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Epidemiological and Clinical Characteristics of Urogenital System Cystic Echinococcosis: A Systematic Review

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Background: *Echinococcus granulosus sensu lato (E. granulosus s.l.)* is globally distributed and highly prevalent in parts of South America, the Mediterranean, Eastern Europe, the Middle East, Africa, China, and Australia. This systematic review explores the epidemiological and clinical features of urogenital system-cystic echinococcosis (UGS-CE).

Aims: To systematically review and synthesize available evidence on the epidemiological patterns, clinical presentations, diagnostic methods, and treatment approaches for UGS-CE.

Study Design: Systematic review.

Methods: The study followed the Preferred Reporting Items for Systematic Reviews and Meta-Analyses Guidelines and was registered in PROSPERO (CRD42024613418). A comprehensive literature search was conducted in the PubMed, Scopus, and Web of Science databases for articles published between 1990 and 2024. Studies based on individual data were included if the UGS-CE diagnosis was confirmed by imaging or laboratory methods.

Results: After screening 1976 records, 490 studies were finally included to comprise the following 698 UGS-CE cases: 561 (80.4%) involved the urinary system; 110 (15.8%) involved the female genital system; and 27 (3.9%) involved the male genital system. The kidney (478, 68.5%) was the most commonly affected organ, followed by the adrenal gland (64, 9.2%), the ovary (51, 7.3%), and the uterus (24, 3.4%). Radiological imaging, especially ultrasonography, was the primary diagnostic method used, which is often supported by computed tomography (CT). Treatment was mainly surgical and in combination with antiparasitic therapy. Among the 155 patients who underwent surgical treatment, 134 were initially misdiagnosed with either malignant or benign tumors.

Conclusion: Though rare and often non-specific, UGS-CE must be considered in patients with abdominopelvic or flank pain, hydatiduria, urinary symptoms, infertility, or dysmenorrhea, especially in endemic areas. Early detection and appropriate management are crucial to improving the outcomes. A radiological assessment is the primary diagnostic method, with ultrasonography as the most effective, often supported by CT. Treatment is mainly surgical and complemented by antiparasitic therapy to reduce the recurrence/relaps risk.

INTRODUCTION

Echinococcosis is a widespread zoonotic disease that is caused by the tapeworms of the genus *Echinococcus* belonging to the Taeniidae family.[1] The two major species that are of medical and public health importance are *Echinococcus granulosus sensu lato (E. granulosus s.l.)* and *Echinococcus multilocularis* (*E. multilocularis*) that cause cystic echinococcosis (CE) and alveolar (AE) echinococcosis, respectively. Humans are accidental intermediate hosts that become infected through the ingestion of eggs. After egg ingestion, oncospheres are released in the intestine and develop into hydatid cysts in various organs, most commonly the liver and the lungs (> 90%). Cysts occasionally occur in the kidney, spleen, peritoneal cavity, skin, or muscles (2-3%) and rarely in the heart, brain, vertebrae, or ovaries (< 1%).[2]

Genitourinary involvement is rare and often secondary to hepatic or pulmonary CE. The kidneys are most frequently affected, as shown in 1-5% of cases.[3] Approximately 80% of pelvic CE involves the genital organs, especially the ovaries, followed by the uterus.[4] To better understand urogenital system (UGS)-CE, we performed herein a systematic review of the relevant cases reported between 1990 and 2024. To the best of our knowledge, this is the first systematic review of UGS-CE.

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MATERIALS AND METHODS

Search strategy

This systematic study was conducted according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) Guidelines.[5] The study review protocol was registered in PROSPERO with CRD42024613418. The literature search was performed in the PubMed, Scopus and Web of Science databases published between 1990 and 2024 (last updated date: 05/01/2025). Various keyword combinations with Boolean operators were used to find the articles that involve CE of the UGS, including terms like "renal", "adnexa*", "seminal vesicle*", "bladder", "unusual loca*", and "*echinococcus granulosus.*" The supplementary file presents a detailed description of the search strategies in the databases. No language restrictions were applied. Studies based on individual data were included. The reports were considered eligible if the UGS-CE cases were diagnosed through imaging techniques or laboratory methods.

The exclusion criteria were as follows: articles not related to CE; articles not related to UGS-CE; experimental or veterinary studies; studies with insufficient data; commentaries; books/chapters, meeting papers, and reviews without original data; duplicated data; articles lacking full-text accessibility; articles related to *E. multilocularis* (AE); and serological or community-based studies.

The search was conducted in the PubMed, Scopus, and Web of Science databases. All records were exported to EndNote. The data were transferred to Excel after the duplicate removal. The reference lists of the included articles were also manually reviewed to identify additional studies. Two reviewers assessed the full-text articles to determine the final eligibility, resolving any contradictions among studies through discussion and consensus. One author extracted the required data, which were then cross-checked by a second author. Additionally, the reference lists of the selected full-text papers were manually examined to identify the articles not retrieved by the database search. The information extracted from these reports included the patients' demographic details, namely, age, sex, and region of origin, medical history, presenting signs and symptoms, diagnostic procedures, site, size and developmental stage of the cyst(s), therapeutic interventions, clinical outcomes, and follow-up details. The quality assessment was performed by using the Joanna Briggs Institute (JBI) critical appraisal tolls (https://jbi.global/critical-appraisal-tools) on each study by two investigators. The supplementary file provides comprehensive information covering all aspects of the review process, including the quality assessment (JBI) and the PRISMA abstract checklist.

Statistical analysis

The extracted data were entered into Excel forms and later transformed into Statistical Package for the Social Sciences files for analysis. The findings were presented as descriptive statistics. The categorical variables were summarized using frequencies and percentages, while the continuous ones were reported as mean, standard deviation, median, and interquartile range (IQR). A formal meta-analysis was not conducted considering the predominance of the case reports and the heterogeneity of the available data.

RESULTS

A total of 3423 records were identified in this work. After excluding 1447 duplicates or irrelevant entries, 1976 records were screened, consequently resulting in 472 eligible reports. An additional 18 studies were identified through the reference lists, yielding a total of 490 included studies (Figure 1). These 490 studies published between 1990 and 2024 reported 698 cases from 46 countries, with Türkiye and India contributing 122 reports each (24.9%). Among the studies, 97.1% were case reports; 1.6% were reviews; 1% were letters; and 0.2% were original research. A total of 698 cases were predominantly reported from Türkiye (27.2%), India (19.7%), Iran (7.6%), Greece and Tunisia (each 6.1%), Morocco (5.4%), and Spain (5.1%). One case lacked geographic data (Figure 2). Female genital CE was mostly reported in India, Türkiye, and Iran, while male genital CE was mostly determined in Tunisia, Türkiye, and Greece.

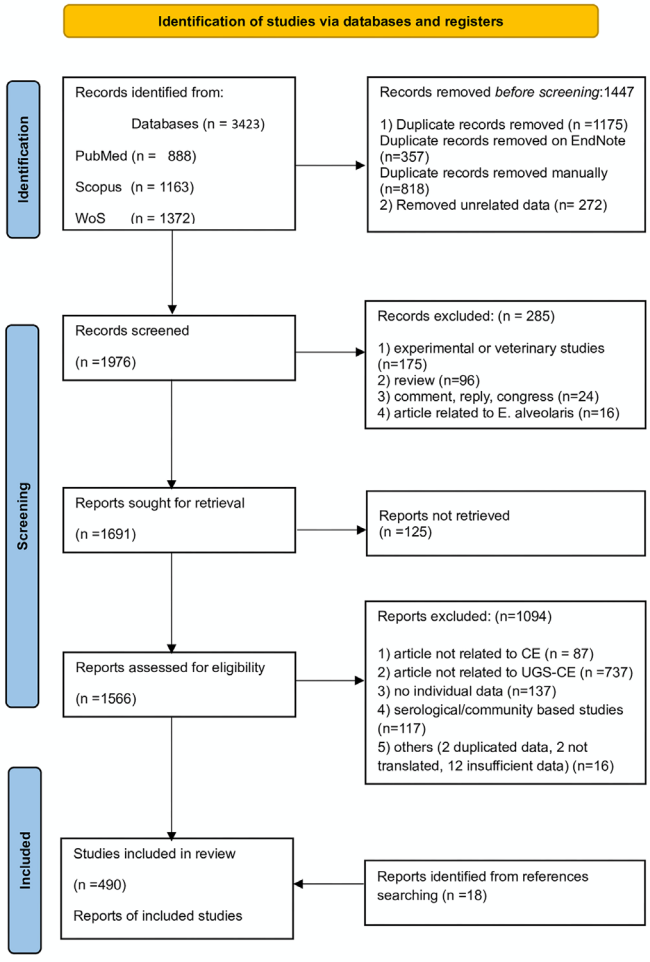

FIG. 1. The PRISMA flow chart summarizing different phases of the systematic review. Source: Page et al.[5].
(The flow diagram adapted from: https://www.prisma-statement.org/prisma-2020-flow-diagram). PRISMA, preferred reporting items for systematic reviews and meta-analyses.

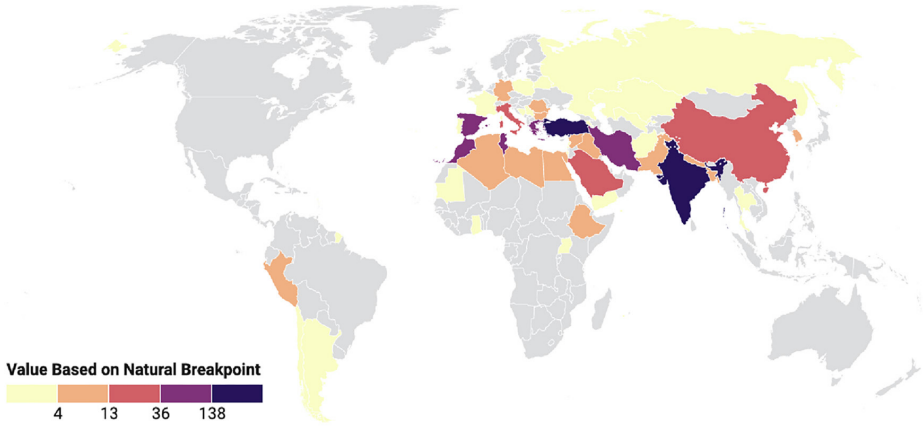

FIG. 2. Geographical distribution of urogenital cystic echinococcosis cases by country. The map adapted from Datawrapper (https://www.datawrapper.de) (free).

Anatomical locations

This systematic review analyzed 698 cases, with 561 (80.4%) pertaining to the urinary system, 110 (15.8%) to the female genital system, and 27 (3.9%) to the male genital system. When the 698 UGS cases were collectively evaluated, involvement was predominantly observed in the following organs: 478 (68.5%) in the kidney, 64 (9.2%) in the adrenal gland, 51 (7.3%) in the ovary, and 24 (3.4%) in the uterus (Table 1).

Urinary system: Most of the urinary cases (85.2%) affected the kidneys, followed by the adrenal glands (11.4%), bladder (2.1%), and ureter (0.5%). Four cases involved multiple urinary organs, and one case had seminal vesicle involvement. Of the 478 renal cases, 49.2% were on each side; 1.7% were bilateral; and laterality was unspecified in 63 cases. Among the localized cases, the upper pole was the most affected. Multiple cysts were seen in some cases, with the localization details lacking in others. Among 64 cases, 26 were left-sided; 35 were right-sided; and one was bilateral.

TABLE 1. Anatomical Locations of Cystic Echinococcosis in Urogenital System.

	Organ	Overall in urogenital system (%)	Within system %	Localization (%)			
				Left	Right	Bilateral	No data
Urinary system (n = 561)	Kidney	478 (68.5)	85.2	204 (49.2)	204 (49.2)	7 (1.7)	63
	Adrenal gland	64 (9.2)	11.4	26 (41.9)	35 (56.5)	1 (1.6)	2
	Bladder	12 (1.7)	2.1	-	-	-	-
	Ureter	3 (0.4)	0.5	1 (100.0)	2 (50)	-	2
	Involvement of ≥2 urinary sys organs	4 (0.6)	0.7	2 (50)	-	-	-
Female genital system (n = 110)	Ovary	51 (7.3)	46.4	21 (52.5)	12 (30.0)	7 (17.5)	11
	Uterus	24 (3.4)	21.8	-	-	-	-
	Adnexa	15 (2.1)	13.6	5 (33.3)	9 (60.0)	1 (6.7)	-
	Fallopian tube	8 (1.1)	7.3	3 (37.5)	4 (50.0)	1 (12.5)	-
	Broad ligament	5 (0.7)	4.5	1 (100.0)	-	-	4
	Cervix	2 (0.3)	1.8	-	-	-	-
	Involvement of ≥ 2 female GS organs	5 (0.7)	4.5	1 (100.0)	-	-	4
Male genital system (n = 27)	Seminal vesicle	9 (1.3)	33.3	2 (50.0)	2 (50.0)	-	5
	Scrotum	7 (1.0)	25.9	1 (33.3)	1 (33.3)	1 (33.3)	4
	Spermatic cord	6 (0.9)	22.2	-	-	-	6
	Prostate	3 (0.4)	11.1	-	-	-	2
	Vas deference	1 (0.1)	3.7	1 (100)	-	-	-
	Involvement of ≥ 2 male GS organs	1 (0.1)	3.7	-	-	-	1

Two lacked a side specification. Of the 12 bladder cases, localization was determined in detail in six.

Female genital system: Among 110 cases, ovaries were involved in 46.4%, uterus in 21.8%, adnexa in 13.6%, fallopian tubes in 7.3%, broad ligament in 4.5%, and cervix in 1.8%. Five cases depicted multiorgan involvement. The ovarian cysts were more often left-sided; seven were bilateral. The uterine cysts were mostly located in the posterior wall. The adnexal cases were predominantly right-sided (Figure 3).

Male genital system: Among the 27 cases, the seminal vesicles were affected in 33.3%, scrotum in 25.9%, spermatic cord in 22.2%, prostate in 11.1%, and vas deferens in 3.7%. One case involved both the seminal vesicle and the prostate. Five cases showed multiple cysts mainly in the scrotum, ureter, and vas deferens.

Extraurogenital involvement: The extraurogenital CE screening was conducted in 163 cases, and cysts were found in 132 (81%). The most common concomitant site was the liver. The lung and spleen involvement was frequent in the renal cases, while the abdominopelvic cysts were common with the genital involvement. A history of previous surgery existed in 33 patients with extraurogenital CE (Table 2).

Clinical features and diagnostic characteristics

The mean age in the 698 reported cases was 39.3 ± 19.1 years. Among the 691 cases with gender data, 47% were male, and 53% were female. Out of the 622 urogenital hydatid cyst cases, 94.1% were symptomatic, and 5.9% were asymptomatic. Multiple symptoms were common (n = 571). Flank/lower back pain was the most frequent at 42.6%, followed by abdominal pain at 38.3% and palpable masses at 30.8%. The median symptom duration for the abdominal and flank pain was 6 months. The less common symptoms included weight loss (3.7%) and hypertension (3.6%). On physical exam (n = 231), a palpable mass was noted in 68.6%; abdominal tenderness was observed in 18.6%; and flank tenderness was found in 13.4% (Table 2).

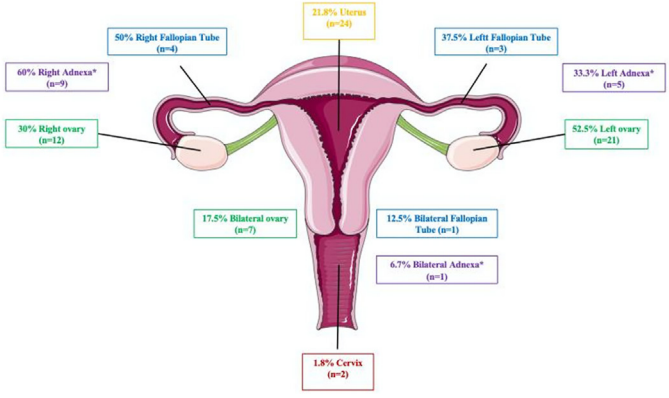

FIG. 3. The location of cysts in the female genital system (Percentages are given as the proportion within each specific location. Locations shown as "no data" in Table 1, for which the exact localization was unknown, were not included). *Cysts included as adnexa are known to be part of the adnexal system, although their exact location remains unidentified. The graphic adapted from Servier Medical Art (https://smart.servier.com). (Free).

Imaging was performed in 682 patients, with most undergoing multiple modalities: computed tomography (CT) (76.5%), ultrasound sonography (USG) (73.2%), magnetic resonance imaging (MRI) (11.8%), X-ray (24.9%), and intravenous pyelogram (IVP) (19.2%). Serology was positive in 69.7% of the 333 tested individuals. Eosinophilia was present in 50.8% of 193 cases. The risk factors were identified in 133 patients: rural residence (24.1%), endemic area (23.3%), animal contact (23.3%), dog contact (11.3%), and farming (9.8%).

Staging was reported in 55 cases. The World Health Organization classification was used in 10 cases: stages 3 (50%), 2 (30%), and 1 (20%). The Gharbi classification applied to 45 cases showed stages 1 (35.6%), 2 (26.7%), 3 (22.2%), 4 (11.1%), and 5 (4.4%).

The cyst size was reported in 478 cases: 85.4% were ≥ 5 cm. Most of these cysts (n = 559) were confined to one organ. Ten had multiorgan involvement within the UGS.

Urinary system: This study evaluated 561 urinary system CE cases. The mean age of the 553 patients was 39.0 ± 19.4 years; eight lacked age data. Among the 554 cases with known gender, 53.8% were male, and 46.2% were female. Gender was unspecified in seven cases. Of the 491 cases assessed for symptoms, 92.7% were symptomatic. The most frequent symptoms were flank/back pain (53.1%), abdominal pain (30.3%), palpable mass (26.5%), hydatiduria (8.8%), and dysuria or urinary retention (7.9%). The less common findings included respiratory symptoms (n = 21), weight loss (n = 19), fatigue/weakness (n = 14), and hypertension (n = 7). On physical examination (n = 161), a palpable mass was found in 69.5%; abdominopelvic tenderness was observed in 16.7%; and flank tenderness was noted in 18.6%. Imaging was conducted in most cases with the following results: CT: 82.4%; USG: 71.7%; X-ray: 26.0%; IVP: 23.7%; and MRI: 9.7%. Additional methods included angiography (n = 14) and scintigraphy (n = 6). Serological tests were performed in 284 cases, yielding 69.4% positivity. Eosinophilia was present in 50.3% of 175 cases. The cyst size was available for 358 cases: 85.2% were ≥ 5 cm. A single-organ involvement was noted in 560 cases. The extraurinary CE screening was done in 117 cases, with positive findings found in 94 (80.3%) (Table 2).

This study analyzed 478 renal hydatid cyst cases with a mean age of 37.5 ± 19.5 years (n = 471). Among the 474 patients with a known gender, 53.6% were male, and 46.4% were female. Of the 414 evaluated cases, 93.3% were symptomatic. The common symptoms included flank/back pain (53.8%), abdominal pain (28.7%), palpable mass (28.5%), hydatiduria (9.8%), and dysuria/urinary retention (6.9%). The less frequent symptoms were respiratory complaints (n = 20), weight loss (n = 15), fatigue (n = 12), and hypertension (n = 2). On physical exam (n = 138), palpable mass was found in 72.4%; abdominopelvic tenderness was observed in 13.7%; and flank tenderness was noted in 18.8%. CT was the most used imaging method (78.9%), followed by USG (71.1%), IVP (26.5%), X-ray (25.1%), and MRI (9.3%). Among the 243 tested patients, serology was positive in 70.8%. Eosinophilia was present in 50.3% of 175 tested cases. The cyst size was ≥ 5 cm in 85.5% of 290 cases. The extrarenal cysts were observed in 81.5% of 108 screened patients (Table 2).

TABLE 2. Location of Extraurogenital Cystic Echinococcosis.

		Location of extraurogenital cysts** patients (%)								
	Numbers of cases*	Liver	Lung	Spleen	Abdomino pelvic cavity	Cardiac	Brain	Muscle	Pancreas	Others
Urinary sytem										
Kidney	84	65 (77.3)	23 (27.3)	16 (19.0)	8 (9.5)	10 (11.9)	8 (9.5)	7 (8.3)	5 (5.9)	4 (4.7)
Adrenal gland	1	1 (100.0)	-	-	-	-	-	-	-	-
Bladder	3	2 (66.7)	-	-	1 (33.3)	-	-	1 (33.3)	-	-
Ureter	1	-	-	-	1 (100.0)	-	-	-	-	-
Female genital system										
Ovary	17	9 (52.9)	-	2 (11.7)	9 (52.9)	-	-	-	-	1 (5.8)
Uterus	5	5 (60.0)	-	-	2 (40.0)	-	-	-	-	-
Adnexa	8	7 (85.7)	-	-	1 (14.3)	-	-	-	-	-
Fallopian tube	3	3 (100.0)	-	-	-	-	-	-	-	-
Broad ligament	3	3 (100.0)	-	-	-	-	-	-	-	-
Male genital system										
Seminal vesicle	2	2 (100.0)	-	1 (50.0)	1 (50.0)	-	-	-	1 (50.0)	-
Scrotum	3	3 (100.0)	-	-	1 (50.0)	-	-	-	-	-

*In 3 cases with multiple organ involvement in the urogenital system, the additional cysts observed were redistributed to each affected organ and recalculated.
**Due to the simultaneous involvement of multiple organs per patient, the frequency of extraurogenital cysts reported exceeds the total number of patients.

This study examined 64 adrenal CE cases with a mean age of 48.5 ± 16.1 years (n = 63). Among the 61 cases with gender data, 54.1% were male. Most patients (86.5%) were symptomatic, with the flank/back pain (52.9%) and abdominal pain (33.3%) being common. The other symptoms included hypertension (n = 5) and weight loss (n = 4). The imaging primarily involved CT (95.2%) and USG (66.6%). Of the 31 serology-tested patients, 58.1% returned positive results. Cysts ≥ 5 cm were observed in 85.2% of the 54 measured cases.

Female genital system: Among 109 cases (mean age: 40.4 ± 17.6), 99% were symptomatic. The common symptoms included abdominal pain (81.7%), palpable mass (42.3%), and urinary complaints (18.2%). The female reproductive symptoms (e.g., infertility and dysmenorrhea) were reported in 16 cases. On physical exam (n = 56), a mass was detected in 76.7%. The imaging primarily involved USG (80.1%) and CT (59.4%). Serology was positive in 78.4% of the 37 tested patients. Most of the cysts (90.8%) were ≥ 5 cm. The extraurogenital involvement was found in 81% of the 42 screened cases.

A total of 51 ovarian CE cases were analyzed (mean age: 41.4 ± 16.9). Nearly all were symptomatic and primarily with abdominal pain (89.3%) and palpable mass (34.0%). The female-specific symptoms occurred in five cases. During the exam (n = 24), a mass was detected in 70.8%. The imaging included USG (72.9%) and CT (64.5%). Serology was positive in 71.4% of the 14 tested cases (Table 3).

Male genital system: Among the 27 male genital CE cases with a mean age of 41.9 ± 18.3, 96.3% were symptomatic. Palpable mass (57.6%) and urinary symptoms (34.6%) were the most frequent symptoms found. On the physical exam (n = 14), a mass was detected in 92.8%. The imaging involved USG (75.0%) and CT (80.1%). Of the 12 serology-tested patients, half were positive. Cysts ≥ 5 cm were found in 63.6% of the 22 measured cases. All four screened cases showed extraurinary CE.

Treatment

The treatment data were available for 669 patients, representing 95.8% of the total cohort. Of these, 630 (94.2%) underwent either surgery or another intervention, while 32 (4.6%) received only medical therapy. The main reasons for the non-surgical treatment were patient refusal (n = 13) and poor clinical condition (n = 2). Among those who underwent surgery or intervention, organ removal was performed in 273 patients (43.3%); cystectomy (surgical removal of the cyst) was done in 270 (42.9%), while puncture, aspiration, injection, and re-aspiration (PAIR) and percutaneous evacuation of residual cavity were conducted in 19 cases, primarily for renal cysts. For the renal involvement in 451 patients, 56.3% received both surgery and medical therapy; 36.4% underwent surgery alone; and 6.7% were treated with medication only. The surgical removal of an organ (n = 188) and cystectomy (n = 157) were the most common procedures. In cases involving the adrenal gland (n = 63), 66.7% underwent surgery; 28.6% received combined treatment; and three were monitored without intervention. For the ovarian cases (n = 50), 60.8% received both surgery and medical therapy, while 38% were treated with surgery alone. Albendazole was the most commonly used drug pre- and post-op. The median preoperative treatment duration was 1.2 months (IQR: 1-3), while the postoperative duration was 3 months (IQR: 1-3). Albendazole was used in 85 among the 88 patients treated both before and after surgery (Table 4). Notably, 134 of the 155 surgically treated patients were initially misdiagnosed with malignancy or benign tumors.

TABLE 3. Clinical Features and Diagnostic Characteristics.

	Numbers of cases (%)			
	All cases (n = 698)	**Urinary system (n = 561)**	**Female genital system (n = 110)**	**Male genital system (n = 27)**
Age mean ± SD [no data]	39.3 ± 19.1 [9]	39.0 ± 19.4 [8]	40.4 ± 17.6 [1]	41.9 ± 18.3
Gender [no data]	[7]	[7]	-	-
Female	325 (47.0)	256 (46.2)	110 (100.0)	-
Male	366 (53.0)	298 (53.8)	-	27 (100.0)
Symptom*	(n = 585)	(n = 455)	(n = 104)	(n = 26)
Abdominopelvic pain	224 (38.3)	138 (30.3)	85 (81.7)	1 (3.8)
Flank/back pain	249 (42.6)	242 (53.1)	5 (4.8)	2 (7.6)
Lump/mass sensation	180 (30.8)	121 (26.5)	44 (42.3)	15 (57.6)
Nausea/vomiting	49 (8.3)	41 (9.0)	8 (7.6)	-
Fever	43 (7.3)	37 (8.1)	5 (4.8)	1 (3.8)
Hematuria	35 (5.9)	34 (7.4)	1 (0.9)	-
Hydatiduria	41 (7.0)	40 (8.8)	-	-
Urinary semptoms	55 (9.4)	36 (7.9)	19 (18.2)	9 (34.6)
Others**	116 (19.8)	86 (18.9)	28 (26.8)	3 (11.5)
Asymptomatic n/N (%)	37/622 (5.9)	36/491 (7.3)	1/105 (0.9)	-
Physical examination*	(n = 231)	(n = 161)	(n = 56)	(n = 14)
Palpable mass	168 (72.7)	112 (69.5)	43 (76.7)	13 (92.8)
Abdominopelvik tenderness	43 (18.6)	27 (16.7)	16 (28.5)	-
Flank/lumbar tenderness	31 (13.4)	30 (18.6)	-	1 (7.2)
Serology	(n = 333)	(n = 284)	(n = 37)	(n = 12)
Positive	232 (69.7)	197 (69.4)	29 (78.4)	6 (50.0)
Negative	101 (30.3)	87 (30.6)	8 (21.6)	6 (50.0)
Eosinophilia	(n = 193)	(n = 175)	(n = 15)	(n = 3)
Positive	98 (50.8)	88 (50.3)	9 (60.0)	1 (33.3)
Negative	95 (49.2)	87 (49.7)	6 (40.0)	2 (66.7)
Imaging techniques performed*	(n = 682)	(n = 552)	(n = 106)	(n = 24)
CT	522 (76.5)	445 (82.4)	63 (59.4)	14 (58.3)
USG	499 (73.2)	396 (71.7)	85 (80.1)	18 (75.0)
MRI	81 (11.8)	54 (9.7)	20 (18.8)	7 (29.1)
Plain radiography	170 (24.9)	144 (26.0)	22 (20.7)	4 (16.6)
Intravenous pyelography	131 (19.2)	131 (23.7)	-	-
Others***	25 (3.6)	25 (4.5)	-	-
Maximum cyst diameter	(n = 478)	(n = 358)	(n = 98)	(n = 22)
< 5 cm	70 (14.6)	53 (14.8)	9 (9.2)	8 (36.4)
≥ 5 cm	408 (85.4)	305 (85.2)	89 (90.8)	14 (63.6)
Cyst structure	(n = 157)	(n = 114)	(n = 38)	(n = 5)

*Since multiple symptoms or physical examination findings may be present simultaneously in a single patient, and more than one imaging modality may be used concurrently, the reported frequencies of symptoms and imaging methods exceed the total number of patients.
**Other symptoms: inguinal/scrotal pain, gastrointestinal system symptoms, neurological symptoms, fatigue/weakness, weight loss, hypertension, respiratory symptoms, dyspareunia/dysmenorrhea/infertility, headache.
***Other imaging tecniques: angiography, scintigraphy, retrograde ureteropyolography, PET/CT.
SD; standard deviation, CT; computed tomography, USG; ultrasound sonography, MRI; magnetic resonance imaging, PET; positron emission tomography.

Outcome

Among the patients, 98.9% fully recovered; 0.2% partially did; and 0.9% died. A complete recovery was observed in 98.9% of the urinary cases, 98.8% of the female genital cases, and 100% of the male genital system cases. The deaths were linked to surgical complications, cyst rupture, or unrelated causes. The follow-up data were available for 319 patients, with a median duration of 12 months (IQR: 6-36). The follow-up was the longest in the multiorgan urinary cases and the shortest in the broad ligament involvement. A recurrence/relaps occurred in 1.9% of 321 cases, typically around 10.5 months (IQR: 3-21) (Table 5).

DISCUSSION

Over 90% of the hydatid cysts occur in the liver and lungs, while the renal, splenic, peritoneal, and muscular involvement is less common.[2] In our review (n = 698), the kidneys were affected in 68.5%; the adrenal glands were affected in 9.2%; the ovaries were affected in 7.3%; and the uterus is affected in 3.4%. Of the 622 symptomatic cases, flank/lower back pain was the most common complaint at 42.6%.

Renal CE is often asymptomatic or presents with non-specific symptoms. Hydatiduria pathognomonic but rare (10-20%) may occur with the cyst rupture into the collecting system. The less common symptoms include urinary retention, anuria, or subcostal pain.[6-11] In this review, 93.3% of the renal CE cases were symptomatic. The common symptoms included flank pain (53.8%), palpable mass (28.5%), hydatiduria (9.8%), and urinary symptoms (6.9%). A mass was the most common finding on examination.

Most of the renal cysts were unilateral and localized to the renal poles. The bilateral cases were rare (1.7%). While the plain radiographs are non-specific, CT is found to be more sensitive for renal CE and useful in pre-op planning or detecting urinary tract communication. MRI offers no added advantage over CT.[12] In this review, CT is the most common modality, followed by USG, IVP, direct X-ray, and MRI.

The treatment aims to stage the disease, prevent recurrence, and preserve the renal function. Active/transitional cysts require intervention, while inactive ones may be monitored.[13] Relevant options include medication, surgery (open/laparoscopic), and PAIR, although PAIR is rarely used in renal CE.[14,15] Surgery remains the mainstay and is often paired with albendazole. Of the 451 renal cases investigated most received surgery plus medication. Nephrectomy was performed in 188 cases, cystectomy in 157, and interventional radiology in 17. Adrenal involvement is < 1% of the cases[16,17], and more than 90% of the cases are unilateral. They are rarely complicated and become symptomatic most of the time because of symptoms developing as a pressure consequence. Most cases are treated surgically via total cyst excision.[18-22] In our review, 86.5% were symptomatic (flank pain: 52.9%). The surgical treatment included organectomy (n = 25), cystectomy (n = 29), and interventional methods (n = 2).

TABLE 4. Overview of Treatment Methods for Cystic Echinococcosis.

	Number of cases (%)	Duration of medical treatment in months median (IQR)/n
Treatment methods (n = 669)		
Surgery/IR only	267 (39.9)	
Surgery/IR and medical treatment	363 (54.3)	
Medical treatment only	32 (4.6)	
Watch and wait	7 (1.0)	
Surgery/interventional radiology methods (n = 630)		
Surgical removal of the organ	273 (43.3)	
Surgical excision of the cyst	270 (42.9)	
Surgery with unknown method	68 (10.8)	
PAIR/PERC	19 (3.0)	
Preoperative medical treatment (n = 157)		**1.2 (1-3)/100**
Albendazole monotherapy	150 (95.5)	1.1 (1-3)/94
Mebendazole monotherapy	6 (3.8)	2 (1.2-5)/5
Albendazole + mebendazole	1 (0.6)	1 (1-1)/1
Postoperative medical treatment (n = 294)		**3 (1-3)/208**
Albendazole monotherapy	267 (90.8)	2.2 (1-3)/188
Mebendazole monotherapy	25 (8.5)	3.5 (1-5)/18
Albendazole + mebendazole	2 (0.7)	9 (6-9)/2
Medical treatment only (n = 32)		**5.5 (3-6)/14**
Albendazole monotherapy	22 (68.8)	5 (3-6)/13
Mebendazole monotherapy	6 (18.8)	-
Albendazole + mebendazole	4 (12.5)	6 (6-6)/1

IQR, interquartile range; PAIR, puncture, aspiration, injection, and re-aspiration; PERC, percutaneous evacuation of residual cavity. IR, interventional radiology.

TABLE 5. Clinical Outcome, Follow-up and Recurrence/Relapse Rates.

		Clinical outcome			Follow-up duration in months		Recurrence/relapse		
	Numbers of cases	Complete recovery	Death	No data	Median/IQR	No data	+	-	No data
All cases	698	454 (98.9)	4 (0.9)	239	12 (6-36)	379	9 (2.8)	312 (97.2)	377
Urinary system	561	352 (98.9)	3 (0.8)	205	14 (6-36)	302	5 (2.0)	249 (98.0)	307
Kidney	478	285 (99.0)	2 (0.7)	190	12 (6-105)	267	5 (2.4)	202 (97.6)	271
Adrenal gland	64	54 (98.2)	1 (1.8)	9	24 (16-36)	22	-	40 (100.0)	24
Bladder	12	10 (100.0)	-	2	18 (12-66.7)	8	-	5 (100.0)	7
Ureter	3	1 (100.0)	-	2	-	3	-	-	3
Involvement of ≥2 organs	4	2 (100.0)	-	2	51 (6-96)	2	-	2 (100.0)	2
Female genital system	110	83 (98.8)	1 (1.2)	26	6 (3-22.5)	66	2 (4.0)	48 (96.0)	60
Ovary	51	36 (97.3)	1 (2.7)	14	6 (3-18)	32	1 (4.2)	23 (95.8)	27
Uterus	24	17 (100.0)	-	7	12 (2.5-28.5)	14	1 (10.9)	9 (90.0)	14
Adnexa	15	11 (100.0)	-	4	6 (2.5-18)	10	-	7 (100.0)	8
Fallopian tube	8	7 (100.0)	-	1	15 (6-51)	4	-	4 (100.0)	4
Broad ligament	5	5 (100.0)	-	5	1 (1-1)	4	-	1 (100.0)	4
Cervix	2	2 (100.0)	-	-	2 (2-2)	1	-	1 (100.0)	1
Involvement of ≥2 organs	5	5 (100.0)	-	-	7.5 (3-12)	1	-	3 (100.0)	2
Male genital system	27	19 (100.0)	-	8	12 (6-23.7)	11	2 (11.8)	15 (88.2)	10
Seminal vesicle	9	8 (100.0)	-	1	17.5 (7.5-24)	1	-	8 (100.0)	1
Scrotum	7	3 (100.0)	-	4	6 (6-6)	4	-	3 (100.0)	5
Spermatic cord	6	3 (100.0)	-	3	18 (12-24)	4	1 (50.0)	1 (50.0)	4
Prostate	3	3 (100.0)	-	-	9.5 (3-19)	1	-	2 (100.0)	1
Vas deference	1	1 (100.0)	-	-	3 (3-3)	-	-	1 (100.0)	-
Involvement of ≥2 organs	1	1 (100.0)	-	-	12 (12-12)	-	1 (100.0)	-	-

IQR, interquartile range.

The bladder and ureter involvement usually occurs secondarily after the renal cyst rupture. Primary cases are extremely rare (< 1%).[12,23] The presentations may include urinary retention, obstructive azoospermia, and hydatiduria.[24,25] In this review, 12 patients (2.1%) showed bladder involvement; three (0.5%) had ureter involvement, and two had both involvements.

Female genital CE is exceptionally rare.[26,27] The ovaries and the uterus are the most affected, with the ovaries accounting for 0.2-1% of the cases.[28] The symptoms are non-specific and include the following: abdominal pain, menstrual irregularities, infertility, and pressure effects.[29,30] It may mimic malignancy or endometriosis and causes obstructed labor.[31-34] Primary broad-ligament CE is extremely rare at only 0.037%.[35,36] In our data, abdominal pain was present in 81.7% of the female genital CE cases; palpable mass was found in 42.3%; and urinary symptoms were observed in 18.2%. Infertility, dysmenorrhea, and dyspareunia were also common. During the exam, 76.7% showed a palpable mass. Most of the cysts (90.8%) were ≥ 5 cm. USG and CT were the most frequently used imaging methods. The median follow-up was 6 months (IQR: 3-22.5). CE can primarily occur in the ovary. More frequently, it is in association with liver or lung echinococcosis or in context of multiorgan hydatidosis, considering that only rare cases with primary ovarian CE have so far been reported.[37,38] In cases involving unexplained infertility, persistent pelvic pain, or an adnexal mass, targeted ultrasound (with CT/MRI) and the consideration of the hydatid specific features are observed. Serology and screening for UGS-CE concurrent with hepatic or pulmonary cysts can support the diagnosis.

Meanwhile, male genital CE is rare but documented in the prostate, seminal vesicle, scrotum, testis, epididymis, and spermatic cord.[39-42] Its symptoms often mimic tumors or tuberculosis, consequently leading to misdiagnosis. The endoscopic and laparoscopic techniques are used similarly to those in other sites.[42,43] In this review, the seminal vesicle and the scrotum were the most commonly involved organs, and USG and CT were the main diagnostic tools.

This study has several limitations. Although a meta-analysis was initially considered, it was not performed because of the clinical and methodological heterogeneities among the included studies. The majority (> 95%) were case reports or small case series that were inherently descriptive and showed a variation in design, patient demographics, diagnostic methods, treatment approaches, and outcome reporting. Under these circumstances, statistical pooling would not have been methodologically sound and could have produced misleading results, which is consistent with those in established guidance.[44,45] Reporting the quality across the case reports had been inconsistent. Not all case reports were written with the same level of detail. Many lacked standardized information as regards the clinical presentation, diagnostics, treatment, and follow-up. Consequently, certain variables were missing and had to be excluded from the corresponding analyses considering the lack of imputation method applied. This limited the completeness of some

findings. There existed a potential risk of reporting or publication bias. UGS-CE is a rare localization; hence, published literature often focuses on unusual or clinically striking cases, while more typical ones may not be reported or may have missing details. Therefore, the distribution of symptoms, diagnostics, and outcomes in our review reflects published evidence rather than the full clinical spectrum. This limitation is inherent to systematic reviews of rare conditions because analyses are necessarily restricted to the available literature. Finally, a large proportion of the included cases originated from regions like Türkiye, Iran, and India, where CE is more prevalent. This geographical clustering may have influenced the patterns of the symptoms, diagnostics, and management observed in our review, potentially limiting the findings' generalizability.

In summary, CE in the UGS predominantly affects the urinary tract, having the kidney as the most commonly involved organ. Flank pain is the most frequent symptom. Palpable mass is the most common physical finding. Among the female genital organs, the ovary is the most often affected. Meanwhile, in males, the seminal vesicle is the primary site. Ultrasonography and CT are the most frequently used imaging tools, with most of the cysts exceeding 5 cm at the diagnosis. Hepatic involvement is the most common concomitant finding. A combined surgical and medical therapy is the preferred treatment, with the postoperative albendazole monotherapy more commonly administered than the preoperative treatment likely because of the intraoperative diagnosis following the surgery performed for other indications. Due to the rarity and the non-specific presentations of UGS-CE, it should be included in the differential diagnosis of patients presenting with abdominopelvic or flank pain, hydatiduria, urinary symptoms, and infertility, particularly in endemic regions. CE can closely mimic urogenital tumors on clinical exam and imaging; hence, a cautious differential diagnosis is essential. Imaging, especially USG supported by CT or MRI, is a key to a diagnosis. The favorable outcomes rely on early detection and timely intervention to reduce the recurrence/relaps risk. Clinicians practicing in endemic regions must maintain a high index of suspicion to enable a timely diagnosis, reduce misdiagnoses, and prevent unnecessary interventions. Future multicenter studies with standardized reporting are necessary in strengthening the evidence base and better guiding clinical practice.

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