



Cystic Echinococcosis of the Breast – Diagnostic Dilemma or just a Rare Primary Localization

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Abstract

Introduction: Although the liver and lung are the most frequently affected organs in cystic echinococcosis, the cysts may develop in any viscera and tissues. Breast is a rare primary localization with few cases described in the literature. We present an updated and systematic review and discuss the possible mechanisms of spreading, diagnostic and treatment options.

Materials and methods: We performed a literature search in PUBMED using the key words 'hydatid disease', 'cystic echinococcosis' and 'breast echinococcosis' without time limitation. Only studies reporting breast cystic echinococcosis were included.

Results: Overall, 121 cases with cystic echinococcosis and 2 with alveolar echinococcosis were reported. A total of 52 cases were included in the analysis. The mean size of cysts was 5.5 cm (range 1.7-12). The most common clinical presentation was painless lump presented from 4 months to 19 years before the final diagnosis. Most cases had isolated breast CE, few cases had synchronous localizations – femoral, thigh and lung, and previous liver CE. Most were active CL and CE1-2 cysts (72%). Ultrasound was used in 83%, followed by mammography (35%). Fine needle aspiration was reported in 27 cases with positive finding in 59%.

Conclusions: In cases with cystic breast lesions from endemic regions we recommend the US as a gold standard. CT and MRT are more accurate but expensive tools without the potential to change the surgical tactic. In contrast to the other localizations of CE, complete excision of the cysts is the best diagnostic and treatment approach.

Keywords

breast, cystic echinococcosis, hydatid disease

INTRODUCTION

Cystic echinococcosis (CE) is a zoonotic disease with high prevalence in South-Eastern Europe, the Middle East, South Africa, South America and Australia. Although the liver (70%) and lung (20-30%) are the most common primary sites, the cysts may develop in all viscera and tissues (8-10%).¹⁻³ Recently, there have been reports from Turkey of unusual locations in 22%, based on 10-year experience.⁴ Breast is an extremely rare primary site and the most frequently cited rate in the literature is 0.27%.⁵ However, we believe this rate to be incorrect because most authors mechanically cited the study of Abi et al., based on 40 cases with rare localizations, published in 1989.³ The aim of the present work was to perform an updated and systematic review of the published literature reporting breast CE and to discuss the possible diagnostic and treatment dilemmas.

MATERIALS AND METHODS

Literature search in PUBMED and Google using the key words 'hydatid disease', 'cystic echinococcosis', and 'breast echinococcosis', without time limitation was performed. Only studies reporting breast CE were included. The following variables were analysed: distribution by continents and country of origin, age, gender, type and duration of the complaints, size and stage of the cysts, synchronous localizations and diagnostic modalities used.

RESULTS

Using all key words, we identified a total of 2646 papers written between 1948 and 2018. Using only 'breast echinococcosis' as a key word, we found a total of 127 papers. Of the latter, 40 were excluded because of reporting different other localizations, and 87 were considered eligible. The analysis included a total of 53 full text papers, whereas 34 full texts papers were not available, but the cases were included in the total count. The PRISMA flow chart is shown in Fig. 1.

Overall, 121 cases with CE and 2 with alveolar echinococcosis were reported in the literature up to October 2018. The distribution of the cases by continents and countries is given in Figs 2, 3.

There was available information for demography, clinical presentation and diagnostic work-up for 52 cases and they were included in the analysis. All cases but one⁶ concerned women. The mean age was 40.5 years (range 18-70 yrs). The size of the cysts was 5.5 cm (range 1.7-12 cm). In the vast majority of the cases there was only one cyst, whereas multiple cysts were found in two cases.^{7,8} The most common clinical presentation was slowly growing painless lump. Abscess was initial presentation in 2 cases.^{9,10} The lump was presented from 4 months to 19 years before the referral to a

specialist. Most cases had isolated breast CE and only in few cases synchronous localizations were presented as follows – femoral (n=1)¹¹, thigh and lung CE (n=1)¹², liver (n=2)^{13,14}, lung¹⁵, pancreas², spleen¹⁰ in two cases with previous liver¹⁶ and lung CE¹⁷. Ultrasound (US) was the most frequently used diagnostic modality, whereas computed tomography (CT) and magnetic resonance tomography (MRT) were used in few cases (Fig. 4). Mammography was used in 35% of the cases and has been gradually replaced by US in the last two decades. Fine needle aspiration (FNA) with cytologic examination was positive (scattered hooklets, scolices and laminated membrane) in 59% of the cases (16/27). Excision biopsy was reported in only two cases. In one case elevated CA 19-9 level was noted.¹⁸

The cysts were classified according to WHO classification^{19,20} based on the statement of authors and the available US images or gross specimens (n=50). Most of them were active CL and CE1-2 cysts (72%), 14% were transitional, whereas 14% were inactive. (Fig. 5).

One case refused operation and was followed-up for two years without progression of the cyst.²¹ There was no sufficient and consistent information regarding the treatment with albendazole – most cases were not treated, some authors reported a 4-week postoperative course, whereas few reported preoperative course. Only few papers reported follow-up studies – there were no recurrences after a mean follow-up of 38 months (3-60).^{5,10-12,16,22-28}

Additionally, two cases with alveolar echinococcosis were reported in the literature (from Bulgaria and Turkey).^{29,30}

DISCUSSION

According to World Health Organization (WHO) the term Cystic Echinococcosis (CE) designates the disease caused by *E. granulosus*. Unfortunately, the terms 'hydatid disease' and 'hydatid cysts' are still commonly misused in the literature.³¹

Bulgaria is a classical endemic region with prevalence 2.37/100 000 and steady increase in incidence from 2.0 to 3.1/100 000 for males and from 2.0 to 3.4/100 000 among women over a span of 45 years (1950-1995). Within the same period, 12343 operated cases were recorded, whereas 556 newly diagnosed cases was registered in 1995. Only two cases with cystic and alveolar breast echinococcosis were reported (0.016%).^{29,32}

The most commonly cited frequency of breast CE is 0.27% of all cases. In fact, we found only 121 cases for the search period. In a series with 661 cases Akcam et al. reported unusual localizations in 20% with breast CE in 0.15% (n=1).³³ Depending on the developmental stage, the cysts may mimic simple cysts, fibroadenoma, phylloides tumor, inflammatory cancer, mastitis, abscess or rare mammary abnormalities, which should be included in the differential diagnosis.^{9,10,21,34}

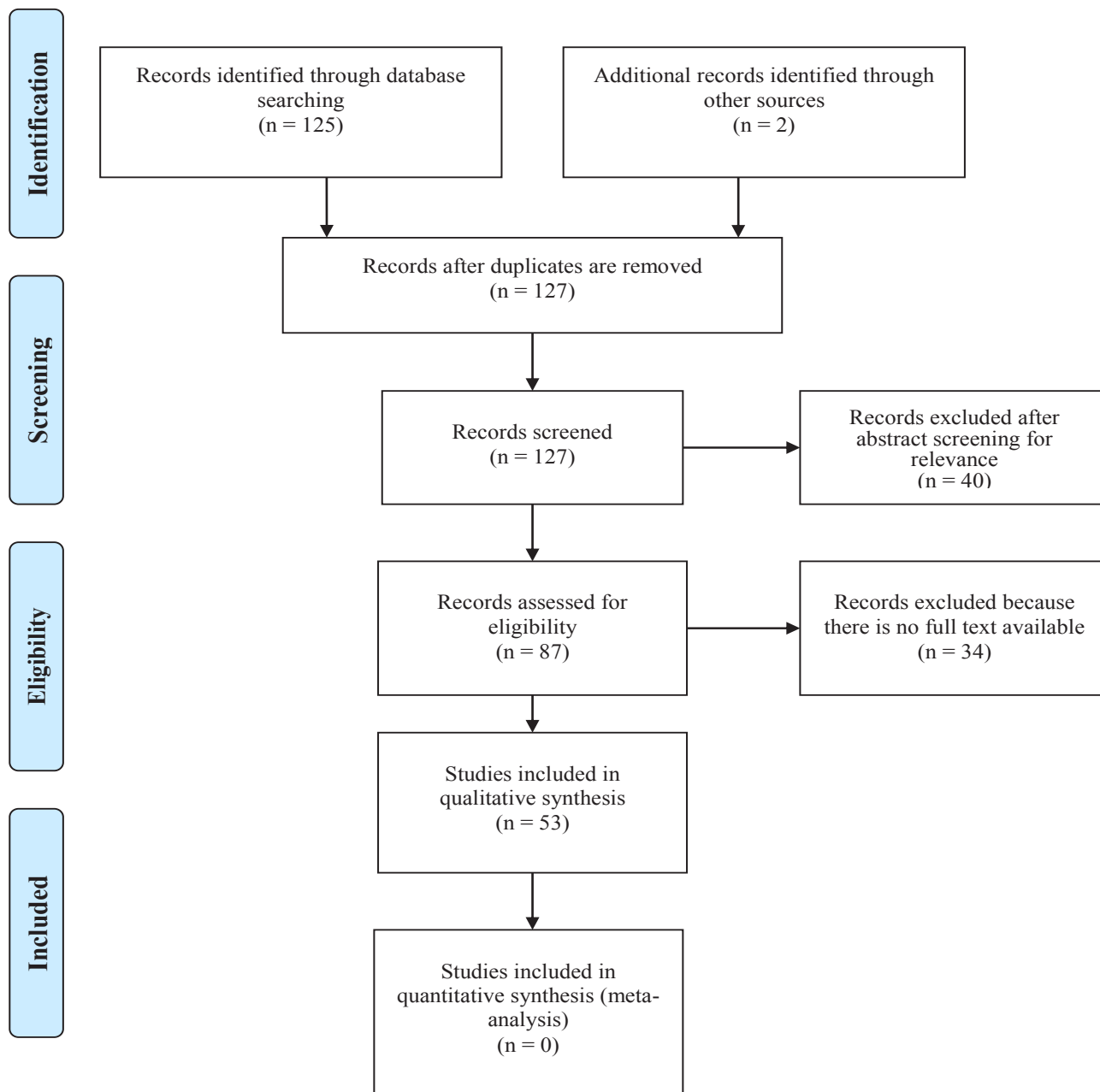


Figure 1. PRISMA 2009 Flow Diagram.

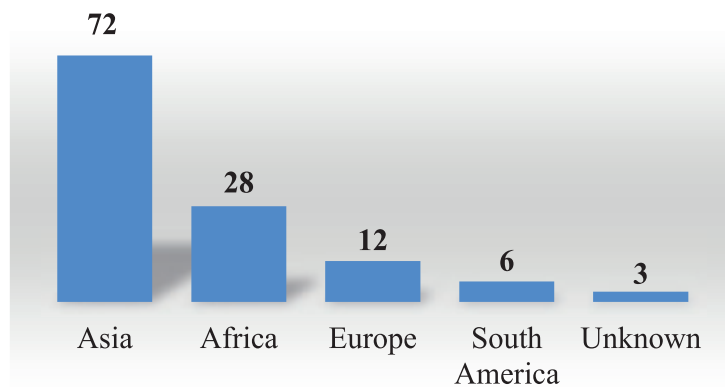


Figure 2. Distribution of cases by continent (n = 121).

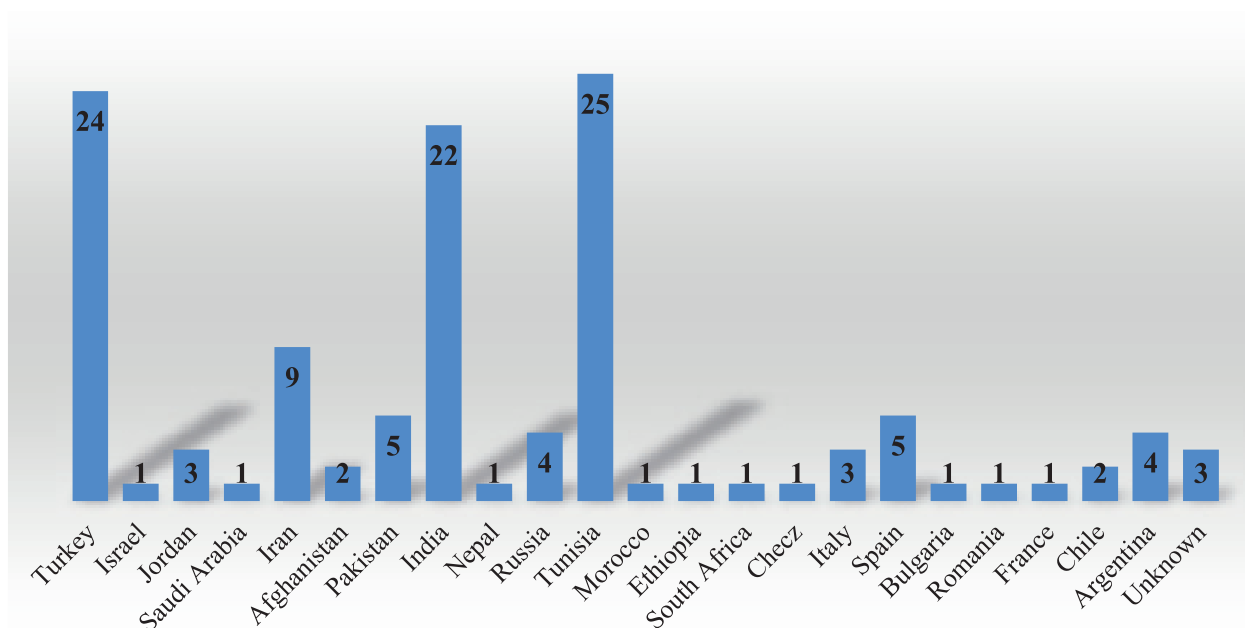


Figure 3. Distribution of cases by country.

Turkey^{2,5,7,12,17,18,22,33,34,37-39,45,47-49,50-54}, Israel⁵⁵, Jordan^{23,56}, Saudi Arabia^{36,57}, Iran^{24,25}, Afghanistan^{25,58}, Pakistan^{8,11,59,60}, India^{9,10,13,16,26,40,44,46,61-72}, Nepal²⁷, Russia^{14,73-75}, Tunisia^{6,15,43,68,76,77}, Morocco⁷⁹, Ethiopia²⁸, South Africa⁸⁰, Czechia⁸¹, Italy⁸²⁻⁸⁴, Spain^{21,41,42,85,86}, Bulgaria³², Romania⁸⁷, France⁸⁸, Chile^{89,90}, Argentina⁹¹⁻⁹³, Unknown country⁹⁴⁻⁹⁶

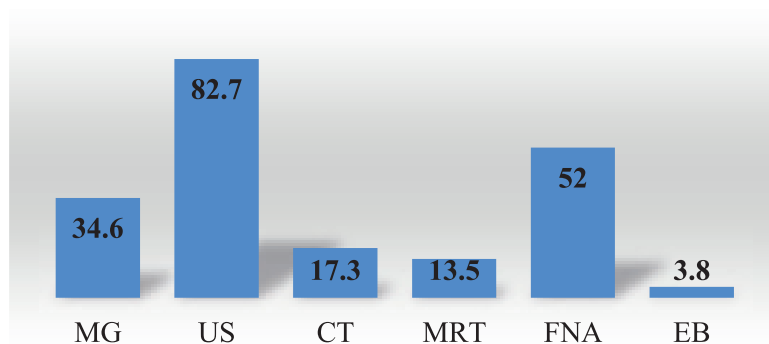


Figure 4. Distribution of the diagnostic modalities (% of 52 patients).

(MG: mammography, US: ultrasound, CT: computed tomography, MRT: magnetic resonance tomography, FNA: fine needle aspiration, EB: excision biopsy).

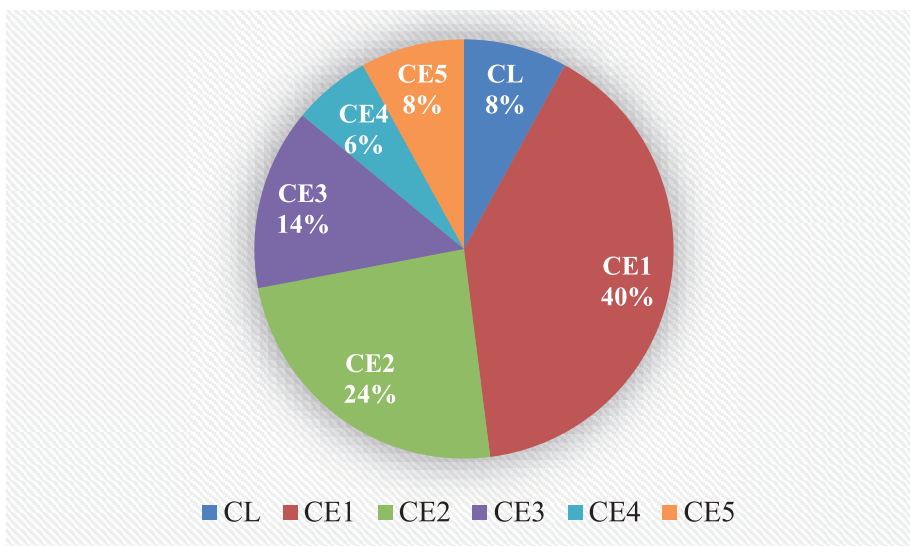


Figure 5. Distribution of cysts according to World Health Organization classification (n=50).

In humans, oncospheres released in the bowel penetrate through the intestinal mucosa and enter into the portal venous system. Due to its location, the liver is the most affected organ (70%). In 20-30% of the cases, oncospheres enter systemic blood stream and initially reach the lung, but in fact all tissues could be affected (10%). Another possible way to reach the lung is to enter the lymphatics of the bowel and through the thoracic duct they pass into the superior vena cava. A report from Turkey described an interesting case with extrapleural parasternal dissemination of liver CE in line with the right internal thoracic artery and intercostal vessels.³⁵ In our view, another possible way for breast involvement is through the lymphatic pathways between liver and breast (Gerota's pathway), similarly to the liver metastasizing of the breast cancer. Only one case in the present series, however, had previous liver CE and another one had previous lung CE. This finding is in contrast to Akcam et al. who reported synchronous liver CE in 24%, lung CE in 5% and both localizations in 1.5% of all cases with unusual CE.³³ Thus, the passing of the systemic blood stream through bowel lymphatics seems to be the most probable route for dissemination.

Mammography is a widely accepted screening tool for breast diseases, despite the fact that it has low specificity. The most common finding in mammography is a ring-shaped dense lesion.³⁶ The recent consensus statement published by WHO Informal Working Group on Echinococcosis (WHO-IWGE) recommended US examination as a very useful screening tool, especially for CE with sensitivity and specificity of 93-98% and 88-90% in endemic areas, respectively.^{19,20} It is considered a gold standard because it is safe and fast bedside procedure which provides an exact description of the developmental stage of CE and reliable comparison of the results. FNA with cytologic examination could reveal scattered hooklets, scolices and

laminated membranes.^{17,36-43} However, it is non-specific (52% in the present series) and carries a risk for dissemination and serious allergic reactions. Computed tomography usually reveals well-defined uni- or multilocular cyst with hypodense fluid and peripheral enhancement, whereas on magnetic resonance breast CE appears as hypointense mass with peripheral isointense capsula on T1-weighted and hyperintense with solid hypointense capsula on T2-weighted images.^{16,36,37,44} Both modalities allow for more precise imaging of the lesions, but actually they are expensive and more importantly, the finding could not change the surgical tactic.^{5,16,36,37,44-46}

In general, the management of CE depends on localization and type of the cysts. For example, inactive (type CE 4 and CE 5) and uncomplicated liver cysts require no treatment. Besides, the total pericystectomy is not always feasible in liver echinococcosis because of the complex liver anatomy. Due to accessibility and relatively simple anatomy of the breast, along with the need for exact differential diagnosis, all suspected cysts in endemic regions are indicated for surgery and the total pericystectomy should be the gold standard. We strongly consider excision biopsy to be the most appropriate diagnostic and therapeutic approach. There was no consistent information in the literature regarding the postoperative treatment with albendazole. In our opinion, in cases with isolated breast CE it is not necessary.

An important limitation of the present study is the lack of search of databases other than PUBMED.

CONCLUSION

The most probable way for an isolated breast involvement is the passing of the systemic blood stream through bowel

lymphatics. The lymphatics between liver and breast (Gero-ta's pathway) represents an unique route for dissemination. In cases with cystic breast lesions from endemic regions we recommend the US as a gold standard. FNA yield frequently equivocal results and carries a risk for dissemination, CT and MRT are more accurate, but expensive tools without the potential to change the surgical tactic. In contrast to the other localizations of CE, the complete excision of the cysts is the best diagnostic and treatment approach and should be the gold standard. In case with isolated breast CE post-operative treatment is not necessary.

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Кистозный эхинококкоз молочной железы диагностическая дилемма или только редкая первичная локализация.

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Абстракт

Введение: Хотя при кистозном эхинококкозе чаще всего поражаются печень и лёгкие, кисты могут развиваться во всех внутренних органах и тканях. Грудь является редкой первичной локализацией, по поводу которой в литературе сообщается лишь о нескольких случаях. Мы представляем обновлённый и систематический обзор и обсуждаем возможные механизмы распространения, диагностические и терапевтические варианты.

Материалы и методы: Мы провели поиск в базе данных PUBMED по ключевым словам «водянка», «кистозный эхинококкоз» и «эхинококкоз молочной железы» без каких-либо ограничений по времени. Были включены только случаи, сообщающие о муковисцидозе.

Результаты: Всего было зарегистрировано 121 случай муковисцидоза. Всего в анализ было включено 52 случая. Средний размер кист составлял 5,5 см (диапазон 1,7-12). Наиболее распространённой клинической картиной была безболезненная опухоль, появившаяся в период от 4 месяцев до 19 лет перед установлением окончательного диагноза. В большинстве случаев имел место изолированный кистозный эхинококкоз (КЭ) молочной железы, в нескольких случаях были синхронные локализации - бедра, бедра и лёгкого, а также ранее существовавший рак печени. Наиболее активными были кисты CL и CE1-2 (72%). УЗИ было использовано в 83%, а затем маммография (35%). Тонкоигольная аспирационная биопсия была зарегистрирована в 27 случаях с положительными результатами в 59%.

Выводы: В случаях с кистозными поражениями молочной железы из эндемичных районов мы рекомендуем применение любого метода диагностики, кроме маммографии и / или ультразвука. КТ и МРТ являются более точными, но дорогостоящими методами без возможности изменения хирургической практики. В отличие от других мест локализации КЭ, тотальное удаление кист является лучшим диагностическим и терапевтическим подходом.

Ключевые слова

Кистозный эхинококкоз, водянка, молочная железа