir	PM	15/45 (33.3%)	10/30 (33.3%)	N/A	[19]
Tanzania	Abattoir	PM	41/90 (45.6%)	39/90 (43.3%)	N/A	[20]
Egypt *	Community	PM	26/747 (3.5%)	-	Of the neurological cases examined, 26/43 sheep were confirmed as *T. multiceps*	[31]
Egypt	Community	PM, PCR	111/420 (26.4%)	-	N/A	[32]
Tanzania *	Community	PM, PCR	1407/5962 (23.6%)	1218/4722 (25.8%)	Of the neurological cases examined, 16/20 sheep and 16/19 goats were confirmed as *T. multiceps*	[26]
Egypt	Community	PM	11/60 (18.3%)	-	N/A	[33]

* These studies reported prevalence of neurological disease and the porportion of neurological cases that were confirmed at *T. multiceps*. These figures therefore represent an apparent prevalence rather than the true prevalence and are excluded from the summary statistics, N/A = No commentsPCR = Polymerase chain reaction, PM = post-mortem conducted out of the survey for reported cases with neurologic syndromes, *n*/N = proportion of positive individuals.

2.3. Prevalence of *T. multiceps*

Reported prevalence ranged from 3.5% in sheep in an abattoir study in Egypt [31] to 45.6% in sheep in an abattoir study in Tanzania [20] (Table 1). The descriptive summary statistics revealed a median (IQR) prevalence of coenurosis of 11.7% (range: 8.5 to 43.3 percent) in goats and a median prevalence of 19.5% (range: 3.8 to 45.6 percent) in sheep. Furthermore, a prevalence of coenurosis of 22.3% (range: 18.3–26.4 percent) was revealed or community based and 14.8% (range: 3.8–45.6) for abattoir-based studies (Figure 2).

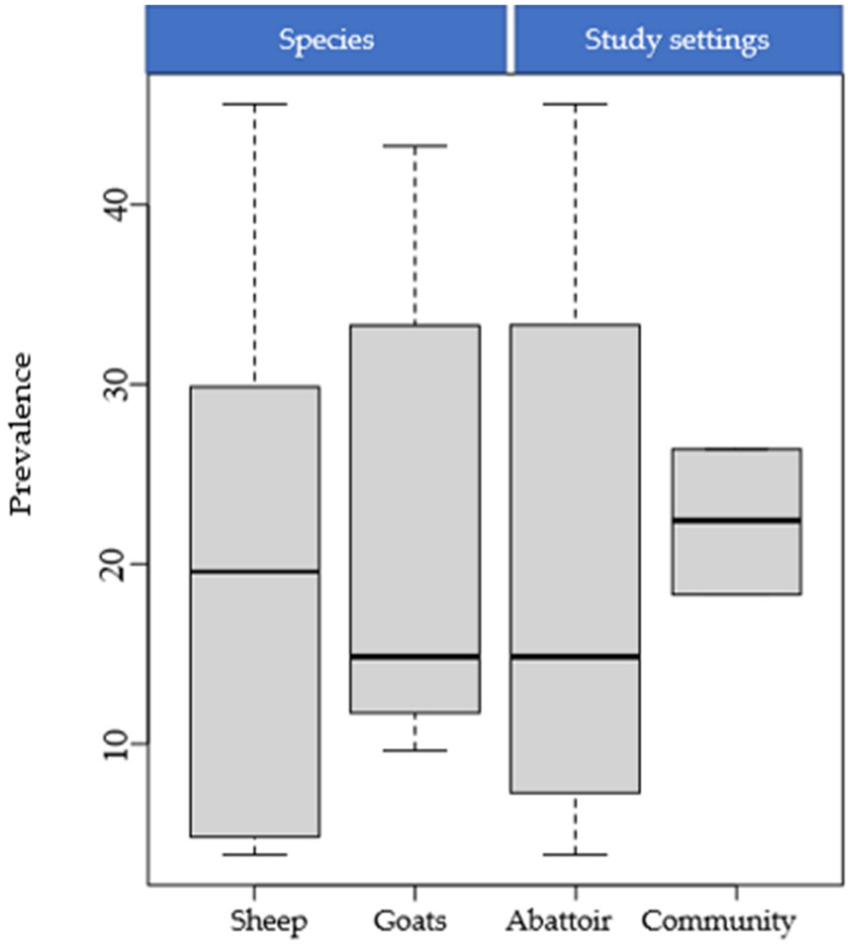

Figure 2. Median prevalence by species and study settings for coenurosis in sheep and goats in Africa from 2000 to 2019.

2.4. Determinants of Prevalence of T. multiceps

Six (54.5%) out of eleven studies mentioned determinant factors for coenurosis prevalence in small ruminants. Of the six studies, only three involved these determinants in their study design and analysis, and of these only two studies; each study from Tanzania [26] and Egypt [31] respectively involved advanced statistical analysis (regression analysis) as shown in Table S5. The stated determinants were grouped according to the strength of the evidence, and the frequency percentage of each determinant factor was computed. The outcome of the summary statistics revealed that six (23%) determinants were very strong (Figure S1). These strongly supported determinants were: feeding brains to dogs, dog ownership, livestock production systems, the presence of wild host canids, not deworming dogs and poor dog feces management.

3. Discussion

This review aimed to synthesize epidemiological evidence on prevalence and risk factors for *Taenia multiceps* coenurosis infection in sheep and goats in Africa from the peer reviewed literature from the year 2000 to 2019. Our search found only 11 eligible articles but despite this small number, evidence of coenurosis was identified from countries in northern, eastern and southern Africa, indicating a wide geographic distribution of the disease; however, the low number of publications found reveals a gap in the reporting of this disease. This further highlights a lack of attention to coenurosis in Africa, despite losses and burden of the disease reported in several countries.

This review found no marked difference in prevalence across species. Sheep and goats are fed and kept together, so this result is not surprising. The median prevalence of coenurosis was higher in community studies compared to abattoir studies, but the maximum prevalence recorded (45.6%) was from an abattoir survey. This this likely reflects the fact that small ruminants suffering from coenurosis are are both sold for meat as well as culled for home consumption. Home slaughter is a practice of concern as this is often linked with deliberate feeding of brains of affected sheep and goats to dogs, which was identified as a determinant of prevalence in of the recent study in east Africa [26]. The habit of "throwing brains to dogs" is reported as a common practice whereby any part of the carcass not used for human consumption is given to dogs [20,26,31]. Similarly, traditional backyard slaughter is likely to be practiced by livestock keeping communities during circumcision, traditional weddings, and birth ceremonies to venerate ancestors and funerals [34]. Such slaughter practices could leadto increased risk of *T. multiceps* coenurosis transmission due to unsuperviseddisposal of offal and other rejected parts of the carcass. Even for everyday slaughtering, the shortage of human capacity to carry out extension services and meat inspection in rural settings is limited [35]. Inadequate meat inspection services are characterized by poor or lack of slaughter infrastructure could lead to the unsupervised disposal of brains and offal, rendering them accessible to dogs and exaggerating taeniid infections. In addition, poor feeding of dogs results in scavenging behavior which further increases the risk of infection in dogs [36].

Taenia multiceps infection of dogs can be treated with anthelmintic drugs such as Praziquantel. Indeed, limited deworming practices have been mentioned in studies involved in this review [6,15,21,22,26]. *T. multiceps* has been controlled in some developed countries through the regular deworming of dogs, and through keeping dogs and wild canids away from farms [37]. However, in Africa, scaling up a sustainable deworming program is likely to be hindered by financial constraints in resource-poor communities. Embedding cestode deworming programs in other dog disease control programs, such as rabies vaccination programs, would offer an exciting opportunity. Such programs could potentially target a large dog population and pave the way for the sensitization of issues related to the welfare of dogs in aspects of health. Furthermore, deworming programs could go hand in hand with behavior change for risk habits which enable the *T. multiceps* life cycle. These should include avoiding the unsupervised slaughter of small ruminants, proper disposal of offal and not feeding brains to dogs. In addition to the husbandry means of controlling *T. multiceps*, there are promising developments for a vaccine against *T. multiceps* in goats and sheep [38,39]; however, to date there are no commercial vaccines available. Outstanding areas for further study include extensive field trials to determine the effectiveness, immunogenicity and safety of vaccine candidates that have shown promising results in early development studies. The availability of an effective vaccine in Africa could dramatically impact the prevalence of *T. multiceps* in goats and sheep in countries where the disease is known to be endemic.

The presence of wild canid hosts has been suggested as a risk factor was for coenurosis in small ruminants in one study in Egpt [33] but further evidence is required to investigate the role of wild carnivores in the epidemiology of coenurosis in different settings. Wildlife-livestock interaction inevitably poses animal disease risks in rangelands around conservation areas. For example, in east Africa, mixed wildlife-livestock rangelands comprise an important part of protected area ecosystems, and the diversity and abundance of wildlife grazing in proximity to livestock and dogs provides many potential opportunities for infection transmission between wild canids and ungulate herbivores. Similarly, outside Africa, a study by Varcasia et al., 2019 in Italy isolated *T. multiceps* in red fox (*Vulpes vulpes*) [14]. In addition, *T. multiceps* cysts were isolated in wild ungulates in the French Alps [40]; Ibex Nubian wild in Israel [41], and wild goats (*Capra aegagrus*) in Iran [42]. This implies that the sylvatic circulation of the parasite *T. multiceps* remains possible however, the role and contribution of wildlife to the transmission and maintenance of *Taenia multiceps* coenurosis remains unclear.

Limitation of the Study

Findings on the prevalence of *T. multiceps* from most abattoir studies should be cautiously interpreted due to the possible bias, since sick animals might be positively selected to be culled due to health problems [19,20,22,27–30]. Although this type of bias might exaggerate the prevalence estimates, these studies remain an important indicator of the magnitude of the problem in small ruminant-keeping communities. Community based studies with random selection of farms provide a more robust estimate, but these remain very rare, with only studies identified from Egypt. The prevalence of suspected coenurosis cases has also been used to demonstrate the likely importance of *T. multiceps* [26,31–33], but these studies are also limited as cases were identified on the basis of neurological syndromes with only a small proportion of cases confirmed by postmortem. The small number of eligible articles identified by this review is also an important limitation. Furthermore, for the majority of studies, the strength of evidence for determinants for *T. multiceps* coenurosis was weak. Most determinants were only mentioned and not included in the study design and analysis. Therefore, possible conclusions on the relationship between *T. multiceps* infection prevalence and these contributing factors remain unsubstantiated with epidemiological evidence. In addition, our literature search did not retrieve any literature on *T. multiceps* coenurosis in small ruminants from western Africa, and it remains unclear whether this is due to a lack of infection or a lack of detection and reporting.

4. Materials and Methods

4.1. Study Design and Review Protocol

Electronic search involved the following databases, EBSCOhost, Google Scholar, Research4life (AGORA), and PubMed. Key strings used during the search were: "Coenurosis" or "Cenurosis" or "Cerebral coenurosis" or "*Taenia multiceps*" or "Coenurus cerebralis" or "Ormilo and Africa" (Table S1). In this search, Africa referred to any country within the United Nations definition of Africa (United Nations (UN) Statistics Division, 2020). The term "Ormilo" is a local term in the Maasai language referring to circling or neurologic syndrome, commonly used by pastoral communities from Kenya and Tanzania. The study protocol and management of this review was performed through CADIMA, an open-access online tool for supporting the conduct of a systematic review [43]. A detailed protocol for this review can be found in CADIMA. The review process ensured compliance to Preferred Reporting Items for Systematic reviews and Meta-analyses, PRISMA (checklist Table S3) [44].

4.2. Search Strategy and Study Selection

A detailed literature search was conducted through structured databases. The review process was divided into two steps: (a) abstract and title review (b) full text review. Article titles and abstracts were initially reviewed by TK for suitability of inclusion. Articles were included for full text review based on the following criteria: (i) reported primary data; (ii) reported risk factors and/or prevalence of *T. multiceps* coenurosis; and (iii) if species involved were goats and/or sheep. Full text articles were excluded if (i) the full article could not be retrieved; (ii) the article lacked data on prevalence and risk factors for *T. multiceps*; (iii) the article had no primary information on *T. multiceps*; or (iv) articles were excluded that did not include data from Africa. Articles eligible for inclusion were retrieved in full format and assessed based on set criteria. Only abstracts and articles in the English language were included in the review process. This review process was performed by two independent reviewers, T.K. and K.N.

4.3. Data Extraction and Synthesis

Data extraction was performed on all articles eligible for inclusion in the study. Two independent team members (T.K. and K.N.) extracted pre-determined quantitative and qualitative data including the author and date (year) of the study, country of origin of the study, study settings (Abattoir or community), diagnostic tests used, species (goat and/or sheep), sample size, prevalence, or any risk factors or determinants of the disease mentioned in relation to the epidemiology of *T. multiceps* coenurosis.

4.4. Data Analysis Methods

Data extracted were collated using Microsoft Excel® (Microsoft Corporation, Washington, DC, USA) with the prevalence in each individual study recorded. The number of goats and or sheep included in the study and number of positive individuals were also extracted (Table 1). In addition, the prevalence was descriptively summarised and reported as medians and interquartile ranges (IQR) based on study type and species.

Similarly, determinants of prevalence mentioned and or investigated in each study were extracted and listed and summarised qualitatively. Descriptively, the quality and strength of evidence of determinants of coenurosis prevalence were examined focusing on methodology (study design) and data analysis. Furthermore, strength of evidence for each determinant of prevalence, as reported by the studies, was assessed and categorized into three categories: strong evidence, medium evidence and weak evidence. The evidence was considered strong when the determinants of the prevalence were part of the study design, the analysis employed advanced statistics such as regression models, and the results were significant. In addition, the evidence was considered of medium quality when the determinants of the prevalence were part of the study design, and the analysis only employed basic descriptive statistics such means and proportions. The evidence was considered weak when the determinants of the prevalence were only mentioned, and none were incorporated in the study design and analysis (Table S4). The frequency of each determinant based on its strength was summarized using the dplyr package [45] in the R statistical environment version 4.0.5, R Core Team (2021). The total frequency yielded the percentage of each determinant mentioned in studies included in this review.

5. Conclusions

Overall, the results from this review indicate that drivers of the endemicity of cerebral coenurosis in small ruminants is likely to be strongly influenced by behavioral practices by farmers and poor standards of dog management. The evidence generated from this review highlights the magnitude of the *T. multiceps* infection problem in Africa. The high prevalence reported by studies included in this review suggest that cerebral coenurosis is endemic in several countries in northern and eastern Africa. Future studies need to emphasize the importance of effective study design so that the relationship between prevalence and contributing factors can be determined. Furthermore, to help reduce production losses associated with this infection, it is important to understand how population patterns and the management of domestic dogs and the presence of wild canid hosts affect the prevalence of *T. multiceps*.

Supplementary Materials: The following supporting information can be downloaded at: https://www.mdpi.com/article/10.3390/parasitologia2020013/s1, Figure S1: Strength of evidence of determinants of prevalence of *T. multiceps* coenurosis in goats and sheep in Africa year 2000 to 2019; Table S1: Full search strategies for databases searches; Table S2: Full-text articles excluded, with reasons; Table S3: PRISMA 2009 Checklist; Table S4: Table of criteria for quality and strength of evidence for determinants of prevalence of coenurosis as described by Alonso et al., 2016 with modification; Table S5: List of identified determinants for *T. multiceps* coenurosis prevalence in small ruminants in Africa from years 2000–2019 as described from previous individual literature based on criteria mentioned in Table S4.

Author Contributions: All authors contributed the concept or study design: T.K., organized literature search and populated the database. T.K. wrote the first draft and subsequent revisions of the manuscript. J.B., G.S., F.L., K.N., E.H., S.C. and K.J.A. made substantial contribution to the manuscript. All authors contributed to manuscript improvement, read and approved the submitted manuscript. All authors have read and agreed to the published version of the manuscript.

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