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Thyroid involvement in cystic echinococcosis: a systematic review

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Abstract

Background Thyroid Hydatid Cyst (THC), a pathological state induced by the larval form of *Echinococcus granulosus*, represents a multifaceted clinical entity with nonspecific symptoms, making both diagnosis and treatment intricate. The current understanding of THC's attributes is somewhat limited. To gain a broader perspective on the disease's clinical and epidemiological characteristics, we have systematically reviewed the existing literature.

Methods We performed an extensive review of articles on THC across four key scientific databases: PubMed, Scopus, Web of Science, and Google Scholar. Our study encompassed all patients diagnosed with THC through post-surgical pathology or Fine Needle Aspiration Cytology (FNAC) examinations, extracting clinical, epidemiological, and therapeutic data of THC patients from publications up to October 2023.

Results From 770 articles, 57 met our criteria, detailing 75 THC patients. The gender ratio was 2.36 females per one male. The patients averaged 36.1 years old, with common symptoms including neck mass, hoarseness, shortness of breath, and dysphagia. The left lobe was involved in most patients, and only 21.3% had extrathyroidal involvement. Cysts averaged 36.4 mm in diameter, with cystic nodules being the most frequent imaging finding (91.2%). Serological tests were performed for 42.6% of cases, of which 62.5% were positive. Surgery was undertaken in 71 patients (94.6%).

Conclusion Cystic echinococcosis (CE) of the thyroid should be considered as part of the differential diagnosis in patients with cervicofacial mass, especially in endemic countries. The present study provides reliable data to improve our understanding of the features of the disease for a better diagnosis and management.

Keywords Hydatid disease, Thyroid nodule, Neck mass, Thyroidectomy, Fine needle aspiration cytology, Albendazole therapy

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Introduction

Cystic echinococcosis (CE) is a serious and potentially life-threatening zoonotic infection caused by the larval stage of the tapeworm *Echinococcus granulosus*. CE affects human as well as different livestock species. It impacts over a million individuals globally and is distributed all over the world except Antarctica [1, 2]. In some endemic areas, the incidence of CE may reach 50 per 100,000 people, with a high public health and economic impact on societies in low-middle-income countries [3].

The parasite eggs excreted from dog feces can infect human and livestock. The eggs hatch in the human small intestine, and the embryos penetrate through the intestinal wall, move through the portal vein to different organs and develop into a fluid-filled space-occupying lesion known as Hydatid Cyst (HC) [4].

Clinical manifestations of CE are related to the organ location, size, and number of cysts. In 90% of CE patients HC develops in the liver and lungs. Other organs are less frequently affected, including the spleen, brain, kidneys, heart, orbit, muscles, and both endocrine and exocrine glands [5, 6]. Thyroid is one of the very rare locations of HC compared to other organs [7]. It is estimated that thyroid hydatid cyst (THC) accounts for approximately 2% of all thyroid nodules in some medical centers [8]; however, due to the difficulties in diagnosis, this percentage may be variable in different centers. THC commonly manifests as a neck mass, alongside symptoms such as hoarseness, dysphagia, and dyspnea, with less frequently encountered yet more severe symptoms such as the development of tracheal fistula. These symptoms may closely mimic the clinical presentation of other critical disorders, including thyroid cancer [9, 10]. Moreover, the lack of typical inflammatory signs, including fever and erythema, presents another challenge in identifying THC as a differential diagnosis of thyroid nodules. Without careful evaluation, THC may cause diagnostic and therapeutic challenges [11].

Approximately 3–7% of adults have nodular thyroid disease, based on the findings in physical examination ; however, ultrasonography (US) examinations reveal thyroid nodules in approximately half of adults, most of which are smaller than one centimeter [12]. Among the various conditions that can be diagnosed in thyroid nodules, THC stands out as a distinct possibility, albeit a rare one.

The clinical nature of THC needs to be better understood. Unfortunately, our knowledge of the characteristics, clinical course, diagnosis, treatment outcome, and prognosis of THC is limited, and most of the data published on THC are case reports. In this study, we aimed to conduct a systematic review of the literature published on THC in the scientific databases to identify different clinical and para-clinical aspects of the disease.

Methods

This systematic review followed the PRISMA guidelines for 2020, with the relevant details provided in Supplementary Table 1 [13]. The study review protocol was registered in the International Prospective Register of Systematic Reviews (PROSPERO) with CRD42022378700.

Search strategy

We conducted a comprehensive search for articles on THC in PubMed, Scopus, Web of Science, and Google Scholar. From inception to October 21, 2023, various keyword combinations utilizing Boolean operators were employed to find related research articles involving CE of the thyroid, including terms such as 'thyroid', 'echinococcosis', 'hydatidosis', 'hydatid disease', 'hydatid cyst', and 'Echinococcus'. The first 100 records from a Google Scholar search for "thyroid hydatid cyst" or "thyroid echinococcosis" were also included. We also scrutinized the reference lists of included studies to uncover any additional potentially related reports on the CE of the thyroid. Supplementary file 1 presents a detailed description of the search strategies in the four databases.

Eligibility criteria, study selection, and data extraction

This study includes definite cases of individuals with either a confirmed post-surgical pathology examination or a fine needle aspiration cytology (FNAC) test confirming the presence of THC.

The review included any article that featured definite patients diagnosed with THC, provided the title and abstract were in English. Non-English articles with English titles/abstracts were also included, with the original content translated using Google Translate. Articles reporting animal studies, editorials, latter to the editors, cellular and molecular studies, and narrative reviews were excluded. We tried to contact the authors of the articles that needed to contain adequate data and the articles for which we needed access to the full text. After removing duplicate records, two authors independently evaluated each article's title, abstract, and full text. The third author assisted in resolving disagreements.

If available, the following information was extracted from the articles: demographic data (patient age and sex, location of residence i.e. urban, rural), clinical and diagnostic data (signs and symptoms, imaging findings, cyst location, FNAC findings, other diagnostic tests, postoperative outcome and follow up), treatment method and bibliographical data (authors and year of publication).

Quality assessment

For the quality assessment, the Joanna Briggs Institute (JBI) critical appraisal tools were used [14]. According to the article type, the papers were categorized into case

reports or series. Two authors carefully evaluated the included papers, and discussions were held to determine the final scores (supplementary Table 3).

Statistical analysis

We presented the findings using descriptive statistics. Categorical variables were summarized using frequency and percent, and continuous variables were summarized using median, mean, and standard deviation (SD). We used SPSS software (ver. 27, IBM, Armonk, NY) to analyze the data.

Results

As shown in Fig. 1, 770 papers were found by searching in four major databases. After removing duplicates and careful screening of the articles, 75 THC patients reported in 57 articles were included in the study (Supplementary Table 2). The range of publication dates for the articles included in our review spans from 1948 to 2023. The quality assessment was done using the JBI tool for 47 case reports and 10 case series articles. The most frequent cause of bias in the articles included in our study was the lack of adequate reporting of demographic and treatment data, including surgical procedures (Supplementary Table 3).

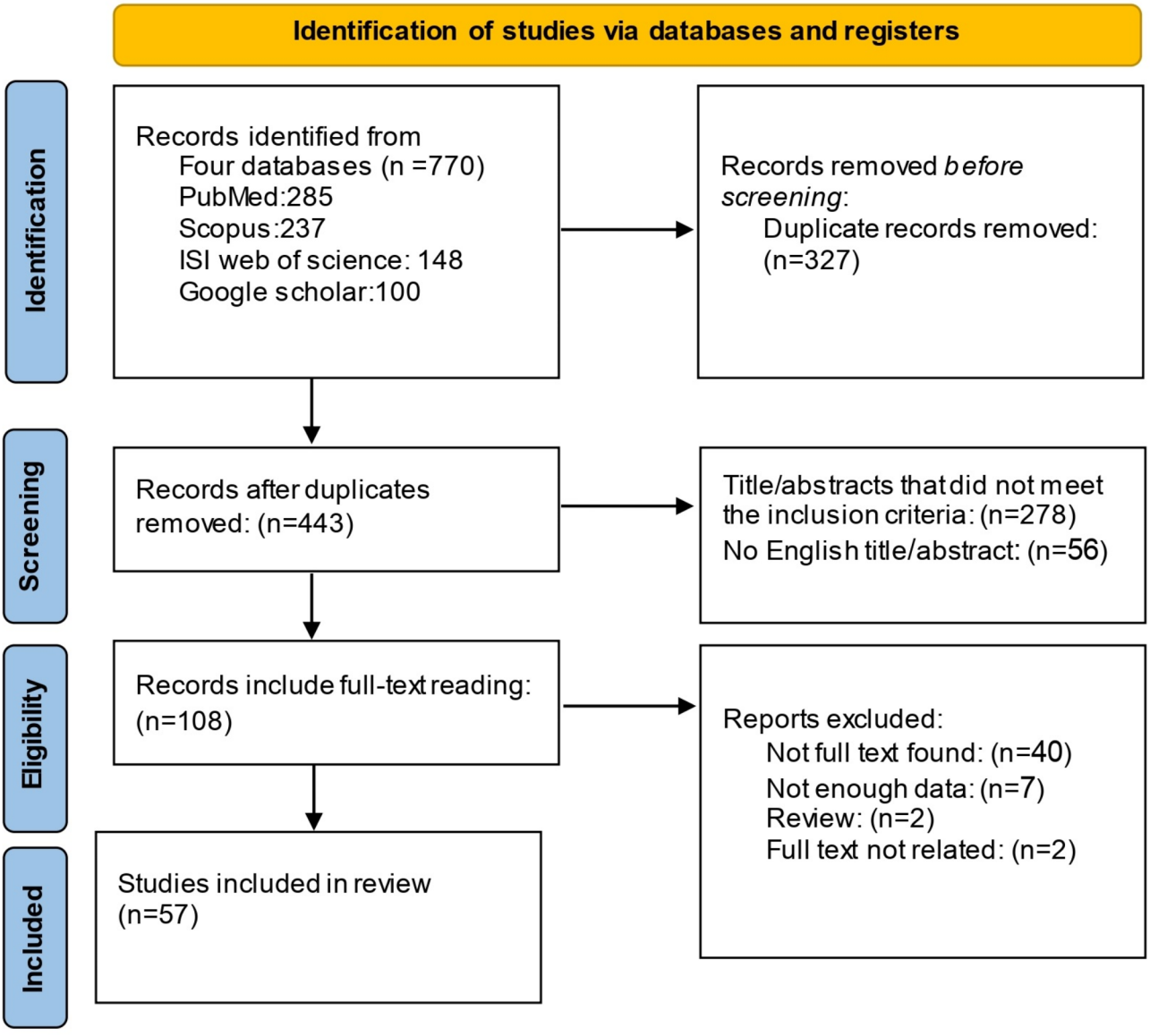

Fig. 1 The PRISMA flow chart summarizing different phases of the systematic review

Table 1 Demographic and epidemiologic features of 75 patients with cystic echinococcosis of thyroid

Variable		Frequency (%)
Age group (year) (*n* = 74)	0–20	15 (20.3)
	21–40	30 (40.5)
	41–60	20 (27)
	Above 60	9 (12.2)
Gender (n = 74)	Male	22 (29.7)
	Female	52 (70.3)
Residency (n = 75)	Rural	24 (32)
	Urban	9 (12)
Country(n = 75)	Turkey	25 (33.2)
	Morocco	8 (10.7)
	Iran	8 (10.7)
	Tunisia	6 (8)
	India	6 (8)

Table 2 Clinical features and cyst locations in 75 patients with cystic echinococcosis of thyroid

Variable		Frequency (%)
Symptom (*n* = 73)	Neck mass	69 (94.5)
	Hoarseness	9 (12.3)
	Breath-shortness	9 (12.3)
	Dysphagia	5 (6.8)
	Pain	4 (5.5)
Location (*n* = 75)	Left lobe ± isthmus	38 (50.7)
	Right lobe ± isthmus	27 (36)
	Both lobes	7 (9.3)
	Isthmus	1 (1.3)
Organ involvement (*n* = 75)	Only thyroid	49 (65.3)
	Liver	5 (6.7)
	Neck tissue	5 (6.7)
	Lung	2 (2.7)
	Liver and lung	2 (2.7)
	Kidney	1 (1.3)
	Disseminated	1 (1.3)

Demographic and clinical features

Table 1 summarizes the demographic and epidemiological characteristics of patients diagnosed with THC. The mean age of THC patients was 36.1 ± 18.64 years, ranging between 6 and 79 years, with a female to male ratio of 2.36 to 1. Most of the THC cases were reported from Turkey (33.2%, Table 1). The clinical characteristics of THC patients are outlined in Table 2.

The most common clinical finding in THC was neck mass (94.5%). In the majority of THC patients (65.3%), the thyroid was identified as the primary location of infection.

The predominant location of thyroid involvement was the left lobe, with or without the isthmus involvement, accounting for 50.7% of cases. The average size of the largest diameter of the cyst in each person, as measured by imaging, was reported to be 36.4 ± 15.3 mm.

Table 3 Diagnostic and treatment characteristics of 75 patients with cystic echinococcosis of thyroid

Variable		Frequency (%)
Radiologic findings (*n* = 68)	Cystic nodule	62 (91.2)
	Multi-vesicular nodule	12 (17.6)
	Calcification	11 (16.2)
	Heterogeneous	7 (10.3)
FNAC[1] (*n* = 75)	Clear fluid	8 (10.7)
	Diagnostic	8 (10.7)
	Benign findings	5 (6.7)
	Not done	26 (34.7)
	N/d[2]	28(37/2)
Serology (*n* = 32)	Positive	20 (62.5)
	Negative	12 (37.5)
Treatment (*n* = 75)	Surgery	36 (48)
	Surgery + BMZ[3]	35 (46.7)
	BMZ	2 (2.7)
	PAIR + BMZ	1 (1.3)
Surgery(*n* = 71)	Subtotal thyroidectomy	39 (54.9)
	Total thyroidectomy	13 (18.3)
	Cystectomy	12 (16.9)
	N/d	7 (9.8)
Follow up (*n* = 75)	NER[4]	36 (48)
	Complication	2 (2.7)
	N/d	37 (49.3)

1: Fine Needle Aspiration Cytology; 2: not defined; 3: benzimidazole; 4: no evidence of recurrence

Diagnostic workup and management approach

Table 3 presents diagnostic findings, treatment approaches, and patient follow-up details. Cystic nodule was the most common finding in diagnostic imaging (91.2%). In our study, 21 out of the 75 patients (28%) underwent FNAC. Contrarily, the authors noted that FNAC was intentionally omitted in 34.7% of cases to prevent the potential occurrence of anaphylactic shock. Parasite materials, including protoscoleces, brood capsules, daughter cysts, or hooklets, were found in the cytology of 10.7% of the cases. Out of the 43 patients having thyroid function tests, two exhibited elevated TSH levels. Hypereosinophilia was found in four patients.

In patients with THC, the median duration from the onset of symptoms to the commencement of treatment was 12 months, ranging from 2 to 240 months. Thyroid surgery was performed for 71 patients (94.7%). Different types of surgery include total thyroidectomy, subtotal thyroidectomy, lobectomy, and cystectomy (Table 3).

Prognosis and follow-up

The median duration in those patients with follow-up was 14 months (ranged 2-264 months). On follow-up, no evidence of recurrence was found in 48% of the patients. Complications were reported in two patients (2.7%), including THC recurrence and a pseudotumor developed following the puncture, aspiration, injection,

and re-aspiration (PAIR) procedure on the cyst (Supp. Table 2).

Discussion

This study presented a systematic review on the literature published on THC to identify different clinical and para-clinical aspects of the disease. Our knowledge on the characteristics, clinical course, diagnosis, treatment outcome and prognosis of THC is limited and most of the data published on THC are case reports. Fifty-seven studies, reporting 75 cases of THC were reviewed in our study. Our findings indicate that majority of the patients were between 21 and 40 years old, and this is in line with the overall age-specific data for human CE [15]. Despite previous research indicating no significant gender difference in CE [16], our study reveals a marked gender imbalance in the occurrence of THC. Specifically, women are found to be 2.36 times more prone to THC than men. This discrepancy can be attributed to the higher prevalence of thyroid nodules among females, which in turn increases the likelihood of diagnostic investigations for thyroid-related illnesses encompassing THC [17].

The majority of existing literature on THC is derived from case reports originating from endemic areas. Over half of the studies we identified in our literature search, were carried out in the endemic countries such as Turkey, Morocco, and Iran. THC is particularly prevalent in rural settings, where individuals engage more frequently with livestock [18]. In the context of our study, the rural population was approximately twice as large as the urban population, with a rural-to-urban ratio of 2.66 to 1.

CE is frequently regarded as a chronic disease that may remain undetected for a considerable period of time, exhibiting minimal or no symptoms. Only when the disease advances and exerts enough pressure on surrounding tissues causing neck mass, hoarseness, dyspnea, dysphagia, and neck pain [3]. Lymphadenopathy and inflammatory symptoms such as fever and redness are relatively uncommon in cases of THC [19]. In our study, the most common clinical presentation of THC was neck mass. The median duration of presentation of neck mass was 12 months, based on the patient's history.

The most common differential diagnoses of THC mentioned in the articles include multi-nodular goiter, colloid cyst, and malignancy [9, 10, 20]. Clinical history, physical examination, TSH measurement, and FNAC are among the main methods for evaluating thyroid nodules and reaching a definitive diagnosis. Typically, thyroid cancer manifests as a firm or solid nodule, which may lead to symptoms such as cervical lymphadenopathy and hoarseness. A history of rapid nodule growth is a risk factor for developing thyroid cancer [21]. One case presented with two concurrent conditions: THC and papillary thyroid carcinoma. This highlights the importance of complete evaluation for thyroid cancers in patients with THC who have risk factors for cancer, ensuring that all potential malignancies are identified and managed appropriately [22]. Similar to thyroid cancer, a THC may become attached to neighboring anatomical structures as it expands, encompassing the trachea, esophagus, carotid sheath, recurrent laryngeal nerve, and strap muscles. As a result, the patient might exhibit pressure symptoms and signs such as vocal cord paralysis, shortness of breath, hoarseness, or dysphagia [20, 23].

Nodular goiter and colloid cysts are frequently identified solely as a mass in the neck region. An expanding nodule may occasionally exert pressure, a characteristic often seen in THC. Consequently, the differentiation between THC, nodular goiter, and colloid cysts poses a significant challenge, necessitating additional diagnostic evaluations including FNAC, imaging studies, and serological tests. Maintaining high suspicion regarding CE is essential when attempting to distinguish it from solid thyroid nodules [24].

Diagnosing THC involves primarily the patient's history, physical examination, clinical evaluations, and imaging studies, including US, MRI (magnetic resonance imaging), and CT (computed tomography) scans, along with serological tests and FNAC. US was the most common imaging technique used for the diagnosis. While MRI and CT scans provide complementary diagnostic [25, 26]. Although there are various diagnostic methods available, post-surgical histopathology and molecular parasitology are still considered the gold standards for confirming a THC diagnosis [27, 28]. Among imaging techniques, the US is especially capable of identifying germinal vesicles, which is vital for a preoperative diagnosis of THC. In our study the most common finding in imaging was cystic nodule (91.2%). Our study has identified that calcification was present in 16.2% of the THC cases. Both thyroid nodules and THC frequently exhibit macroscopic calcifications. Thus, the detection of calcification alone is not particularly informative for differentiating between these two conditions. However, the presence of microcalcifications within a nodule is probably not suggestive of THC [29].

In the evaluation of thyroid nodules, FNAC is a standard procedure. In our study, FNAC was conducted on 28% of patients with THC, while it was intentionally omitted from 34.7% of cases to mitigate the potential risk of anaphylactic shock. The practice of FNAC in THC patients is generally discouraged because of the associated risks of severe anaphylactic reactions, including life-threatening shocks. Interestingly, despite these concerns, there is only one documented case report of allergic reactions following FNAC [30]. Further studies are required to evaluate the use of FNAC for THC diagnosis.

In our study, in most cases, thyroid function tests were normal. However, two THC patients showed high TSH, indicating hypothyroidism. Serology was not performed for more than half of the patients. Positive CE serology was found in 62.5% of the tests. Nevertheless, our findings suggest that serodiagnostic tests are beneficial to rule out other differential diagnosis [31].

Two distinct forms of THC can occur within the thyroid gland [25, 32]. Primary THC refers to the condition where the cyst is confined exclusively to the thyroid gland without any indication of involvement of other organs or tissues beyond the thyroid. In contrast, secondary THC involves the presence of cysts in multiple locations throughout the body [32]. The current review indicates that the majority of cases (65.3%) are classified as primary form. The underlying cause for this predominance of primary THC remains unknown. For all THC patients, an imaging study is necessary to evaluate the involvement of other organs before the operation [33]. Given the relative smallness of the thyroid gland, there is a considerable circulatory flow through it, which theoretically increases the likelihood of the parasite reaching the gland. However, THC is an infrequent condition. This rarity can be linked to the narrow diameter of the thyroid arteries and their unique anatomical arrangement, which places it at a right angle to the carotid arteries. This positioning provides the thyroid gland with a degree of defense against the invasive parasite [23].

The standard approach to treating THC is through surgical removal, specifically the complete excision of the cyst. There is a general agreement that, wherever feasible, hydatid cysts should be completely removed via total or subtotal thyroidectomy [23, 24, 34]. In our review, surgical treatment, alone or combining with albendazole, was administered in 94.7% of the patients. In a minority of cases, albendazole therapy served as the sole treatment method.

Surgical interventions carry risks such as seroma formation, hematoma, injury of the recurrent laryngeal nerve, and hypocalcemia. In the context of this review, there was one documented case of anaphylactic reaction resulting from cyst leakage during the procedure. Consequently, caution is advised during the removal of THC [19]. For smaller cysts, preserving the thyroid tissue is advantageous to sustain thyroid hormone [22]. The PAIR technique is one of the alternatives to the surgical method. In this technique, the hospitalization time is less, and it does not have the side effects of thyroidectomy [35]; however, in our study, PAIR was only performed for one patient, and pseudotumor development was reported during the follow-up [26].

It is recommended to perform post-operative US examinations, as these are vital for assessing the long-term effectiveness of the treatment and identifying any potential recurrence of the disease [36]. The median duration of follow-up in the present study was 14 months; in which complications were reported in 2.7%.

The results of the current study should be taken into account with caution, considering the typical limitations present in all systematic reviews, particularly regarding the quality and completeness of the data available. Several studies included in our review failed to offer thorough information on the diagnosis and treatment of THC, including the precise details of the surgical interventions utilized.

In conclusion, our systematic review of patients with THC addressed various facets of the condition, encompassing its location, clinical manifestations, diagnostic methods, therapeutic approaches, and post-treatment complications. The insights gained from this study can serve as a foundation for future longitudinal investigations and the development of evidence-driven strategies for managing CE of the thyroid. It is essential to consider THC within the differential diagnosis for patients presenting with cervicofacial masses, particularly in populations from endemic regions.

Abbreviations

Computed	Tomography scans (CT scans)
Cystic	Echinococcosis (CE)
Fine	Needle Aspiration Cytology (FNAC)
Hydatid	Cyst (HC)
Magnetic	Resonance imaging (MRI)
Puncture	Aspiration, Injection, and Re-aspiration (PAIR)
Thyroid	Hydatid Cyst (THC)
Thyroid	Stimulating Hormone (TSH)
Ultrasonography	(US)

Supplementary Information

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Supplementary Material 1: Search strategy

Supplementary Material 2: Supplementary Table 1. PRISMA 2020 checklist showing locations where different sections of the systematic review are reported

Supplementary Material 3: Supplementary Table 2. The master table showing data extracted from 57 articles reporting 75 cases of cystic echinococcosis of thyroid

Supplementary Material 4: Supplementary Table 3. Quality assessment of 47 case report and 10 case series studies using the Joanna Briggs Institute (JBI) critical appraisal tools

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Author contributions
AAD and MFH: conceptualization and study design, AAD, MM, AA: data curation, AAD, MM, AA, AS: data analysis, AAD, AS, MFH: data validation, AAD, MFH: writing – original draft preparation, AAD, MM, AA, AS, MFH: revising and final approval of the manuscript. All authors approved the final version for submission.

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Data availability
All data generated during this study are included in this published article and its supplementary information files.

Declarations

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The authors declare no competing interests.

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